THE IOWA ORTHOPAEDIC JOURNAL

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Matthew R. Lavery, M.D.

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Jose A. Morcuende, M.D.

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INSTRUCTIONS FOR AUTHORS, 2008

Any original article relevant to orthopaedic surgery, orthopaedic science or the teaching of either will be considered for publication in The Iowa Orthopaedic Journal. Articles will be enthusiastically received from alumni, visitors to the department, members of the Iowa Orthopaedic Society, residents, and friends of The University of Iowa Department of Orthopaedics and Rehabilitation. The journal is published every June. The deadline to receive articles for the 2008 edition is Friday, January 11, 2008.

Published articles and illustrations become the property of The Iowa Orthopaedic Journal. The journal is peer reviewed and referenced in Index Medicus. Articles previously published will not be accepted unless their content has been significantly changed.

When submitting an article, it is essential to include the following:

1. **The original manuscript with illustrations and abstract.**
   The corresponding author must be clearly identified with mailing address, telephone/fax numbers and e-mail address. Manuscripts for accepted articles will not be returned.

2. **References**, presented in the text by superscript numbers. The bibliography should list references in the order of their appearance in the text, and be double-spaced.

3. **Legends** for all illustrations, listed in order of appearance and double-spaced.

4. **Illustrations/ Images**:
   a. One set of 5 x 7-inch, black-and-white, glossy prints of all photographs.
   b. Original drawings or charts.
   c. Color illustrations may not be used unless it is the opinion of the journal that they convey information not available in black and white.
   d. Each image should be sent to diana-johannes@uiowa.edu as an individual .tif or .jpg file. If images are embedded in

the manuscript, individual image files should still be sent. All images must have resolution of 300 pixels per inch (ppi). Web page images are to be avoided. Set digital cameras to their highest quality (ppi) setting for photographs.

5. **Electronic copies of all items above**, sent to diana-johannes@uiowa.edu. Special illustrations and photographs may be exempted from this electronic requirement and should be mailed to the address below.

**Preparation of manuscripts**: Manuscripts must be typewritten and double spaced using wide margins. Write out numbers under 10 except percentages, degrees or numbers expressed as decimals. Direct quotations should include the exact page number on which they appeared in the book or article. All measurements should be given in SI metric units. In reporting results of surgery, only in rare instances can cases with less than two years of follow-up be accepted.

**Preparation of photographs/illustrations**: On the back of each photo and illustration, write the figure number, author’s name and indicate the top. Send prints unmounted - paste or glue will damage them. Drawings, charts and lettering on prints should be done in black with white backgrounds. Put dates or initials in the legends, not on prints. Make lettering large enough to be read when drawings are reduced in size. When submitting an illustration that has appeared elsewhere, give full information about previous publication and credit to be given, and state whether or not permission to reproduce it has been obtained.

Additional copies of these instructions may be obtained at www.uihealthcare.com/depts/med/orthopaedicsurgery/research/ioj.html or by writing to Diana Johannes, University of Iowa Hospitals and Clinics, Department of Orthopaedics and Rehabilitation, 200 Hawkins Drive – 01006 J.P.P., Iowa City, Iowa, 52242-1088 or by emailing diana-johannes@uiowa.edu.

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ERRATUM
IOWA ORTHOPAEDIC JOURNAL
VOLUME 26, JUNE 2006

In the 2006 Iowa Orthopaedic Journal, Volume 26, both in the table of contents and in the article titled “Proximal Tibiofibular Synostosis as a Source of Ankle Pain: A Case Report,” page 127 et al, the first author’s name should read Vinayagam Lenin Babu in all instances. This correction is typographic only and content is not affected. We extend our thanks to Dr. Lenin Babu for kindly bringing this to our attention. All rights reserved.
EDITORS’ NOTE

We are pleased and honored to present the 2007 edition of The Iowa Orthopaedic Journal (IOJ). In keeping with the tradition of the IOJ, this issue contains publications of scientific articles, reviews, case reports, and essays of both historical and philosophic interest. We hope those who read this edition will come away with increased knowledge and enthusiasm for the field of Orthopaedics.

This 2007 edition marks the beginning of an evolution for the IOJ. For the first time, the IOJ is being made available in an electronic format. It will also be the first time that the articles contained herewithin will be available for global perusal via full-text links through PubMed. It is our hope that the time and dedication our authors have put in their manuscripts may thus be appreciated by a worldwide readership.

In keeping with the theme of globalization, this edition of the IOJ expands upon its submitter base as well. As always, we are pleased to have a significant contribution from the faculty, alumni, and residents of our own Orthopaedic Department. However, we also have many contributions this year from other quality institutions and clinicians from both across the country and around the world. This broad range of submissions greatly enriches the IOJ and to our many submitters worldwide, we wish to express our endless gratitude.

We would also like to recognize the departing senior residents. Drs. Amiritharajah, Altenburg, DeWall, Jones and Ward are exemplary clinicians, dedicated academics, role models for excellence, and most importantly, great friends. They will be missed dearly but their inspiration to us will remain.

This edition is dedicated to Don Shurr. He is not only an exceptionally gifted orthotist, but also a great teacher and friend. We are forever indebted to his vast knowledge of prosthetic and orthotic care in Orthopaedics and his infinite patience for putting up with our own lack of finess in said field. It has been a privilege to work with and learn from him, and our department is undoubtedly strengthened through his contributions.

The IOJ would not be possible without the dedicated support of Diana Johannes, who has worked tirelessly on the monumental task of coordinating manuscripts, corporate advertisements, and department photos, as well as craftily keeping us focused on our task (which is easily her most impressive accomplishment). Our predecessors can undoubtedly empathize with our gratitude for her efforts.

A journal of this size and quality could not be possible without the tremendous financial contributions of our corporate sponsors. We thank them for their generosity and continued support, both in the care of patients and educational forums.

Finally, we would like to thank Dr. Jose Morcuende and Dr. Joseph Buckwalter for their continued support and leadership as faculty advisors for the IOJ as well as all of the previous editors, who have set the high standard to which we aspire.

We hope you find this edition of the IOJ as enjoyable to read as it was to edit. Congratulations and thanks to all who have had a part in making The Iowa Orthopaedic Journal a lasting icon.

Michael S. Chang, M.D.
Matthew R. Lavery, M.D.
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The publication of the 2007 edition of the *Iowa Orthopaedic Journal* provides us with another opportunity to thank someone who has profoundly impacted Orthopaedic Surgery resident education. This man has been a part of the University of Iowa Community since 1964, and has served in his current capacity for more than 20 years. It is with great pleasure that we dedicate the 2007 *Iowa Orthopaedic Journal* to Donald Shurr, C.P.O., PT.

Don grew up in Pontiac, Illinois, where he was a three-sport athlete. He earned all-state honors in football and basketball, and holds the distinction as a baseball player of pitching a no-hitter in a high school All-Star game. He went on to attend the University of Iowa, playing football for the Hawkeyes in 1964. Don was sidelined with a severe knee injury early in his Big Ten career. He underwent intensive Physical Therapy in an attempt to return to the game, but a football career was not to be. He maintained his scholarship working as a student assistant coach. Don continues to enjoy football, and has served as a high school referee for well over 20 years.

Although his injury kept him from playing collegiate football, it served as an introduction to Don’s eventual vocation. After completing his undergraduate work, Don attended physical therapy school for 2 years then went on to earn his Masters Degree in PT a year later. In 1971 he began working as a therapist at UIHC, eventually becoming the director of the PT department. Through Don’s patient interactions, he discovered that his true calling lay elsewhere. He went back to school in 1983 to earn his CPO degree at Northwestern University. He returned to Iowa City, and has worked in prosthetics and orthotics at the University ever since. Don currently serves as a regional manager, overseeing four other offices as well.

Don was married in 1967 to his wife Marilyn. They have two daughters, Molly and Carrie. Molly works as an Occupational Therapist at the hospital, and Carrie previously worked with AmPro before taking a job as a school administrator. Don and his wife have been blessed with three grandchildren, and are expecting their fourth soon.

Through his years of involvement with the Orthopaedics Department, Don has been involved in numerous research studies. He has been coauthor on papers with Drs. Saltzman, Cooper, and Buckwalter to name but a few. Much of Don’s research has focused on the unique prosthetic needs and physiologic demands of the amputee. He is the author of a Prosthetics and Orthotics Text, and has contributed to numerous other books on the subject.

Don has served as a lecturer for our department on many occasions, teaching residents basic concepts about Orthotics and Prosthetics as part of our didactic curriculum. Many residents have also been treated to an evening at the Brown Bottle where Don takes the time to review P & O principles in a less formal setting. He clearly loves his role as a teacher.

Don is the “can-do” man. Despite sometimes unreasonable requests from Resident Physicians on Saturday afternoons, Don will almost always find a way to provide patients with great care and our department with great service. Many of the challenges we take to Don do not have standard solutions; using his creativity and experience, he nearly always will find one, though.

Although Don is clearly dedicated to his work, his passion outside the confines of UIHC has long been hunting. He frequently travels to Montana to hunt deer, elk, bighorn sheep and other large game. He has claimed trophy bucks, one of which is the mule deer pictured here. Over the years he has spent a fair amount of time hunting with the residents, and has come to befriend many.

Don’s love of the outdoors and love of history find common ground in the Lewis and Clark Expedition. He has been very active in teaching this part of American history to both children and adults all over the state of Iowa.

It is our honor and great pleasure to dedicate the 2007 *Iowa Orthopaedic Journal* to Don Shurr. Don has provided our department with much more than Orthotic and Prosthetic service; he has served as an outstanding example of dedication, hard work, and service to others. We consider ourselves privileged to work with him on a daily basis.
2007-2008
DEPARTMENT OF ORTHOPAEDICS AND REHABILITATION
SCHEDULE OF LECTURESHIPS AND CONFERENCES

(Larson Conference Room, 01090 JPP)

2007 Senior Residents Day
June 8 & 9, 2007
Dr. Marc Swiontkowski
Dr. Regis O'Keefe

Iowa Orthopaedic Alumni Meeting
October 4-6, 2007
Dr. Lew Schon
Greater Chesapeake Memorial Hospital, Baltimore

Dr. Elizabeth “Liza” Arendt
Ruth Jackson Memorial Lecturer
Department of Orthopaedic Surgery
University of Minnesota

23rd Annual Hawkeye Sports Medicine Symposium
November 30 & December 1, 2007
Marriott Hotel and Conference Center
300 East 9th Street, Coralville
*Contact Kris Kriener (319) 353-7954

Carroll B. Larson Shrine Memorial Lecture
Spring, 2008

Eighth Biennial Johnston Lectureship
In Hip Reconstruction
October, 2008*

Reginald R. Cooper
Orthopaedic Leadership Lectures
April, 2009

*Please check with us later for exact dates, times and speakers.
Department of Orthopaedics

The University of Iowa
Roy J. and Lucille A. Carver College of Medicine
DEPARTMENT OF ORTHOPAEDICS AND REHABILITATION STAFF 2006-2007

Dr. Brian Adams  Dr. John Albright  Dr. Ned Amendola  Dr. Thomas Brown  Dr. Joseph Buckwalter  Dr. John Callaghan

Dr. Joseph Chen  Dr. Charles Clark  Dr. Reginald Cooper  Dr. Fred Dietz  Mr. Paul Etre  Dr. John Femino

Dr. Ernest Found  Dr. Nicole Grosland  Dr. Richard Johnston  Dr. Erika Lawler  Dr. J. Lawrence Marsh  Dr. Todd McKinley

Dr. Sergio Mendoza  Dr. Jose Morcuende  Dr. James Nepola  Dr. Michael O’Rourke  Dr. Ignacio Ponseti  Dr. Neil Segal

Dr. Joseph Smucker  Dr. Stuart Weinstein  Dr. Brian Wolf  Dr. Robert Yang
Matthew DeWall, M.D.

Matt DeWall grew up on a farm in northwestern Iowa. He completed his undergraduate as well as medical school training at the University of Iowa. It was during his last year of medical school that he met his wife Jennifer, who moved to Iowa City to join him for his residency. In July, Matt and Jen will travel to Sydney, Australia, for his one-year fellowship in knee surgery. After the fellowship, they plan to return to Iowa where Matt will join a private-practice orthopaedic group.

He would like to thank his classmates for being great colleagues and friends, the faculty for all of their contributions to his training, and most importantly, his wife Jen for her support and understanding.
Kevin Jones, M.D.

Kevin was born in southern California and raised in St. Louis. He came to Iowa after earning a degree in English literature at Harvard University and his medical degree from Johns Hopkins. He and his wife, Arden, have had simultaneous residencies while in Iowa City, hers in child-rearing and his in orthopaedics. They both count many blessings from the last five years, most importantly, the three children who have joined their family, Caelen, Ewin, and Lillien.

Next year they will move to Toronto for Kevin’s fellowship in musculoskeletal oncology. The year after that, Kevin will begin a faculty position at the University of Utah, including basic science research and patient care in the Huntsman Cancer Center.

Kevin has relished the unique environment of the department, which encouraged—never stifled—curiosity and healthy skepticism. He also enjoyed the open and inviting scientific environment of the University overall, where requests for multidisciplinary collaborations were always welcomed. Most importantly, he is grateful for the mentorships he received from so many in the Department. He was always surprised to see that his education was of sincere interest to the faculty, even in the midst of their many other departmental, institutional, national, and international involvements and responsibilities.

Christina Ward, M.D.

Christina Ward was born and raised in Kansas. She received her undergraduate degree from Grinnell College in Grinnell, Iowa, where she was introduced to the wonderful Iowa winters. On a positive note, Grinnell was also where she was introduced to her husband, Nathan Lueck. She received her medical training at Washington University in St. Louis. Christina will spend next year completing hand fellowship training at the University of Iowa.

Christina and Nate have appreciated the kindness and support of all of the orthopaedics department faculty and staff, and have especially enjoyed the friendship and support of the residents.
2007 GRADUATING FELLOWS

Bryce Bederka, M.D.

Bryce Bederka was born in Bangkok, Thailand, but unfortunately has no memory of that time in his life. The family moved to Chicago, Illinois, when he was 2 1/2, and he has spent the rest of his life in the Midwest. He attended the University of Iowa and, although he initially enrolled in a premed track, received his undergraduate degree in business administration. Bryce worked for several years in Chicago before realizing that he really did indeed want to be a doctor, and began medical school at the University of Illinois at Chicago with the intention of becoming an orthopaedic surgeon specializing in sports medicine. He completed medical school and then residency at that same institution.

While looking at fellowships, Bryce was very impressed with what he saw at the University of Iowa, and was excited about having the chance of working intimately with the Hawkeye athletics program that had given him so many years of excitement in the past. He has greatly enjoyed his year here at Iowa, working with the Orthopaedic staff and residents, and all of the athletics staff.

This fall, Bryce will be moving with the family to Portland, Oregon, to start up practice with a private group. He hopes to establish a relationship with one of the small local colleges or universities in town, but has not ruled out migrating south to Eugene and becoming a Duck. He also plans on taking advantage of all the Pacific Northwest has to offer in terms of outdoor lifestyle. He will miss the Midwest and Chicago, but, as he has been known to say, "Chicago is a great city, it's just in a crappy location."

Danielle Conaway, M.D.

Danielle Conaway is the current Hand Fellow in the Department of Orthopaedics and Rehabilitation at The University of Iowa Hospitals and Clinics. She received her undergraduate degree at Eastern Michigan University where she competed in Cross Country and Track. Dr. Conaway then obtained her medical degree from Wayne State University in Detroit, Michigan. From 2001 to 2006, Danielle completed her Orthopaedic Surgery Residency training in Grand Rapids, Michigan. She recently accepted a hand surgery position in Traverse City, Michigan. She and husband Scott are also expecting their first child in September.

Mauricio Campos Daziano, M.D.

Mauricio Campos Daziano is an Associate Instructor in Orthopaedic Surgery, Pontificia Universidad Catolica de Chile. He attended medical school and later completed his orthopaedic residency at the same institution. He has been an attending orthopaedic surgeon since 2003 at PUC, dedicated completely to the field of Spine Surgery. He sought further training abroad, completing a one-year Fellowship in Pediatric Spine Surgery at The University of Iowa under the guidance of Dr. Stuart L. Weinstein and next year will complete a year of Fellowship in Spine Surgery at the University of California, San Francisco.
2007 DORIS DUKE FELLOW

Anjan Kaishik

Anjan Kaishik is a native of Bangalore, India, who has lived in Wisconsin and Virginia for most of his life. He graduated from the University of Wisconsin - Madison in 2003 with Bachelor of Science degrees in French and Biology. He now attends the University of Virginia School of Medicine. In 2006-07, Anjan took a year to pursue research at the University of Iowa Department of Orthopaedic Surgery under the Doris Duke Clinical Research Fellowship. This fellowship has allowed him to work closely with Dr. Jose Morcuende on a basic science project examining the cell biology of articular cartilage. He is studying the functions of the chondrocyte primary cilium. He received the Iowa Orthopaedic Society Award for Student Research in 2007 and presented his research at the spring 2007 IOS meeting. Anjan's avocations include playing racquet sports, sketching, hiking, and bicycling on Iowa country roads.

Upon completing his year-long fellowship, he will return to Virginia to complete his final two years of medical school. He intends to pursue an Orthopaedic Surgery residency in the Midwest and possibly specialize in pediatric or hand surgery.
NEW ORTHOPAEDIC FACULTY

Ericka Lawler, M.D.
Ericka Lawler joined The University of Iowa Department of Orthopaedics and Rehabilitation in August of 2006 as part of the Hand and Upper Extremity Division.

Originally from Michigan, Dr. Lawler received her undergraduate degree from Yale University in 1996. She obtained her medical degree from George Washington University School of Medicine in 2000. In 2005, she completed her residency in orthopaedics at New York University - Hospital for Joint Diseases. She subsequently served as a fellow in hand surgery from 2005-2006 here at the University of Iowa where she accepted an invitation to stay on as a faculty member.

Dr. Lawler’s clinical interests include congenital hand deformities and soft tissue reconstruction. She lives here in Iowa with her husband, Judd, and daughter, Claire. Outside of work she enjoys hiking, cooking and soccer.

Robert Yang, M.D.
Robert Yang joined the University of Iowa Department of Orthopaedics and Rehabilitation in 2007 as part of the Spine Center. He originally hails from the Washington DC area and received his undergraduate degrees in biochemistry and mathematics, and medical degree from Washington University in St. Louis. He completed residency training in Physical Medicine and Rehabilitation at Mayo Clinic in 2000 and was on the faculty at Mayo Clinic Rochester from 2000 – 2004, and the University of North Carolina from 2004 – 2006.

His clinical interests are musculoskeletal disorders and medical acupuncture. He completed additional training in acupuncture in 2003 and is board certified in medical acupuncture by the American Board of Medical Acupuncture. He also has an active interest in health policy and is currently pursuing an advanced degree through the University of North Carolina School of Public Health, Division of Health Policy and Administration.
The Michael Bonfiglio Award for Student Research in Orthopaedic Surgery

The Iowa Orthopaedic Society Medical Student Research Award for Musculoskeletal Research

As has been the tradition, the University of Iowa Department of Orthopaedics and Rehabilitation, along with the Iowa Orthopaedic Society, sponsor two research awards involving medical students.

The first, the Michael Bonfiglio Award, originated in 1988 and was named in honor of Mike, who had an avid interest in students, teaching and research. The award is given annually at medical convocation. It consists of a plaque and a stipend. It is awarded to a senior medical student in the Carver College of Medicine who has done outstanding orthopaedic research during his or her tenure as a medical student. The student has an advisor in the Orthopaedic Department; however, the student must have played a major role in the design, implementation and analysis of the project. He or she must be able to defend the manuscript in a public forum. The research project may have been either a clinical or basic science project, and each study is judged on the basis of originality and scientific merit. The winner presents their work at the spring meeting of the Iowa Orthopaedic Society, as well as at a conference in the Department of Orthopaedics and Rehabilitation.

The second award is the Medical Student Research Award for Musculoskeletal Research. This award is for students in the Carver College of Medicine who complete a research project involving orthopaedic surgery during one of their first three years of medical school. The award consists of a $2000 stipend, $500 of which is designated as a direct award to the student, and $1500 of which is designated to help defray continuing costs of the project and its publication. The student must provide an abstract and a progress report on the ongoing research. The aim of this award is to stimulate research in the field of orthopaedic surgery and musculoskeletal problems. This award is also presented at a medical convocation. In addition, the student presents their work at the spring meeting of the Iowa Orthopaedic Society and at a conference in the Department of Orthopaedics and Rehabilitation.

This year, the awards committee, consisting of a member of the Iowa Orthopaedic Society (Dr. Joseph Martin, President) as well as members of the Orthopaedics and Rehabilitation Department (Dr. Charles Clark, Dr. Joseph A. Buckwalter, Dr. Brian Wolf, Dr. Jose A. Morcuende, and Dr. John Femino), recommended that awards be given to the following students:

Dr. Andrea E. Buckwalter and Dr. Matthew Lovell were the co-recipients of the 2007 Michael Bonfiglio Student Research Award. Andrea’s award was based on her project, “Results of Charnley Total Hip Arthroplasty With the Use of Improved Femoral Cementing Techniques at a Minimum of Twenty-Five Years.” Her research advisor was Dr. John Callaghan, and co-authors of the study were Dr. Lori Dolan and Dr. Ignacio Ponseti.

The second Dr. Michael Bonfiglio Award winner was Dr. Matthew Lovell, whose award was based on his project “Natural History and the Effects of Foot Hyperabduction in Clubfoot Relapses.” His research advisor was Jose Morcuende, M.D., Ph.D., and his co-investigators were Dr. Lori Dolan and Dr. Ignacio Ponseti.

This year’s award winner of the Iowa Orthopaedic Society Medical Student Research Award is Anjan P. Kaushik, M3. Anjan’s study is titled, “Mechanosensory Functions of the Chondrocyte Primary Cilium in Bardet-Biedl Syndrome and Conditional Polaris Knockout Mice.” His advisor was Jose Morcuende, M.D., Ph.D., and his co-investigators are Dr. Martin Zhang and Dr. Val Sheffield.

The Michael Bonfiglio Award and the Iowa Orthopaedic Society Medical Student Research Award for Musculoskeletal Research are very prestigious, recognizing student research on the musculoskeletal system. These awards have indeed attained their goal of stimulating such research and have produced many fine projects over the years.

Charles R. Clark, M.D.
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ANTIOXIDANTS BLOCK CYCLIC LOADING INDUCED CHONDROCYTE DEATH

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ABSTRACT

Articular cartilage in congruous joints benefits from the moderate stresses and strains associated with normal cyclic loading. However, loading of joints with surface incongruities can lead to local stress and strain elevation at “step-off” sites where cartilage is not fully buttressed by surrounding matrix. Excessive stresses and strains predicted to occur at such sites may induce apoptosis, a process thought to promote cartilage degeneration and osteoarthritis (OA) through chondrocyte attrition. We hypothesized that the induction of apoptosis is mediated by oxidants, and that antioxidants can reduce elevated stress-induced chondrocyte attrition. To test this we exposed cylindrical cartilage explants from human articular cartilage to radially unconfined cyclic axial compression (3600 cycles, 1 Hz, 50% duty cycle) using two different physiologic loads (2 MPa and 5 MPa). We found that 30% of chondrocytes in the superficial zone died within 24 hours of exposure to loading with 5 MPa axial compression, whereas mortality was limited to less than 15% with 2 MPa axial compression. Similarly, lactate accumulation in the medium was suppressed by compression with 5 MPa, but not 2 MPa. Approximately 80% of cell death induced by 5 MPa compression was blocked by pre-incubation of the explants in a variety of anti-oxidants including vitamin E, n-acetyl cysteine (NAC), and a superoxide dismutase mimetic (SOD). SOD and NAC also prevented the suppression of lactate secretion after 5 MPa compression. These observations support the hypothesis that the harmful effects of abnormal cyclic loading are mediated by oxidants and suggest that treatments to prevent OA may include methods of minimizing oxidative damage to chondrocytes.

INTRODUCTION

High-energy joint injuries that disrupt articular surfaces often lead within a few years to post-traumatic OA.13,15,32 Although the overall risk for post-traumatic OA varies among different joints and with patient age at the time of injury, mechanical factors play a significant role in determining outcome.4,5,32 These factors include the energy delivered to the articular surface at the time of injury and, in joints with residual surface incongruity or instability, the potential for chronically abnormal mechanical stress. The development of treatments to forestall post-traumatic OA may be aided by a better understanding of the biologic responses to mechanical conditions in injured joints.20,38,40

The effects of mechanical injury on cartilage stability have been investigated in a number of in vitro studies using cartilage from various animals and from humans. Physical effects of impact loading were demonstrated by Jeffrey et al.22 This study of bovine cartilage showed a linear increase in chondrocyte death three days after impact, indicating a partial loss of ECM integrity. A follow-up study showed that biosynthetic activity, as assessed by metabolic radiolabeling, was significantly impaired in the days post-impact.23 Similar results were found in canine cartilage after impact: Hexosamine content in impacted cartilage declined after impact loading, coincident with structural disruption and death of chondrocytes.9 Proteoglycan synthesis was also impaired and water content declined in bovine cartilage exposed to severe impacts of 25-75 MPa.2 These authors also reported the detachment of cartilage surrounding an impact site as a significant source of destabilizing collateral damage. Other authors studying the metabolic and physical effects of impact loading reported that proteoglycan synthesis declined at 24 hours post-injury in concert with loss of chondrocyte viability, collagen network disruption, and increased tissue hydration.39 They suggested that the threshold for these effects was near 20 MPa and that loads equal to or greater than that caused irreparable structural damage. Though not as severe as the effects of impact loading, cyclic loading may also contribute to matrix damage and instability.11 Lin et al., reported that cyclic loading with 5 MPa (0.5 Hz) for 1 or more hours caused increased stromelysin-1 (MMP-3), proteoglycan degradation and collagen damage in cyclically load-injured articular cartilage.36 These studies demonstrate unequivocally that mechanical insult is associated with chondrocyte death, either by apoptosis or necrosis, loss of biosynthetic activity, and with structural disruption of the cartilage ECM.

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Several studies show an association between mechanical stress and increased production of reactive oxygen species (ROS), as well as decreased antioxidant capacity. Although low levels of oxidants are required for normal cartilage metabolism, exposure to elevated ROS levels is associated with chondrocyte death and matrix degeneration. There is evidence to suggest that apoptosis associated with intra-articular fractures is due, at least in part, to elevated levels of ROS induced by impact loading. Apoptosis and the resulting reduction in cell density is thought to play a significant role in the development of post-traumatic OA.

Exogenous antioxidants provide some protection to chondrocytes from the harmful effects of elevated oxidants. There is evidence that a superoxide dismutase (SOD) mimic combined with methotrexate protects rats with collagen-induced arthritis from the development of OA. Previous studies in our laboratory established that mechanical compression in an unconfined configuration leads to extensive chondrocyte death and apoptosis in cartilage explants. Cell death was blocked by the free radical scavenger n-acetyl cysteine (NAC), suggesting that oxidative damage mediated the harmful effects of compression. Another published study indicated that apoptotic chondrocyte death induced by a single high impact load could be inhibited by treatment with a SOD mimic that detoxifies oxygen free radicals, but not by the anti-lipid peroxidation activity of α-tocopherol (vitamin E). These findings suggested that lipid oxidation plays a minor role in impact-induced death. Nitric oxide (NO) also appears to play a role in oxidative apoptosis: The nitric oxide synthase inhibitor N-Nitro-L-Arginine methyl ester (L-NAME) has been shown to inhibit chondrocyte apoptosis when oxygen free radicals are present. One study suggests that a peroxynitrite scavenger decreases GAG loss in articular cartilage implicating an oxidative mechanism for GAG breakdown.

Estimates of contact stresses at cartilage surfaces in ankle and knee joints during routine physical activity range from a few to several MPa. Within this range of contact stresses it is thought that articular cartilage strain in normal, congruent joints rarely exceeds 15%. However, in incongruent joints, the same moderate contact stresses can lead to local strains of 30% or greater at “step-off” sites due to the partial loss of transverse support normally provided by surrounding matrix. Repetitive exposure to strains near this magnitude has been shown to induce chondrocyte apoptosis in cyclically-compressed cartilage explants. We hypothesized that apoptosis induced by cyclic axial compression is mediated by oxidants, and is therefore subject to inhibition by antioxidants. To test this we determined the effects of antioxidants on chondrocyte viability in human cartilage explants exposed to unconfined cyclic axial compression (1 Hz, 3600 cycles) in a mechanically-active bioreactor. Compression amplitude was varied (2 MPa or 5 MPa) to produce near physiologic strains (10-15%), and super-physiologic strains (20-30%), representing conditions that might occur in and around step-off sites in vivo.

METHODS

Human cartilage explants (4 mm diameter) were harvested from non-osteoarthritic ankle joints from 10 donors. The explants were incubated in culture medium (40% Dulbecco’s modified Eagle medium, 40% Ham’s F12, 10% alpha-MEM, 10% fetal bovine serum) in a 5% O₂, 5% CO₂, 90% N₂ atmosphere at 37°C. Some explants were incubated in antioxidants for 24 hours prior to mechanical stress exposure. These antioxidants included n-acetyl-cysteine (NAC) at a concentration of 2.5 mM, superoxide dismutase (SOD) (MnTBAP) at a concentration of 50 μM, catalase (Aspergillus niger) at a concentration of 15 mg/mL, and vitamin E at a concentration of 100 μM. Additional explants were incubated for 24 hours with the nitric oxide (NO) synthase inhibitor N-Nitro-L-Arginine Methyl Ester (L-NAME), which blocks NO-induced apoptosis. L-NAME was used at a concentration of 1.0 mM. Untreated controls were also included. All explants were placed in the bioreactor for mechanical stress treatment. The bioreactor is a mechanically active culture device capable of imposing variable shear stress states at quasi-physiologic levels. Here, the bioreactor was used to apply unconfined cyclic axial compression to cartilage explants (30-40 MPa/sec loading, 3600 cycles at 1 Hz, 50% duty cycle) to simulate the lack of support at joint reduction incongruities. The amplitude of compression was either 2 MPa or 5 MPa. Non-compressed controls were included in each experiment.

Data obtained from DVRTs and caliper measurements of initial and final explant height were used to calculate Green-Lagrange strains over 3,600 cycles of loading. Strain versus cycle number plots revealed that the rate of change in overall strain, and the greatest peak-to-peak strains were greatest over the first few hundred cycles of loading. The rate of change declined significantly after 1000 cycles, when near maximum steady strain was reached. Linear regression analysis over early (75 to 250 cycles) and late (1,000 to 3,500 cycles) intervals was used to derive slopes and y-intercepts to characterize the rate of change in overall strain, the maximum strain within each cycle, and maximum overall strain. These parameters were calculated for individual explants and the data were pooled to find means for 5 MPa compression (n = 35) and 2 MPa compression (n = 41). Student's
t-test was used to determine the significance of the differences between these groups.

Following mechanical compression, explants (n = 6 per group) were removed from the bioreactor and incubated overnight in calcein AM to stain viable chondrocytes and ethidium homodimer to stain nonviable chondrocytes. Explants were cryoembedded and an aliquot of the medium was removed for lactate assay, which was performed using a colorimetric assay kit according to the manufacturer’s directions (Biomedical Research Services).

Explants (6 per group) were cryosectioned and imaged with 488 nm light using an Olympus BX60 epifluorescence microscope equipped with a stepper motor-driven stage. Individual frames were tiled to form high-resolution composite images. Viable cells (calcein-stained) and nonviable cells (ethidium homodimer stained) were counted using a custom designed automated MATLAB based image analysis program to determine percent viability. The program automatically identifies and counts fluorescent-labeled cells in high-resolution composite images of full thickness cartilage sections (4 mm wide x 1-3 mm thick). The program also automatically segments images into superficial (top 15%), middle (16%-45%), and deep (46-100%) zones. Percent viability was normalized to non-stressed controls in each experimental group. Duplicate sections were analyzed from each of the 6 explants representing an experimental group (12 sections total).

Replicate cryosections from explants were stained for apoptosis using a commercial in situ TUNEL assay kit with tetramethyl rhodamine- or fluorescein isothiocy-anate-labeled dUTP suitable for epifluorescence imaging (Roche). These stains were imaged and analyzed as described above for viability stains.

Each experiment was performed with at least 6 explants. Kruskal-Wallis One Way Analysis of Variance on Ranks was used to evaluate the statistical significance of differences between treatment groups.

RESULTS

Representative plots illustrate strain behavior of explants over 3600 cycles of 2 MPa or 5 MPa axial compression (Figure 1). Maximum overall strain averaged $19 \pm 4.5\%$ with 2 MPa compression and $27 \pm 3.4\%$ with 5 MPa, a significant difference ($p < 0.001$). Dynamic strain increased significantly ($p <0.001$) from $5.9 \pm 2.7\%$ with 2 MPa compression to $9.0 \pm 2.6\%$ with 5 MPa axial compression.

Fluorescence stains for chondrocyte viability revealed that most chondrocytes (>85%) in all cartilage zones were alive in control explants and in explants after 2 MPa compression, but there was significant loss of viability in explants after 5 MPa compression (Figures 2A, 2B, 2C). The greatest losses occurred in the superficial zone, where viability was reduced to 40%. However, there was also significant death in the middle zone, in which viability was reduced to 55% (Figure 2D).
Post-compression histological assays for DNA fragmentation (TUNEL reaction) revealed that much of the cell death observed in viability assays was attributable to apoptosis (Figure 3). The frequency of TUNEL reaction positive cells in uncompressed controls was less than 5% in all zones but increased to >20% in the superficial and middle zones after 5 MPa axial compression. NAC pre-treatment reduced the number of positive cells to less than 10% in both zones. The primarily superficial/middle zone distribution of TUNEL reaction-positive chondrocytes was similar to the pattern of dead cells detected by calcein AM/ethidium homodimer staining.

Figure 2. Effects of Axial Compression on Chondrocyte Viability. Representative photomicrographs show living chondrocytes (green) or dead chondrocytes (red) in explants that were either unloaded (A), or exposed to 2 MPa axial compression (B), or 5 MPa axial compression (C). The surface of the explant faces upward in each panel. The bar in panel C represents a length of 500 μm. (D) The columns show average viabilities (mean and standard error based on 6 explants) in the superficial (S), middle (M) and deep (D) zones.
Chondrocyte mortality induced by high she stress was dramatically affected by pre-incubation with antioxidants prior to stress treatment (Figure 4). Post-compression viability in NAC-, vitamin E-, and SOD-treated explants increased in the superficial zone from ~40% to ~80%, a statistically significant change (p<0.005). The same reagents improved viability significantly (from 53% to >80%) in the middle zone, whereas only NAC improved viability in the deep zone (from 83% to >87%) (Figure 4B). L-NAME, a nitric oxide synthase inhibitor that has been shown to block chondrocyte apoptosis, had effects similar to those of antioxidants. Catalase appeared to have little if any beneficial effects in the superficial zone, as post-stress viabilities remained low (60%) in the presence of the enzyme.

Lactate accumulation in the culture medium was measured to determine compression and antioxidant effects on chondrocyte metabolism (Figure 5). Compression with 5 MPa caused a significant reduction in lactate (p < 0.05) relative to unloaded controls, but compression with 2 MPa had no significant effect. Lactate production in explants compressed with 5 MPa was maintained at control levels when NAC or SOD was used for pre-treatment, but not when catalase was used.

DISCUSSION
Chondrocyte viability after 3600 cycles of compression with 5 MPa was significantly higher in explants treated with a variety of antioxidants than in untreated controls exposed to the same compression regime. Post-stress viability in the superficial zones of anti-oxidant-treated explants was near 80%, twice as high as in controls. These findings implicate oxidative stress in the harmful effects of cyclic mechanical stress in articular cartilage and show for the first time that antioxidants protect chondrocytes from those effects.

The TUNEL reaction indicated that compression with 5 MPa induced apoptosis in the superficial and middle zones of cartilage explants. In cyclically compressed explants, nearly 20% of chondrocytes in these zones were positive for the TUNEL reaction, five times higher than in controls. Treatment with NAC before mechanical stress exposure reduced apoptosis to less than 10%. These findings indicate that apoptosis contributes significantly to cyclic compression-induced chondrocyte death and suggest that antioxidants minimize cell death.
Figure 5. Effects of Compression and Antioxidants on Lactate Production. The concentration of lactate in culture medium (mM) was measured in unloaded controls (white columns) or 24 hours after compression with 2 MPa (hatched columns), or 5 MPa (black columns). Explants were not pre-treated (Con) or were pre-treated with SOD, NAC, or catalase (CAT). Columns and bars show means and standard errors based on 6 explants.

by inhibiting apoptosis. The exact mechanism(s) of inhibition are still unclear. Antioxidants might act by mitigating the oxidative damage that would otherwise initiate the apoptosis cascade. Alternatively, antioxidants may block the activation of mitogen activated kinases or other signal transduction pathways involved in the expression of apoptosis-related genes. Further work is needed to determine if this is the case and to assess the effects of antioxidants on chondrocyte necrosis, which might also contribute to cell attrition induced by cyclic compression.

Lactate assays revealed that 5 MPa compression suppressed overall glycolytic activity in explants. These findings paralleled the results of viability assays, suggesting that much of the reduction in lactate production was associated with chondrocyte loss. However, the magnitude of reduction in lactate (~50% loss) was greater than might be predicted from cell losses alone (32% +/- 7%). This suggests that 5 MPa compression inhibited glycolysis in chondrocytes that survived compression.³⁹

Vitamin E, NAC, and SOD all showed similar anti-apoptosis activity in explants exposed to compression with 5 MPa, strongly indicating a role for oxidative damage in this process. The effects of antioxidants were comparable to the effects of L-NAME, the nitric oxide synthase inhibitor that has been shown to inhibit IL-1-induced apoptosis in chondrocytes.³² Although the most significant form(s) of oxidative damage induced by cyclic compression are uncertain, the anti-apoptosis activity of vitamin E suggests that, unlike blunt impact injury, cyclic stress induces lipid peroxidation.²⁶

Compression with 5 MPa was associated with increased accumulated strain, as well as increased peak-to-peak stiffness, which correlated with increased chondrocyte death. Dead cells were found mainly in the superficial zone and the middle zones. The zonel specific distribution of these effects are likely related to zonal variations in the composition of the cartilage extracellular matrix: the relatively low concentration of proteoglycans in the superficial and middle zones leads to reduced compressive stiffness and higher local strains than in the proteoglycan-rich deep zone.⁷

Catalase did not protect chondrocytes in explants from mechanical stress effects. The reasons for this are unclear since catalase is expected to help detoxify hydrogen peroxide, a potentially damaging oxidant produced by chondrocytes. Although it is conceivable that H₂O₂ is not generated during cyclic compression, the failure of catalase to improve viability could be due to more prosaic issues related to its function under the culture conditions we used: the molecular weight of catalase (250 kDa) is more than 10 times as large as the other antioxidants we tested. Thus, it is possible that the inability of catalase to penetrate the cartilage matrix may have impaired any potential chondrocyte-sparing effects.

The mechanisms by which excessive mechanical forces contribute to OA are unknown, stymieing development of effective methods for prevention or treatment. The findings of this study support the hypothesis that the harmful effects of excessive stress and strain are mediated by oxidants. These observations suggest that methods of minimizing acute oxidative damage, including local administration of antioxidants, might decrease the risk for OA, particularly under abnormal mechanical stress conditions that exist at sites of residual articular incongruities following joint trauma.

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REFERENCES


Antioxidants Block Cyclic Loading Induced Chondrocyte Death


ABSTRACT
Indian Hedgehog (Ihh) - Parathyroid related protein (PTHrP) and Fibroblast Growth Factor 3 (FGFR3) signaling pathways are important in regulating endochondral bone formation. In the growth plate, Ihh and PTHrP are involved in a feedback loop to increase proliferation and delay differentiation of chondrocytes. Fibroblast Growth Factor Receptor 3 (FGFR3) conversely decreases proliferation and hastens differentiation with an agonist. Since proliferation is the hallmark of chondrosarcoma cells, we hypothesized that Ihh/PTHrP and FGFR3 pathways may be dysfunctional on these cells. Therefore, we sought to investigate the role of these signaling pathways in the Swarm rat chondrosarcoma cells utilizing expression and functional studies. Semiquantitative RT-PCR analysis demonstrated difference in expression between normal growth plate chondrocytes and chondrosarcoma cells (JWs). JW s had an increased mRNA expression of FGF2 and FGFR3 suggesting a mechanism to reverse the proliferative rate of the cells. Immunohistochemical analysis showed increased staining for FGFR3 and patched-1 (Ihh receptor) in JWs compared to the rat tibia growth plate (p=0.0004 and 0.02 respectively). In vitro functional experiments demonstrated that the use of FGF2, a FGFR3 receptor agonist, dramatically decreased the proliferative rate of Swarm chondrosarcoma cells (LTC). Cyclopamine, a hedgehog inhibitor, did not have a significant effect on their proliferative rate. However, when cyclopamine was used on normal chondrocytes, it effectively decreased the proliferative rate of these cells, suggesting abnormalities in this pathway in the chondrosarcoma cells. In conclusion, our investigation describes dissimilarity in the Indian Hedgehog and FGFR3 signaling pathways between the rat chondrosarcoma cells and native rat chondrocytes. Understanding the underlying mechanisms may provide a target for future therapy for chondrosarcoma.

INTRODUCTION
Chondrosarcoma represents the second most common primary malignant skeletal tumor in adults, accounting for up to 24% of all bone tumors. Despite the new advances in adjuvant therapy, surgical resection is the only effective standardized treatment. Chondrosarcomas do not respond to chemotherapy or radiation therapy, therefore, metastatic disease is rarely amenable to curative treatment. To improve survival rates in patients with high-grade chondrosarcomas, a better understanding of its biology is necessary.

In endochondral bone formation, paracrine ligands Indian Hedgehog (Ihh) and Parathyroid related protein (PTHrP) are important in coordinating the proliferation and differentiation of chondrocytes in the growth plate. They work mutually through an intricate web of local feedback loops. Knockout mice for Ihh or PTHrP have shown pronounced abnormalities of bone growth with histologic evidence of decreased proliferation and increased hypertrophic chondrocytes at the ends of the growth plate. Thus, Ihh and PTHrP regulate the pace of chondrocyte differentiation by keeping the chondrocytes in the proliferative pool and delaying the progression of chondrocytes toward the hypertrophic zone and eventual apoptosis, allowing longitudinal bone growth. Fibroblast Growth Factor Receptor 3 (FGFR3) is also important in the regulation of the growth plate. This is exemplified by three inherited human dwarfism syndromes: hypochondroplasia, achondroplasia, and thanatophoric dysplasia, which are caused by missense mutations in the FGF receptor 3 gene. These mutations lead to different levels of receptor activation, which correlates with the severity of the human phenotypes. The receptor is expressed at the proliferative zone of the growth plate, and is activated by FGF2 and other presently unknown ligands to directly reduce the prolifera-
The current understanding of the PTHrP, Ihh, and the FGFR3 signaling pathways associated with endochondral bone formation is depicted in Figure 1. These pathways play crucial roles in regulating bone growth and differentiation of chondrocytes. PTHrP and Ihh are involved in maintaining the proliferative pool of chondrocytes and suppressing Ihh expression, respectively. FGFR3 directly increases the rate of proliferation and suppresses Ihh expression.

Several lines of evidence suggest that these signaling pathways may be deregulated in cartilage tumors. A mutant PTH/PTHrP type I receptor was identified that signals abnormally in vitro and causes enchondroma-like lesions in transgenic mice. Immunohistochemical studies of human chondrosarcoma showed increased PTHrP ligand expression with increased tumor grade. Another investigation showed co-expression of PTHrP and PTH/PTHrP receptor in chondrosarcoma and that its positivity may be valuable for differentiating between benign and malignant cartilaginous tumors. In addition, treatment of human chondrosarcoma cells in vitro with anti-parathyroid hormone-related protein monoclonal murine antibody accelerated apoptosis of chondrosarcoma cells in a dose-dependent manner.

In this study, we investigated the role of these signaling pathways in chondrosarcoma. Specifically, we focused on the Ihh / PTHrP and the FGFR3 pathways, which are summarized in Figure 1. We hypothesized that their expression and functionality are deregulated in chondrosarcoma cells compared to normal growth plate chondrocytes. We conducted analysis of expression at the molecular and histologic level using a Swarm rat chondrosarcoma cell line and growing epiphyseal cartilage obtained from <5-week-old rats as controls. We also conducted a functional study in vitro to assess whether manipulation of these pathways will affect the proliferative activity of chondrosarcoma cells.

**MATERIALS AND METHODS**

**Isolation of total RNA**
Sprague-Dawley rats were obtained from Harlan Laboratories (Indiana, U.S.) and euthanized between 2-5 weeks of age. The growing epiphyseal cartilage from the head of the femur and proximal tibia was obtained free of soft-tissues. A Swarm rat chondrosarcoma tissue line (JWS) that has been maintained through the years by...
serial subcutaneous injections was used for this study. Total RNA was extracted as previously described. Samples were purified further by using Rneasy (Qiagen, Valencia, CA, U.S.). RNA concentrations were estimated by measurements of absorbance at 260nm.

**Semi-quantitative reverse-transcription polymerase chain reaction**

RT-PCR was performed using the OneStep RT-PCR kit (Qiagen, Valencia, CA, U.S.). Reverse transcription was run at 50°C for 35 min. The PCR condition was 95°C for 15 minutes, followed by 20-35 cycles of 94°C for 1min, 55°C (varied with primer) for 1 min, and 72°C for 1min. Different cycles were used to determine optimal condition for analysis and were all in the linear range of the polymerase chain reaction. B-actin was used as the internal control (Table 1). The amplified material was run on 2% agarose gel and visualized with ultraviolet light and photographed. QIAquick PCR purification Kit (Qiagen, Valencia, CA) was used to purify the PCR product. Product sequence was obtained with an automated sequencer at the University of Iowa DNA facility and analyzed using the NCBI Blast search engine. We compared the intensity of the bands from epiphyseal growing cartilage and chondrosarcoma for each primer set by analyzing the luminosity of the bands using a histogram with Adobe photoshop 6.

**Immunohistochemistry**

Before sacrificing the rats to obtain tibias for immunohistochemistry, Swarm rat chondrosarcoma cells were implanted into the diaphysis of the tibia and allowed to grow and permeate the bone marrow for 10 – 14 days. Rats were then sacrificed and tibias were removed and fixed in 10% buffered neutral formalin. The fixed specimens were dehydrated in a graded series of ethanol, cleared in xylene, and embedded in paraffin. Four-micron thick sections were cut on a rotary microtome and mounted on Fisher Brand Superfrost/Plus microscope slides (Fisher Scientific, Pittsburg PA). Standard peroxidase-labeled streptavidin-biotin detection method was used. Sections were stained with safranin O (The Blakiston Company, Inc. New York, US). The sections were incubated in blocking solution [10% normal horse serum, 1% bovine serum albumin, 0.1% Tween 20 in PBS]. Blocking solution was then removed and sections were incubated overnight at 4°C in a humid chamber with appropriate dilution of primary antibodies (FGFR3, 1:200, Santa Cruz Biotechnology, Inc., California US; and Ptc-1 receptors, 1:500, Orbigen Inc., California, US). The next day, samples were incubated with secondary antibodies (biotinylated goat anti-rabbit, Sigma, Missouri, US) diluted 1:800 in blocking solution. Finally, Vectastain Elite ABC kit (Vector Laboratories, California, US) was utilized and sections were then reacted with the chromagen diaminobenzidine from the DAB kit from Vector.
Labs (Burlingame, CA, US). Negative control slides were incubated without primary antibodies.

**Cell Culture**

The Swarm rat chondrosarcoma cell line LTC\(^{14}\) was cultured in high glucose Dulbecco’s Modified Eagle medium (DMEM) supplemented with fetal bovine serum (12%), 1M HEPES (25mM), and gentamicin (50µg/ml) and grown in humidified air with 5% CO\(_2\) at 37°C. Passage of cells was accomplished using 0.25% trypsin-EDTA (pH 7.2-7.4, concentration = 0.02%, Sigma Chemical Co., Missouri, US). Rat growth plate chondrocytes were obtained from the femoral cartilage cap of 2-week old rats. The cells were extracted from tissue by incubating in CO2 incubator in hyaluronidase solution (testicular hyaluronidase 0.1%, DMEM, HEPES 25mM) for 30 minutes and then in a collagenase solution (collagenase 0.5 mg/ml, fetal bovine serum 10%, HEPES 25mM, gentamicin 50 µg/ml) overnight. The epiphyseal growth plate chondrocytes were cultured in the same way as the LTC cells.

**Cell count assay**

For testing the effect of agonists/antagonists on cell proliferation, LTCs or normal chondrocytes were plated in 24-well plates (75,000 - 100,000 cells/well) in a total volume of 1 ml of culture medium per well. For studies utilizing FGF2, a serum-free medium containing DMEM, 1M HEPES (25mM), 100X ITS (1x, Sigma, MO, US), BSA (1%, Amresco, OH, US), and gentamicin (50µg/ml) was used. Cells were allowed to adhere for 4 hours prior to addition of the agents. Afterwards, variable amounts of agents were added to six replicate wells and incubated for a total of 48 hours. The agents used were FGF2 (PeproTech Inc., NJ, US), Cyclopamine (BIOMOL Research Laboratories Inc., PA, US), and FGFR3 antibody (Santa Cruz Biotechnology, Inc., California US) at varying concentrations. After incubation, the culture media was removed and cells were collected by trypsinisation and counted using a hemocytometer. For cell viability, the trypan blue exclusion assay (0.4%) was used. The effect of agents was determined by evaluation of cell growth and viability in comparison to untreated controls.

**Statistical Analysis**

All gene expression and in vitro cell culture experiment data are expressed as the mean ± SEM and statistical analysis was performed using ANOVA. For the immunohistology study, statistical analysis was performed using the Repeated Measures Analysis of Variance.

**RESULTS**

**Semi-quantitative gene expression analysis**

Semi-quantitative reverse-transcription polymerase chain reaction (RT-PCR) was performed to analyze the expression of Ihh, Ptc-1, PTHrP, PTH/PTHrP receptor, FGF2, and the FGFR3 in Swarm rat chondrosarcoma and in the postnatal growing epiphyseal cartilage. Transcripts for Ihh, Ptc-1, PTHrP, PTH/PTHrP receptor, and the FGFR3 were detected in both the chondrosarcoma cells and the normal chondrocytes with the correct base pair size on gel electrophoresis. Semi-quantitative analysis of gene expression of paracrine ligands Ihh, PTHrP,
FGF2, as well as FGFR3 was higher in the swarm rat chondrosarcoma cells compared to normal chondrocytes (p = <0.01). On the other hand, the expression of PTH/ PTHrP receptor and PTC-1 receptor were lower in rat chondrosarcoma compared to normal chondrocytes (p = <0.01) (Figure 2).

**Immunohistochemical evaluation of FGFR3, PTC-1, and PTHrP/PTH receptor**

Immunohistochemistry was performed to study the expression patterns of the corresponding receptors using tibia obtained from 5 weeks old rats with Swarm rat chondrosarcoma implanted into the tibia. Sections were stained with antibodies specific for PTC-1, PTHrP/PTH and FGFR3 receptors, since the growth plate and the chondrosarcoma tumor tissue are both in the same tibia. The staining was done concomitantly, thus allowing us to do semi-comparative studies. Staining with the antibody against FGFR3 showed higher staining in the chondrosarcoma tumor tissue compared to the growth plate (Figure 3). Using the modified Mankin scoring, an average score of 1.446 ± 0.165 was obtained for chondrosarcoma tumor tissue and a score of 0.458 ± 0.257 for the growth plate was obtained with antibody specific for FGFR3. The p-value was 0.0004. An average score of 1.376 ± 0.215 for chondrosarcoma tumor tissue and a score of 0.987 ± 0.193 for the growth plate was obtained with antibody specific for PTC-1. The p-value was 0.0266.

Figure 3. Immunohistochemical examination using antibodies specific for PTC-1, PTH/PTHrP receptor, and FGFR3. (A) Staining of rat chondrosarcoma (JWS) embedded in tibia. (B) Staining of 5 week old rat tibial growth plate. Staining of both the growth plate and chondrosarcoma were performed concomitantly since they were on the same tibia. Chondrosarcoma tissue had noticeably higher intensity of staining than in the rat growth plate with all antibodies. Also, notice the heterogeneity of staining of the rat chondrosarcoma. Controls were stained without the primary antibody.

Figure 4. Comparative study between rat chondrosarcoma tissue and rat postnatal growth plate with immunohistochemistry utilizing antibodies specific for PTC-1 and FGFR3 receptor utilizing the modified Mankin score. (A) Average Mankin score of 1.446 ± 0.165 for chondrosarcoma tissue and a score of 0.458 ± 0.257 for the growth plate was obtained with antibody specific for FGFR3. The p-value was 0.0004. (B) Average Mankin score of 1.376 ± 0.215 for chondrosarcoma tissue and a score of 0.987 ± 0.193 for the growth plate was obtained with antibody specific for PTC-1. The p-value was 0.0266.
Staining with the antibody against PTHrP/PTH also showed higher staining in the chondrosarcoma tumor tissue compared to the growth plate. In addition, it was observed that there was heterogeneity in staining of chondrosarcoma tumor cells with all three antibodies.

**Functional studies**

*In vitro* studies using Swarm rat chondrosarcoma cells (LTC) treated with fibroblast growth factor basic (FGFb or FGF2), a Fibroblast Growth Factor receptor 3 (FGFR3) agonist, demonstrated that cell numbers after 48 hours of incubation were 40-60% less than in the untreated control suggesting decrease or stasis in the proliferative rate (Figure 5). The average number of cells in the control group doubled to 352,000 ± 25,600 whereas FGF2 5nM treated group had an average cell number of 165,000 ± 3,870. Addition of polyclonal antibody of FGFR3 to FGF2 treated cells did not reverse the effect of FGF2.

LTC treated with cyclopamine, an agent that inhibits the hedgehog signaling pathway, had no statistically significant effect on its proliferative rate. The average number of cells in the control group was 254,000 ± 36,300 whereas as cyclopamine 40uM treated group was 231,000 ± 16,000 at 48 hours (p = <0.01). However, cyclopamine seems to have a statistically significant effect on the proliferative rate of growth plate chondrocytes by decreasing the proliferative rate of these cells. The non-treated control group had an average cell number of 89,700 ± 6,980 at 48 hours whereas the number of cells in the cyclopamine 40 uM treated group was 45,000 ± 9000 at 48 hours (p = <0.01).
DISCUSSION

Many genes and signaling pathways have been identified that are crucial for the proper development of chondrocytes and bone. Indian hedgehog (Ihh) is considered one of the key regulators of bone development by coordinating chondrocyte proliferation, differentiation and osteoblast formation.\textsuperscript{1,2,16,17} Ihh is one of the three mammalian Hedgehog (Hh) proteins and is closely related to Sonic hedgehog, an essential secreted protein involved in regulation of limb outgrowth.

It is currently thought that Ihh is expressed in the prehypertrophic chondrocytes of cartilage elements, where it regulates the rate of hypertrophic differentiation. In one study, Ihh activity was reduced by Cre-loxP approach that specifically removed the Smoothened (Smo) transmembrane protein activity that transduces all Hh signals. It was observed that animals treated by this means developed shorter long bones, and demonstrated a 50% reduction in chondrocyte proliferation.\textsuperscript{18}

Other studies have focused on the use of cyclopamine, a steroidal alkaloid derived from the plant \textit{Veratrum californicum} that effectively binds to Smo, to specifically block the cellular response to Hh signaling in other models.\textsuperscript{19-21} However, no study to date has looked at the involvement of the Ihh signaling pathway in chondrosarcoma mitogenesis. Since previous studies demonstrated that Ihh activity induces proliferation of chondrocytes, we wanted to assess the effect on chondrosarcoma cells if the Ihh function was blocked.

In this study, we used cyclopamine to block the activity of Ihh in cell culture experiments. Interestingly, although cyclopamine effectively decreased the proliferative rate of control growth plate chondrocytes, it did not influence the proliferative rate of chondrosarcoma cells. These results suggest that there may be a deregulation in the Ihh signaling pathway in the Swarm chondrosarcoma cells that alters the signaling cascade associated with this pathway. Since cyclopamine inhibits Smo, it is possible that other mechanisms in these chondrosarcoma cells may be affected downstream this protein to maintain Gli transcriptional activation of the genes in this pathway.\textsuperscript{22}

Recent studies have shown that FGF signaling also crucially regulates chondrocyte proliferation and differentiation, of which signaling involving FGFR3, a tyrosine kinase receptor, is the best understood. FGFR3 is a negative regulator of bone growth\textsuperscript{14} and one of the effects is to down regulate the Ihh pathway.\textsuperscript{15} Knockout of FGFR3 in mice leads to an increased rate of proliferation of chondrocytes and an activating point mutation in FGFR3, found in human chondrodystrophies such as achondroplasia, decreases the rate of proliferation of chondrocytes.\textsuperscript{23,24} In one study, it has been found that activation of FGF signaling inhibits chondrocyte proliferation both in a rat chondrosarcoma cell line and in primary murine chondrocytes through phosphorylation of STAT-1.\textsuperscript{25} Also another study, using microarray and biochemical analyses of FGF-treated rat chondrosarcoma chondrocytes, showed that FGF inhibits chondrocyte proliferation by initiating multiple pathways including dephosphorylation of the retinoblastoma protein (pRb) p107 and repression of a subset of E2F target genes.\textsuperscript{26}

Our study found similar results with the use of FGF2, and FGFR3 agonist, in which it reduced the proliferative rate of chondrosarcoma cells. However, the addition of a polyclonal antibody for FGFR3 did not reverse the affect of FGF2, which may suggest also a deregulation in the FGFR3 signaling pathway. Additionally, our descriptive study found that there was increased expression of FGF2 and the FGFR3 in chondrosarcoma chondrocytes compared to growth plate chondrocytes at the molecular and histologic level. Thus it seems that chondrosarcoma chondrocytes are responding appropriately to try and reduce the proliferative rate, to no avail. This suggests further that there may be a deregulation of the FGFR3 pathway and more studies are needed to further elucidate the mechanism of action.

In conclusion, our investigation describes dissimilarity found in Indian Hedgehog and FGFR3 signaling pathways between the rat chondrosarcoma cells and native rat tissue chondrocytes. Understanding the underlying mechanisms may provide a target for future therapy for chondrosarcoma.

ACKNOWLEDGMENT

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REFERENCES


HABITUAL HIP JOINT ACTIVITY LEVEL OF THE PENNED EMU
(DROMAIUS NOVAEHOLLANDIE)

Karen L. (Reed) Troy*‡#, Hannah J. Lundberg*‡§, Michael G. Conzemius+†, Thomas D. Brown‡*

ABSTRACT
Orthopaedic management of femoral head osteonecrosis remains problematic, partly because of inability to systematically compare treatments in an animal model whose natural history parallels the human in terms of progression to femoral head collapse. Recently, it was determined that collapse could be consistently achieved for cryogenically induced osteonecrosis in the emu. Toward delineating the comparative hip joint biomechanics of emus versus humans, for purposes of establishing the emu as a model for human femoral head osteonecrosis, habitual hip joint activity level was quantified for a group of seven healthy adult emus housed in an outdoor research pen typical of those used in emu farming operations. The daily number of steps taken, and the time spent with the hips loaded (standing, or squatting/sitting) versus unloaded (recumbent), were quantified from 24-hour videotape recordings, analyzed by four independent observers. The average number of steps taken per day was 9,563, which extrapolates to 1.8 million hip loadings per year, a value that falls in the same general range as seen in normal adult humans. On average, the emus spent 4:05 hours per day idly standing, 2:12 hours squatting/sitting, and 10:44 hours recumbent; they underwent an average of 37 transitions per day between the respective posture/activity states.

INTRODUCTION
Osteonecrosis of the hip remains an important unsolved problem in the field of orthopaedic surgery. This disorder is caused by a disruption in blood perfusion to the cancellous bone of the femoral head (occurring for any of several reasons), and results in the death of often large regions of weight-bearing bone. Left untreated, osteonecrosis generally progresses to femoral head collapse and subsequent secondary osteoarthritis.1 Due to the mechanical nature of the collapse phenomenon, interventions to hopefully preclude its occurrence ideally should be systematically evaluated at the whole-joint level, under a loading regime as close as practicable to that in humans.

Many animals models have been developed to study pharmacologically- or surgically-induced osteonecrosis. Species utilized have included dogs, rabbits, pigs, horses, goats, rats, and mice.2-9 However, while (usually) successful in reproducing early tissue-level pathology, none of these models has managed to replicate the clinically all-important phenomenon of progression to femoral head collapse. Plausibly, the reason for non-progression has been that all of these species have been quadrupeds, which has allowed spontaneous load-protection of the affected limb, thereby presumably involving lesser hip joint mechanical demand that that typical of the human.

Recently, Conzemius et al.10 demonstrated that cryogenically-induced osteonecrosis progresses to femoral head collapse in the emu, a large flightless (bipedal) bird native to Australia. Emus (Figure 1) are the second-largest member of the ratite family, which along with their bigger cousin the ostrich also includes rheas, cassowaries, and kiwis. Adult emus typically stand about 5 feet (150 cm) tall, and weigh about 100 pounds (440 N). Although not a species previously utilized in biomedical research, emus are potentially attractive for studying various musculoskeletal disorders where bipedal gait is an important consideration, and where human-size-relevant surgical interventions are of interest. Emus nowadays are farmed commercially in many areas of the US as a source of meat and (especially) of cosmetic oils, so they are inexpensive to procure. They also are relatively easy to maintain, requiring only a penned outdoor enclosure/run with sufficient space to freely ambulate year-round, and with shelter available for times of inclement weather.

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Although osteonecrosis in the Conzemius et al. model was histologically and morphologically similar to that in the human, the time needed to achieve collapse in that series averaged twelve weeks. This is substantially faster than the average time of twenty-three months needed for untreated human femoral head osteonecrosis to progress to collapse. Understanding the reason(s) for the faster-than-human natural history of osteonecrosis progression in the emu is obviously an important consideration for interpreting results from studies potentially using that species as a disease model.

Activity level difference stands out as one obvious possibility. To the casual observer, penned emus appear to relentlessly pace in their enclosure, spending very little time with their hips unloaded. Although most of the lesions induced in that earlier series by Conzemius et al. were more severe than those in many human cases, the emu’s apparent high activity level therefore might have contributed appreciably to pathogenesis acceleration. Moreover, in the Conzemius et al. study, histology for two of the three animals that did not develop lameness showed end-stage osteonecrosis, suggesting that emus either experience less pain with structural collapse, or that they are better able to tolerate hip pain, than humans. Besides osteonecrosis, the emu’s seeming stoic behavior may also prove useful for modeling other musculoskeletal disorders (e.g., osteoarthritis) whose pathophysiology is influenced by unprotected loading of the involved extremity. Therefore, in addition to helping understand the potential role of the number of loading cycles and the duration of static hip loading as contributors to the rapidity of emu necrotic femoral head collapse per se, an appreciation of the daily activity levels of emus would be informative in the broader context of this species being considered for modeling other musculoskeletal disorders.

For these reasons, a study was undertaken to formally catalog the daily activity levels of emus. An important caveat to the high activity level apparent on casual observation is that emus are at best semi-domesticated animals, that react apprehensively to interactions with humans. An observer being visible to the emus would potentially constitute a disturbance that could result in a substantial artifactual increase of their habitual activity level. The study design therefore relied upon a data capture protocol that involved minimal human presence.

**METHODS**

Seven normal adult emus were sedated and marked with spray paint for individual identification. The animals were housed within a 28.2 x 2.8 m outdoor enclosure (Figure 2) that contained a 5.3 x 2.8 m covered shed at one end. This overall enclosure area corresponded to 11.25 m² per animal. They were allowed food and water ad libitum. These conditions were similar to those typically used in emu farming operations, and were approved by the Iowa State University Institutional Care and Use Committee. Three video cameras were placed along the fence of the enclosure: one in the covered shed, one next to the feeder, and one at the opposite end of the pen, looking towards the shed.

A week before the videotaping, one observer (HJL) spent the night inconspicuously observing the emus in their enclosure, to gain an appreciation of their nocturnal activity level. It was determined that around-the-clock videotaping was appropriate, since the animals occasionally got up and walked around at night, although in general such activity was appreciably less than in daylight hours. To facilitate nocturnal videotaping, three small incandescent lights were placed along the enclosure fence, and one light was placed inside the shed, providing just enough illumination to distinguish between emus, while not substantially disturbing their nocturnal behavior.

Videotaping commenced at 3:45 PM. At approximately 9:00 PM the lights were turned on and the cameras were loaded with new tapes. Tapes were exchanged again.
at approximately 4:30 AM, which caused the emus to awaken and stand for several minutes before returning to sleep. At approximately 10:30 AM the tapes were exchanged a third time. Videotaping was stopped at 3:50 PM. The experiment was conducted during a period of clear summer weather.

Quantifying the video recordings proved to be a substantial undertaking. Due to the animals’ frequent intermingling, only one individual could be tracked at any given time. Thus, a total of 504 hours of videotape (3 cameras x 7 emus x 24 hours) needed to be analyzed. Moreover, owing to the seemingly random motions of the animals from site to site within the pen, it was frequently necessary to switch from tape to tape, with corresponding tape rew windings and/or fast forwards to preserve time synchrony. Because of the length and tediousness of videotape analysis, the task was split up among four observers. Each observer processed data (three cameras’ tapes) from one of the four (approximately 6-hour) time segments recorded. For each emu the number of steps taken (both walking and running), and the time spent sitting/squatting and lying (Figure 3) were quantified. Occasionally, animals mingled together into tight clusters, such that the particular animal that was being observed was obscured. During the intervals that a particular animal was not distinctly visible on any of the three tapes, its time spend standing, sitting/squatting, and lying, and its numbers of steps were estimated in context by the observer, based on all of the animals’ activity patterns. Cumulative results of all four observers were then merged to produce a 24-hour compilation of activity levels for each of the seven emus. To verify that observers were counting steps and measuring the standing, sitting/squatting, and lying periods in a consistent manner, three emus in three 30-minute representative segments of video were analyzed by all four observers. The number of steps taken, and the time spent standing, sitting/squatting, and lying, were compared using Kendall’s W coefficient of concordance, a measure of observer agreement. In this statistic, an extremum of 1.0 indicates perfect agreement, and 0.0 indicates absolute disagreement.

As an additional point of reference, two emus that had undergone osteonecrosis-inducing cryo-surgery four weeks previously were videotaped for a two-hour period. These two animals were housed indoors in 3 x 3 m pens, the same management procedure as used in the earlier series. These two surgery animals were videotaped during daytime hours. Their numbers of steps, and their times spent sitting/squatting and lying down, were quantified by one of the four observers (KLT). These data were then extrapolated to estimate an average of these quantities per hour, for comparison to the non-operated emu cohort.

RESULTS

The Kendall’s W statistic for the 30-minute comparisons between all four observers was 0.845 (p=0.0004), which indicates very strong inter-observer agreement. This agreement was judged sufficient to compile the results of the entire 24-hour period as if a single observer had analyzed all the data.

On average, the non-operated emus took 9,653 steps per day (Table 1). This figure extrapolates to 1.8 mil-
lion loading cycles per hip per annum. An average of 10 hours and 45 minutes per day was spent in a recumbent (hips unloaded) position. For this lying posture, since interest was in the hip joint, no distinction was made between the alert (head held up) versus sleeping (head tucked under the wing or on the ground) states. Both idle standing time and sitting/squatting time represent conditions of static hip loading, and plausibly contribute to osteonecrotic femoral head collapse. The emus spent an average of four hours, five minutes each day standing (without walking), and two hours, thirteen minutes sitting/squatting with their hips loaded. Idle standing time was estimated by assuming that a single step took one second to complete. (This one-second average step duration was estimated by measuring the time and step count for several typical walking episodes). On average, emus had 37 transitions per day between sitting/squatting, standing, and/or lying.

Not surprisingly, activity level varied appreciably over the course of the day (Figures 4 and 5). The animals were most active in the afternoon to mid-evening period (recording period 1, and the first part of period 2). For the most part, they slept through the night (most of recording period 2), occasionally rising for short periods of time. Some animals were substantially more active than others, with one taking as many steps during recording period 1 as another did during the entire 24-hour period. The early morning (recording period 3) had the most variability in how much time the animals spent lying down, presumably because some awoke earlier than others.

In the two-hour videotape of the two animals that had undergone cryo-insult surgery, one took 2,955 steps, and the other took 3,981. The animal that took fewer steps spent 28 minutes lying down, but the other one remained standing for the entirety of the two-hour videotape. The corresponding 1,478 and 1,991 steps per hour for these two animals are labeled with by asterisk in Figure 4, for comparison to the non-operated animals.

Over the course of cataloguing the 24-hour videotapes, it was noted that emus often arbitrarily paused during walking, while midway through weight shift from one leg to the next. Often they stood frozen in this posture for multiple seconds, and even tens of seconds, before either continuing on with the next step, or shifting the weight back to the trailing leg to adopt a two-legged stance.

Table 1. Summary of Activity Data Compiled for All Seven Animals over a 24-hour Period

<table>
<thead>
<tr>
<th>Animal #</th>
<th>Steps (L+R)</th>
<th>Sitting Time</th>
<th>Lying Time</th>
<th>Idle Standing Time</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>7894</td>
<td>3:04</td>
<td>12:40</td>
<td>3:36</td>
</tr>
<tr>
<td>2</td>
<td>9375</td>
<td>0:22</td>
<td>13:31</td>
<td>4:25</td>
</tr>
<tr>
<td>3</td>
<td>6926</td>
<td>2:04</td>
<td>10:54</td>
<td>2:50</td>
</tr>
<tr>
<td>4</td>
<td>4371</td>
<td>5:18</td>
<td>9:13</td>
<td>4:05</td>
</tr>
<tr>
<td>5</td>
<td>15809</td>
<td>2:10</td>
<td>7:20</td>
<td>6:10</td>
</tr>
<tr>
<td>6</td>
<td>11488</td>
<td>0:50</td>
<td>12:51</td>
<td>2:55</td>
</tr>
<tr>
<td>7</td>
<td>11708</td>
<td>1:37</td>
<td>8:37</td>
<td>4:31</td>
</tr>
<tr>
<td>Average</td>
<td>9653</td>
<td>2:12</td>
<td>10:44</td>
<td>4:05</td>
</tr>
<tr>
<td>St. Dev.</td>
<td>3742</td>
<td>1:15</td>
<td>2:24</td>
<td>1:05</td>
</tr>
</tbody>
</table>

Not surprisingly, activity level varied appreciably over the course of the day (Figures 4 and 5). The animals were most active in the afternoon to mid-evening period (recording period 1, and the first part of period 2). For the most part, they slept through the night (most of recording period 2), occasionally rising for short periods of time. Some animals were substantially more active than others, with one taking as many steps during recording period 1 as another did during the entire 24-hour period. The early morning (recording period 3) had the most variability in how much time the animals spent lying down, presumably because some awoke earlier than others.

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Over the course of cataloguing the 24-hour videotapes, it was noted that emus often arbitrarily paused during walking, while midway through weight shift from one leg to the next. Often they stood frozen in this posture for multiple seconds, and even tens of seconds, before either continuing on with the next step, or shifting the weight back to the trailing leg to adopt a two-legged stance.

**DISCUSSION**

Historically, formal quantification of activity level has usually been lacking in animal studies where functional loading is important (such as in the case of physical loading stimuli to bones and joints). In the past several years, however, much interest has arisen in cataloging the habitual loading activities both in humans and in experimental animals such as turkeys, dogs, and sheep. For example, Adams et al. used 24-hour videotaping to document the daily activity patterns responsible for mechanical homeostasis of turkey ulnae. That work was expanded by Fritton et al., who placed strain gauges on the tibiae and ulnae of turkeys, dogs, and sheep, to record 12-24 hour loading histories. Both of those studies were conducted to quantify the numbers and magnitudes of strain-producing activities that contribute to bone maintenance versus remodeling. Habitual activity levels in humans have been documented in a number of studies, primarily in the context of assessing polyethylene wear rates in total hip arthroplasty.
The manual cataloguing methodology used in the present study is similar to that used by Adams et al. In each tape, only one emu could be followed at a time, due to their intermingling. Because of the sheer volume of data generated by the videotapes (3 cameras x 7 emus x 24 hours) and the tediousness of the cataloguing measurements, it was necessary to divide the work among several observers, whose consistency it was necessary to verify with Kendall’s W statistic. Adams et al. resorted to similar usage of multiple observers, linked via statistical documentation of inter-observer reproducibility, when categorizing wing activity events of turkeys.

Because of the fast progression to femoral head collapse seen in the pilot study by Conzemius et al., it was initially supposed that emus would be documented to have substantially higher habitual activity levels than humans. However, the measured average number of steps per day taken by healthy emus (9,653) turns out to be strikingly consistent with the average number of steps that a healthy human takes each day (10,400 for men and 8,900 for women, and 8804 for men and 8913 for women, respectively). The present emu data fall near the middle of the range reported by Goldsmith et al. for healthy adult humans (395 to 17,718 steps per day). A Student’s t-test showed that emus do not take significantly more steps than humans (p=0.55), when compared to data obtained by Sequeira et al. Although humans are highly variable, the ~11 hours per day that...
Emus spend with their hips bearing weight empirically seems appreciably greater than the corresponding period for most humans (and may have contributed to the quick progression to femoral head collapse in the pilot study). While the initial supposition that emus are more active (in terms of number of daily loading cycles on the hip) than humans was clearly not borne out by the data, this similarity in activity levels may prove useful in terms of future comparisons between the two species.

Goldsmith et al. and Schmalzried et al. both remarked on the high levels of inter-subject and intra-subject variability in their studies. The ratios of maximum to minimum number of steps per day in these two studies were 5:1 and 45:1 respectively. (The latter high number is likely due to the very low number of steps by the least-active individual in that particular study). The present study in emus shows only a 3.6:1 ratio, and suggests that emus have less variable activity levels than do humans.

While the average number of loading cycles per day for normal emus approximated that seen in humans, it is important to note that the human data described above were for normal (healthy) subjects. As regards human osteonecrosis patients, we are not aware of any studies that have examined their daily loading regime. While osteonecrosis patients tend to be young “high demand” individuals, many have co-morbidities, and almost certainly their activity level tends to decrease when the hip becomes symptomatic.

Concern was allayed as regards operated-upon animals potentially having reduced activity levels, either due to increased pain or due to the change in environmental conditions (indoors versus outdoors). While the initial sample size described here (two animals for a two-hour period) is insufficient to draw formal statistical conclusions about the overall activity level of post-operative versus pre-operative animals, it does provide anecdotal evidence that activity levels after surgery remain reasonably high.

Goldsmith et al., whose study methodology utilized pedometry, noted that activity levels varied seasonally for the younger of two subjects whom they monitored over the course of a year: activity was highest during the summer. The videotaping in the present emu study was performed in midsummer during clear weather. Variations in emu activity due to weather would obviously affect these estimates of emu yearly activity. These animals are typically housed outdoors year-round. It seems a reasonable assumption that they would spend more time in the covered area of their pen during wet or colder weather, which presumably would involve less activity than that presently reported. Although daily life in an emu pen seemingly holds less variety of hip joint loadings than do the lifestyles of most humans (e.g., stair-climbing, sit-to-stand, stooping to pick something up, etc.), the emu videotapes nevertheless showed a range of occasional activities seemingly involving high hip demand. These included episodes of running, jumping, confrontations between birds (kicking, scratching, etc.) plus the numerous (average 37 per day) sit-to-stand transitions. Our impression is that the variety of emu hip loadings is not substantially different from that of humans, and probably exceeds that of sedentary individuals.

Of the various indices of activity level here measured, the number of daily loading cycles seems most relevant to studying osteonecrosis, since femoral head collapse is probably a fatigue-related process, driven by cyclic fluctuation of stress in the at-risk osseous lattice. The measured average number of emu daily loading cycles, which extrapolates to 1.8 million per year, is remarkably similar to the recent estimates for healthy humans of 1.7 million per year by Seedholm and Wallbridge and of up to 2.0 million per year estimate by Silva et al.

Finally, the above observations and data of course apply only to emus housed in a farm-type pen enclosure. However, the activity level of emus in the wild, while presumably higher, is of lesser practical interest in the context of this animal serving as a model for necrotic femoral head collapse, because any individual animal potentially used for surgical research would probably be housed in such a (sheltered) pen, or for short durations in an indoor enclosure, rather than being free-ranging.

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REFERENCES

EFFECT OF CAST REMOVAL TIMING IN THE CORRECTION OF IDIOPATHIC CLUBFOOT BY THE PONSETI METHOD

Gaston Terrazas-Lafargue, M.D., and Jose A. Morcuende, M.D., Ph.D.*

ABSTRACT

Clubfoot correction by the Ponseti method is highly successful with an average of 5 casts in the majority of patients. However, early experience with the technique demonstrated that many cases required more than 5 casts for correction. The purpose of this study was to evaluate the effects of timing of cast removal before subsequent manipulation and casting in the correction of clubfoot using the Ponseti method. We reviewed 90 patients (129 clubfeet) treated between December 2000 and June 2006. Two groups were evaluated. Group A: 44 patients (63 clubfeet) had their cast removed by the parents the night before the visit. Group B: 46 patients (66 feet) had their cast removed in the clinic just before the new cast is applied. Overall, full correction was obtained in 128 (99%) clubfeet, with only 1 clubfoot requiring surgical release. Group A required an average of 10 casts (range: 4 to 22), compared to 5 casts (range: 4 to 10) in Group B (p=0.001). Average time for correction was 13 weeks in Group A and 6 weeks in Group B (p=0.001). There were 20 relapses (22%), 12 (27%) in Group A and 8 (17%) in Group B (p=0.42). In conclusion, removing the cast just before the new cast is applied significantly decreases the number of casts required for correction and shortens the length of treatment.

INTRODUCTION

Idiopathic clubfoot is the most common musculoskeletal birth defect, affecting an average of 1 in 750 newborns. Most orthopedists agree that initial treatment should be non-surgical and started soon after birth. In the past few years, several reports have demonstrated successful correction in >95% of the clubfeet using the Ponseti method.1-8 Interestingly, there is some variation in the reported average number of casts required to obtain full correction. At our institution in Chile, early experience with this method demonstrated that many patients required more than 5 casts for correction. We hypothesized that these feet were more severely involved, therefore, requiring more casts. However, it has been our practice to request the parents to remove the cast the night before the visit to allow for skin care. This recommendation differs from the Ponseti method which requires the cast to be removed just before the new cast is applied. The purpose of this study was to evaluate the effects of timing of cast removal in clubfoot correction by the Ponseti method.

MATERIALS AND METHODS

We reviewed the records of 90 patients with congenital idiopathic clubfoot (129 clubfeet) consecutively treated by one of the authors (GTL) between December 2000 and June 2006. Positional and syndromic clubfeet were not included. Institutional review board approval was obtained. No patient has been lost to follow-up.

The treatment technique followed the principles of correction described by Ponseti.19-21 However, at our institution we have traditionally asked the parents to remove the cast the night before the visit to allow for skin care. Therefore, our method should be considered a modification of the Ponseti protocol that requires the cast to be removed just before the new cast is applied. Importantly, after a visit to the University of Iowa in October 2003, we modified our cast removal protocol to precisely follow the Ponseti recommendations. Based on this change, we evaluated two groups of patients based on the timing of cast removal. Group A: 44 patients (63 clubfeet) were studied retrospectively. These patients had their cast removed by the parents the night before the visit. Group B: 46 patients (66 feet) were studied prospectively. They had their cast removed in the clinic just before the new casts were applied.

We evaluated the following variables: age of the patient at first visit to our institution; previous treatment before referral: type, number of casts, tendoachilles tenotomy; number of casts required at our institution; need for percutaneous tendoachilles tenotomy; degree of ankle dorsiflexion after tenotomy; and compliance with the foot abduction brace. These variables were in turn related to the need for extensive corrective surgery and
the incidence of relapses. Fisher’s exact tests, t-tests, and odds ratios were used as appropriate.

**RESULTS**

Sixty-four patients (71%) were males. The majority of patients were otherwise healthy (72%) and most children (92%) were full-term, without complications during pregnancy or delivery. Seventy-nine patients (60%) were first-born. Thirty-three patients (22%) had a positive family history of clubfoot deformity. Developmental dislocation of the hip was observed in 10% of the patients. At initial Ponseti casting, all patients but three were younger than 6 months old.

Three patients had some manipulations and casting before their initial visit to our institution. None of these patients had a percutaneous tendoachilles tenotomy. These three patients came to the clinic with all the components of the deformity uncorrected. Clubfoot correction was obtained in all clubfeet but one (n=128, 99% correction). This patient from Group A had a very stiff, bilateral deformity.

As shown in Figure 1, there was a significant difference between groups in the average number of casts required for correction: 10 casts (range, 4 to 22) in Group A, and 5 casts (range, 4 to 10) in Group B (p=0.001). In addition, there was a significant difference in the time from first cast to correction. In Group A, it took an average of 13 weeks compared to 6 weeks in Group B (p=0.001). Percutaneous tendoachilles tenotomy was performed in 81% of the cases overall, but the Group B patients required far fewer tenotomies than the Group A patients (Group A, 98% vs Group B, 65%) (Figure 2).

Average ankle dorsiflexion post-tenotomy was 20 degrees (range, 0 to 35 degrees), with no significant differences between groups. The patients started to walk at an average age of 14 months (range, 12 to 20 months).

Four patients (4%) had a cast complication that included skin redness or slight swelling of the toes. These complications were attributed to a deficient casting technique. No infections, skin necrosis, neurovascular compromise or profuse bleeding post-tenotomy were observed. One patient had a slight over-correction that resolved over time and that did not affect when he started walking or the need for orthopedic shoes or inserts.

There were 20 relapses (22%) after initial successful treatment: 12 (27%) in Group A and 8 (17%) in Group B (p=0.42). The average age at relapse was 14 months (range, 4 to 27 months). Relapses were not significantly related to age at presentation or previous unsuccessful treatment at another institution. Relapses were, however, associated with non-compliance with the foot-abduction brace (p=0.001). Relapses were treated with a second series of manipulation and casting, followed by the use of the foot-abduction brace. Two patients required a second tendoachilles tenotomy, and another patient required tendoachilles lengthening and anterior tibialis transfer to the third cuneiform to prevent further relapses. At the last follow up, patients were evaluated for foot/calf shape and range of motion of the ankle. All patients had normal looking feet (although slightly smaller than the normal side) but smaller calf muscle mass. Plantar-flexion (average=36 degrees) and dorsi-flexion (average=22 degrees) were also slightly decreased compared to accepted normal values. No patient had pain or limitation of activities of daily living. All patients used regular shoes.

**DISCUSSION**

This report is the first from Chile demonstrating a very high correction rate (99%) for idiopathic clubfoot using the Ponseti method, and confirms the results of other recently published series. Importantly, we have found that the timing of cast removal is associated with
a dramatic decrease in the number of casts required for correction and also in the length of treatment. Both of these outcomes can have significant financial benefits for the families. Based on the very low complication rate in this series, we see no need to remove the cast the night before for skin care as traditionally recommended at many institutions. In addition, this study also demonstrates that following the published Ponseti technique and protocol to the smallest detail greatly improves the chance of achieving the outstanding results published by many other groups.

We have observed a high rate of relapses (22%) and, as many other groups have pointed out, most of these cases were due to non-compliance with the brace. This problem stresses the need for developing educational programs for the parents as well as other healthcare providers to maximize bracing compliance. It has been our experience that other physicians who are not very familiar with the method and protocol recommendations have allowed parents to discontinue the use of the brace much earlier than required. These cases almost always result in relapse.

We have also changed our clinic schedule to accommodate all clubfoot patients on a single day. This practice allows parents to share their concerns and questions with other parents, and to see that the results are predictable and lead to normal foot function. It also creates an environment of “peer pressure” where parents who are not compliant with the brace are exposed to other parents and children who are wearing the brace. It allows parents the opportunity to share techniques leading to easier brace wear. Furthermore, it has helped in the development of a network of parents in our country that are very supportive of the method.

Finally, it is also very important to work with local orthotic providers to understand the method and to help in the development of a brace suited for the financial conditions of the family as well as the country. Ultimately, it would be ideal if there were a standardized, comfortable and low-cost brace that would work for all families, both in developed and undeveloped countries.

In conclusion, removing the cast just before the new one is applied significantly decreased the number of casts required for correction and shortened the time of treatment. Following the principles and technical details of the Ponseti method will assure optimal results in almost all patients.

REFERENCES


ABSTRACT
This paper evaluates the efficacy of the Moss-Miami System instrumentation for surgical treatment of spinal deformity. Eight-five patients with AIS underwent a posterior spinal fusion with using this system between 1994 and 1998. Radiographs of the spine were taken preoperatively, at discharge, one year after surgery, and at the latest follow-up (average of 2.5 years, range 2 to 6 years). All radiographs were assessed for curve magnitude, coronal balance, kyphosis, lordosis, junctional kyphosis, and sagittal balance. The Scoliosis Research Society instrument was administered at the final follow-up. The average curve correction ranged from 53 to 65 percent. All patients showed solid fusion by final follow-up. Three patients required a second operation for complications related to their scoliosis; two patients showed a rod fracture without evidence of pseudoarthrosis or curve progression. Two transient neurological complications related to the surgery were observed. SRS results were favorable with regard to function and cosmetic appearance.

INTRODUCTION
Hibbs is credited with first using spinal fusion as a treatment for scoliosis in 1914.21 The introduction of instrumentation for spinal fusion started with Harrington in 1960,23 and consequent to this innovation, surgical treatment has evolved from the one-planar, two-dimensional instrumentation of Harrington to the three-planar, three-dimensional instrumentation of Cotrel-Dubousset in 1984.12,14,15 Cotrel-Dubousset instrumentation was designed to provide segmental fixation of the spine and selective distraction and compression at different levels to improve the alignment of the spine in the coronal, sagittal, and axial planes. The stability achieved by this system precludes the need for post-operative immobilization in the majority of cases.1,3,5,32,45,47 Nevertheless, despite reports of satisfactory results with regard to fusion, percentage correction, and minimal complications, problems have been reported with posterior systems, including loss of correction in the lumbar spine, hypokyphosis of the thoracic spine and the presence of a thoracolumbar kyphosis below the level of instrumentation.5,9,30,42,49

The Moss-Miami System instrumentation (DePuy, Warsaw, IN) was introduced in 1994, and shortly thereafter we began using the system for all patients with adolescent idiopathic scoliosis. Advantages of this system include a lower profile, smaller dimensions, top-opening hooks simplifying rod placement, and the ability to use various hooks or screws in the lumbar spine.

The purpose of this study is to describe short-term radiographic, clinical and patient-oriented outcomes after fusion and instrumentation with the Moss-Miami System for the treatment of adolescent idiopathic scoliosis.

MATERIALS AND METHODS

Patient Selection
The indications for surgical treatment of adolescent idiopathic scoliosis included a progressive curve over 40 degrees in skeletally immature patients or a progressive curve measuring greater than 50 degrees or a painful curve in skeletally mature patients. All surgeries were performed using the senior author using Moss-Miami instrumentation. Those who underwent surgery at or before the age of 18, with a minimum of two-year follow-up were included.

Radiographic Analysis
Standing posteroanterior, lateral, and anteroposterior with lateral bending radiographs were taken of all patients prior to surgery; oblique films were taken between six to eight months post-operatively, and posteroanterior films were taken yearly. We do not routinely obtain lateral films on post-operative patients, and these were available for twenty-three of the patients at the final follow-up.
The following radiographic measurements were taken from the posterior-anterior films: Risser grade, Cobb angle, degree of apical rotation of the primary curve, global coronal balance (lateral displacement, in centimeters, of a plumb line drawn from the base of the spinous process of the seventh cervical vertebra to the center of the sacrum) and lowest instrumented vertebra translation (geometric center of the vertebra to the center sacral line measured in millimeters).

The lateral films were used to assess the pre- and post-operative sagittal plane alignment using the method of Cobb. The thoracic kyphosis was measured from the upper end plate of T₃ to the lower end plate of T₉. The thoracolumbar junction was measured from the top of T₁₁ to the bottom of L₁ and lumbar lordosis from the lower end plate of T₁₂ to the upper end plate of S₁.

Global sagittal balance was assessed by drawing a vertical plumb line from the center of the body of C₇ to the level of the sacrum. The distance from this line to the anterior edge of S₁ was measured in centimeters; positive values were assigned to anterior displacement and negative value to lines dropped through or behind the sacrum. All radiographic measurements were made by the same team of surgeons.

Clinical Review
The medical records, including clinical notes and operative reports, were examined for the following: surgical indication, time from diagnosis to surgery, blood loss, blood and blood product replacement, surgical complications, length of stay and spine surgery consequent to the index fusion.

Patient Self-Report
All the patients were asked to complete the original version of the twenty-four item Scoliosis Research Society outcomes instrument. Responses to each item can range from 1 (lower functioning) to 5 (highest functioning). Using the scoring method presented in two previous studies, we calculated average scores within the seven subscales (pain, general self-image, self-image related to surgery, function after surgery, general function, function-activity, and satisfaction with surgery) and for the complete instrument.

Statistical Analysis
Descriptive statistics were calculated for the clinical and radiographic variables over time. Subscale and total scale internal consistency was estimated using Cronbach’s alpha, and Pearson correlations were calculated to estimate relationships between the subscales and also between the subscales and the radiographic variables percent correction, absolute correction, and number of levels fused. Differences in the subscale means between curve types and between lowest level fused were evaluated using analysis of variance. Models predicting satisfaction and pain scores were evaluated using multivariable linear regression.

RESULTS
Sample
Between 1994 and 1998, 133 patients with adolescent idiopathic scoliosis underwent a posterior arthrodesis of the spine using Moss-Miami System instrumentation. Of these, eighty-five patients (seventy females and fifteen males) had a minimum of two-year follow-up. The average age at surgery was 15.64 years (median=14.00; range, 11 to 40). The Risser sign was zero in nineteen patients, one in six patients, two in nine patients, three in fifteen patients, four in twenty patients, and five in sixteen patients. Forty-six patients had single structural curve patterns including thirty-six thoracic curves, three thoracolumbar curves and seven lumbar curves. Thirty-nine patients had two structural curves: five double thoracic curves, two thoracic/thoracolumbar curves, and thirty-two double major curves. Ten patients underwent magnetic resonance imaging prior to surgery due to atypical features: three patients presented before the age of ten, three patients had rapid progression of their curves, one adolescent patient complained of significant, function-altering back pain, one patient presented with an abnormally large curve, one patient had an associated spondylolisthesis, and one patient presented with a left thoracic curve. None of these patients required neurosurgery prior to instrumentation and fusion.

Twenty-nine patients underwent surgery after treatment with a thoracolumbosacral orthosis failed to halt progression of their curves, with an average time between diagnosis and surgery of three years and six months. Fifty-six patients presented at our institution with curves meeting surgical indications and underwent surgery at an average of ten months after presentation. The latest follow-up was performed at an average of two years and seven months after surgery (range two to six years) when the patients were an average of 18.33 years of age (median=16.41; range 13 to 42).

Surgical Technique
Eighty-one patients were treated with the standard construct consisting of two longitudinal rods with two transverse, two-centimeter connector rods. A third shorter rod was used in three patients (one adolescent and two adults) due to unusually stiff curves, and connected to one of the main rods by dominos. Four patients had an anterior release combined with the posterior
instrumentation to achieve maximal surgical correction in light of extremely stiff curves. Nine patients, in whom the thoracic prominence measured greater than four centimeters, had a thoracoplasty in addition to the primary procedure.

Hypotensive anesthesia was used in all cases. The estimated average blood loss at surgery was 604 cc (range, 100 to 1500 cc). Seventy-three patients had intra- or postoperative transfusions, requiring an average of 2.5 units of red blood cells or fresh frozen plasma (autologous units were used in fifty-nine cases). Autografts were taken from the iliac crest, except when a thoracoplasty was also performed; rib grafts were used in those cases. The average number of vertebrae fused was 11.28 (range, 7 to 15 levels). Intraoperative motor and sensory spinal cord monitoring and wake-up tests were conducted in all cases; all were normal. The average length of stay was 5.9 days (range, four to thirteen days).

Radiographic Analysis

Pre- and post-operative radiographic measurements from the anteroposterior views are presented in Table 1. The average correction in the Cobb angle from before surgery to the film taken prior to hospital discharge was 61 percent for all curves, ranging from 21 to 89 percent. The median rotation of the spine was Grade 2 (range 1 to 4) both before and after the procedure using the Nash and Moe classification. A solid arthrodesis was achieved in all patients within eight months as demonstrated on oblique radiograph.

Loss of correction (defined as progression greater than 9 degrees) from surgery to the first year follow-up was noted in twenty-seven patients (32 percent). Loss of correction occurred in 22 percent of thoracic curves (10/46); 47 percent of the patients with double major curves (15/32); in one patient with a double thoracic curve, and in one with a double thoraco-thoracolumbar

![Table 1: Average Radiographic Measurements in the Frontal Plane by Time and Curve Pattern](image-url)

*LIV*: frontal tilting of the last instrumented vertebra

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curve. Of the patients with double major curves, both the thoracic and lumbar components progressed in seven patients; in the remaining four patients, only one of the components progressed. Four patients (one lumbar and three double major curves) had a loss of correction greater than 9 degrees between the first year after surgery and the final follow-up. Of the double major curves, both components progressed in one patient, and in the other two only one component progressed. In each case of progression in the lumbar region the curve had been fused to L₁ or L₂. Loss of correction was not related to Risser grade (Risser 1, 26 percent; Risser 2, 17 percent; Risser 3, 22 percent; Risser 4, 50 percent; Risser 5, 19 percent).

Coronal balance improved on average from 1.80 centimeters before surgery (range, 0-8.0) to 0.89 centimeters at the final follow-up (range 0-3.5). The average change from pre-op to final follow-up was 0.75 centimeters (range, -2.6 to 7.3); 78 percent of the patients demonstrated either maintenance or an improvement in the coronal balance. Deviation of the last instrumented vertebra ranged from 0 to 4.7 centimeters before surgery (mean, 1.39) and was virtually unchanged at final follow-up (mean, 1.4; range 0 to 5.0).

Radiographic measurements in the sagittal plane are summarized in Table 2. The average thoracic kyphosis at the time of the procedure was 28.22 degrees (range, 10 to 60 degrees). Using the range of normal defined by Voutsinas and MacEwen,21 at one year follow-up 16 percent of patients had a normal degree of kyphosis (30.5 to 46.6 degrees), while 84 percent remained hypokyphotic. Hypokyphosis was less frequent in patients with thoracolumbar (one of three patients) and lumbar curves (three of seven curves) than in patients with thoracic involvement and higher fusion levels. Lordosis measured an average of 59.76 degrees (range, 34 to 86) before surgery and 57.96 degrees one-year after surgery (range 30 to 81 degrees). Sixty-three percent of patients were normo-lordotic (normal range, 48-66 degrees) and 16 percent were hypo-lordotic at one-year follow-up.

Junctional kyphosis ranged from -13 to 60 degrees pre-operatively (mean, 3.42) and from -10 to 20 degrees (mean, 3.05) at one-year follow-up. All three patients with thoracolumbar curves had a junctional kyphosis measuring greater than 5 degrees, compared to 0 to 25 percent of patients with other curve patterns. The junctional kyphosis was greater than 5 degrees in four of five patients whose fusion stopped in the thoracic spine, compared to 19 percent (15/80) of those whose fusions extended beyond L₁. Pre-operative measurements of sagittal balance ranged from -11.50 centimeters to 1.1 centimeters for an average of -4.30. At one-year after surgery, the range was -10.40 to 4.4 and averaged -3.30.

### Complications

Two patients (aged thirty-two and thirty-six years) suffered a rod fracture after the first year. In both cases the broken rod was to the left of the spine, on the concave side of the curve, with the fracture occurring at the level of the thoracolumbar junction, directly adjacent to a hook. However, no pseudoarthroses or loss of curve correction was noted at final follow-up in these patients.

Three patients required a second operation for a complication related to scoliosis. One patient required a proximal extension of the fusion for a junctional kyphosis (70 degrees) two years after the initial surgery. A second patient had a progression of a lumbar curve distal to the area of previous fusion with development of a junctional kyphosis and increasing rib prominence (approximately 4 cm) (Figures 1, 2). This required, thirty-three months after the first surgery, an extension of the existing posterior spinal fusion (from T-8 to L-4 on the right and from T-12 to L-4 on the left) and thoracoplasty (Figures 3, 4). The third patient had an excision of the tip of a rod due to bursitis at thirty-two months after surgery.

Four patients had a second surgery after posterior spine fusion for problems not directly related to the index surgery: three patients underwent thoracoplasty (one for pain and cosmesis at twenty-nine months post-operatively, one for rib regrowth post-thoracotomy after twenty-two months, and another for cosmesis at thirty months post-operatively). Two patients had an associated asymptomatic spondylolisthesis that was not treated at the time of surgery and of whom one required posterior-lateral fusion from L₃ to S₁ thirty-seven months after the scoliosis procedure.

Two patients had neurological complications. One patient had an eighth cervical rib that was recognized pre-operatively, requiring careful positioning during the surgery. Despite precautions, this patient developed a partial eighth nerve palsy. A second patient developed a partial paraplegia likely related to spinal cord ischemia during the operation. This patient had normal motor and

### Table 2

**Average Radiographic Measurements in the Coronal Plane by Time**

<table>
<thead>
<tr>
<th></th>
<th>Pre-op</th>
<th>Post-op</th>
<th>1 yr FU</th>
</tr>
</thead>
<tbody>
<tr>
<td>Kyphosis (°)</td>
<td>28.22</td>
<td>22.81</td>
<td>25.16</td>
</tr>
<tr>
<td>Fusion to T12</td>
<td>(10-60)</td>
<td>(5-57)</td>
<td>(10-68)</td>
</tr>
<tr>
<td>Fusion past L1</td>
<td>59.76</td>
<td>55.01</td>
<td>57.96</td>
</tr>
<tr>
<td>(0-4)</td>
<td>(28-79)</td>
<td>(30-81)</td>
<td></td>
</tr>
<tr>
<td>Junctional Kyphosis</td>
<td>3.42</td>
<td>2.91</td>
<td>3.05</td>
</tr>
<tr>
<td>(13-60)</td>
<td>(10-18)</td>
<td>(10-20)</td>
<td></td>
</tr>
<tr>
<td>Fusion to T12 (n=5)</td>
<td>2.00</td>
<td>7.60</td>
<td>11.40</td>
</tr>
<tr>
<td>Fusion past L1 (n=80)</td>
<td>3.51</td>
<td>2.61</td>
<td>2.53</td>
</tr>
<tr>
<td></td>
<td>(13-60)</td>
<td>(10-18)</td>
<td>(10-18)</td>
</tr>
</tbody>
</table>
sensory spinal cord monitoring and a normal wake-up test. Both complications resolved within six months.

Self-Report Data
Fifty-seven patients completed the SRS questionnaire (67 percent). All but one patient currently performs at least light labor and 66 percent reported full activities without restriction. Twenty-three percent of the patients reported an increase in function after surgery and 11 percent an increase in ability to enjoy sports or hobbies. Ninety-one percent felt they look “good” or “very good” in clothes, 14 percent perceived an increased confidence in relationships with others, 46 feel they look better since the surgery, and 18 percent said treatment improved the way others viewed them. Seventy-seven percent of the patients were “extremely satisfied” with the results, and 95 percent said they’d definitely have the treatment again.

The average score over all twenty-four items was $4.10 \pm 0.41$ (range, 2.56 to 5.00). The average percentage total score was $82 \pm 8$ (range 54 to 94). Scores were calculated for seven subscales (general and post-operative self-image, general and post-operative function, activity, pain and satisfaction with surgical results). The subscales did not exhibit high levels of internal consistency, either due to the small number of items within a scale, or due

Figure 1A and 1B. Radiographs of a 14-year-old female with a 56 degree right thoracic curve which corrected to 34 degrees with side-bending, and a left lumbar curve of 45 degrees which corrected to 14 degrees on side-bending.
to the lack of correlation between the items constituting each scale. Cronbach’s alpha coefficients of reliability ranged from 0.12 (general function scale, 3 items) to 0.68 (post-operative self-image, n=3). Subscale scores are presented in Table 3. These scores were not related to the age of the patient, post-operative Cobb angle, the percentage of curve correction obtained, the number of levels fused, the distal extent of the fusion or need for re-operation (all p-values <0.05). Differences between curve types was found for one of the seven scales; the patients with thoracolumbar curves had a significantly higher (p<0.02) mean score on the post-operative self-image scale (4.83) than did patients with double major (3.52), thoracic (3.21), lumbar (3.06) or double thoracic curves (3.00). There was no significant difference in pain scores between the forty-six patients with no junctional kyphosis (-5.00 to 5.00 millimeters) or those with a junctional kyphosis (greater than +/-5 millimeters). Using multivariable regression, pain was modeled as a function of number of levels fused, general self-image, and the three function subscales; significance tests showed that patients with lower levels of activity (p<0.0001), and lower self-image scores (p<0.004) also had higher pain scale scores.

Satisfaction with treatment was modeled as a function of pain, both self-image scales, post-operative function and activity. Of these variables, only post-operative self-image (p<0.03) and pain (p<0.01) were significantly predictive of satisfaction. Patients with an increase in pain or a decrease in self-image tended not to be satisfied with the results of treatment.
TABLE 3. SRS Outcomes Instrument Scale Scores: Reliability and Average Scores by Subscale

<table>
<thead>
<tr>
<th>Scale Score</th>
<th>Alpha Coefficient</th>
<th>Average Item Score</th>
<th>Average of Summed Items</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pain</td>
<td>0.66</td>
<td>4.35±0.52 (2.57, 5.00)</td>
<td>29.30± 5.27 (8.00, 35.00)</td>
</tr>
<tr>
<td>General Self-Image</td>
<td>0.67</td>
<td>4.13±0.61 (2.33, 5.00)</td>
<td>12.18± 2.09 (6.00, 15.00)</td>
</tr>
<tr>
<td>Post-operative Self-Image</td>
<td>0.68</td>
<td>3.36±0.72 (1.33, 5.00)</td>
<td>10.00± 2.07 (4.00, 15.00)</td>
</tr>
<tr>
<td>General Function</td>
<td>0.12</td>
<td>3.11±0.79 (1.00, 5.00)</td>
<td>6.16± 1.65 (2.00, 10.00)</td>
</tr>
<tr>
<td>Post-operative Function</td>
<td>0.31</td>
<td>4.20±0.55 (2.33, 5.00)</td>
<td>12.35± 1.75 (7.00, 15.00)</td>
</tr>
<tr>
<td>Function/Activity</td>
<td>0.57</td>
<td>4.48±0.86 (2.00, 5.00)</td>
<td>13.23± 2.82 (6.00, 15.00)</td>
</tr>
<tr>
<td>Satisfaction</td>
<td>0.53</td>
<td>4.49±0.46 (3.00, 5.00)</td>
<td>13.47± 1.39 (9.00, 15.00)</td>
</tr>
<tr>
<td>Total</td>
<td>0.84</td>
<td>4.10±0.41 (2.71, 4.71)</td>
<td>96.68± 11.72 (63.00, 113.00)</td>
</tr>
</tbody>
</table>

Figure 3A and 3B. Three years later, she presented with a junctional kyphosis below her fusion and adding on to her original curve. At surgical exploration, her fusion was found to be solid. She underwent extension of her fusion distally without complication.
In 1994, after many years’ experience with the original Cotrel-Dubousset system, our service began using the Moss-Miami instrumentation system for posterior spine fusion in adolescent idiopathic scoliosis. This system presents biomechanical and metallurgical attributes similar to other currently used posterior systems. It also offers advanced characteristics such as low profile implants, smaller rod diameter (five millimeters), all top opening implants permitting straight-forward rod placement, a simplified and strong closure mechanism (which can be easily removed or altered if necessary), different hook styles, and pedicle screws.

Since the introduction of three-planar, three-dimensional systems, beginning with the Cotrel-Dubousset system in 1984,12-15 several studies have been published concerning their properties including the amount and stability of coronal plane correction, normalization and maintenance of thoracic kyphosis and lumbar lordosis, derotation, and intra- and post-operative complications.8,9,30,32,42,45,47,48 However, we are not aware of similar studies concerning the Moss-Miami system.
Average correction of the Cobb angle in the coronal plane in this case series was greater than 50 percent for all curve patterns similar to the published range of 54 percent to 68 percent using Cotrel-Dubousset instrumentation or that reported by Cardoso-Monterrubio and Oliveira-Castillo using the Moss-Miami system. In a comparative study, Luk et al. found no significant difference between the amount of correction possible using Texas Scottish Rite, Isola, CD-Horizon or the Moss-Miami systems. Also similar to these studies was the slight loss of correction noted in the first year after surgery. No further loss of correction was observed at the latest follow-up. Only in the seven single lumbar curves did loss of correction occur after the first year, and this was very slight at an average of four degrees. This loss could be attributed either to measurement error or the limited segmental fusion of the lumbar spine in an attempt to preserve mobility.

Unlike the Cobb angle, we were not able to correct rotation, and the median Nash Moe grade was 2 both before and after surgery. This lack of rotational correction was also reported by Lenke et al. and Takahashi et al. Therefore, the cosmetic improvement noted in all patients treated is wholly attributable to the improvement in apical translation.

Risser, Moe, and Tambornino advise that fusion should include the entire major curve and end at the neutral stable vertebra. Harrington, however, advocates extending the fusion one or two vertebrae below the curve. King et al. identified the stable vertebra as the appropriate area to end fusion by using side-bending radiographs to differentiate structural from compensatory curves. Using this latter criteria for choosing fusion levels, at latest follow-up the average coronal balance was 0.8 centimeters, and the translation of the lowest instrumented vertebra was 1.3 centimeters. The average spinal balance observed at the last follow-up was –2.2 centimeters. However, the measurement of sagittal balance was noted to be extremely sensitive to the position of the patient during the radiograph and within-patient variation was frequent and sometimes large. This lack of reliability limits the use of this measure as a meaningful predictor or outcome.

At final follow-up the junctional kyphosis averaged 3.05 degrees compared to 3.42 degrees preoperatively. The fusion ended at or above T12 in five of our patients and one of them developed a junctional kyphosis of 20 degrees. This patient required extension of the distal fusion with lengthening of the instrumentation two years and nine months after the index procedure (Fig. 1). Patients whose fusion extended into the lumbar spine had a significantly smaller average junctional kyphosis that remained essentially unchanged after surgery. Some authors have expressed concern about the risk for postoperative junctional kyphosis if a fusion is stopped at the thoracolumbar junction, although this concern is not universal. Long-term follow-up is necessary before we fully understand the implications of junctional kyphosis in terms of radiographic and clinical outcomes.

The ability to correct or maintain physiologic sagittal contours of the spine is the major advantage of modern instrumentation systems. Several studies have discussed hypokyphosis of the thoracic spine in patients who have had spinal fusion with posterior instrumentation, but as noted by Lee, few have discussed the occurrence of proximal hyper-kyphosis. One of our patients, fused from T8 to L3 for a lumbar curve, developed hyper-kyphosis above the level of fusion. Kyphosis increased from 37 degrees after the first surgery to 70 degrees two years after, requiring a second surgery to lengthen the instrumentation and extend the proximal fusion to T6. Radiographic follow-up fourteen months after the second surgery demonstrated a right curve from T4 to T9 of 8 degrees, a left curve from T10 to L3 of 30 degrees, and 30 degrees of kyphosis.

Complications in this series were infrequent. The two rod fractures noted at follow-up occurred under similar circumstances. Both were in adult patients, on the concave side of the curve, at the level of the thoracolumbar junction, adjacent to a locking device. These circumstances suggest that perhaps repeated tightening or loosening of the device, stress risers from contouring the rod, or the relatively large size of the patient contributed to the rod fracture. And although there were no adverse consequences for either of these patients, it might be reasonable to use 0.25-inch rods in adult patients instead of the 5 millimeter rods typical of this system.

Two transient neurological complications occurred. One patient showed a partial paraplegia after surgery, likely related to spinal cord ischemia during the operation. The intraoperative spinal cord monitoring and the wake-up test were both negative. He had weakness in all muscle groups in both lower extremities and difficulty with bowel and bladder incontinence for six weeks, while his hyperreflexia and a mild clonus disappeared in six months after surgery. The second patient showed a partial eighth nerve palsy. The patient had an eighth cervical rib, and the position of the patient on the surgical table can be related to this symptomatology. Also this complication was completely resolved in six months.

Richards and Grosman described cases of late infection requiring consequent removal of the instrumentation. The Grosman study evaluated infectious complications in 786 patients with idiopathic scoliosis over 24 years. As in Cardoso’s work, no infection was noted after use of Moss-Miami instrumentation.
The infection rate with the Harrington system was 1.1 percent, 5 percent with TSRH and 4.8 percent with Isola. None of the our patients developed an infection, but one patient did require removal of two centimeters of the distal end of the rod due to the development of a painful aseptic bursitis two years and eight months after the spine fusion.

Aurori et al. reported similar percentages of pseudoarthrosis in two groups of patients who underwent spinal fusion with Harrington instrumentation and who received autogenic or allogenic bone grafts. We used only autogenic bone graft and observed no pseudoarthrosis at follow-up. Moreover, we also did not observe the flat-back syndrome consequent to sagittal imbalance described by other authors.

It is difficult to synthesize or compare findings from the SRS Outcomes instrument across published studies. The original version used here was also used by Haher et al., White et al., and Perez-Grueso et al. However, populations differed in terms of length of follow-up, types of instrumentation used, results reported, and method of scoring. For example, Haher et al. added item scores together to create subscale scores, while White et al. reported the average score across items within a subscale. This makes it difficult to compare means or correlations across studies. Additionally, unlike previous studies, the subscales scores from this sample did not demonstrate adequate levels of internal consistency. This could be due differences in the samples, or be just an inherent property of scales consisting of such few items. Although the alpha coefficient for the entire scale was adequate at 0.85, caution should be taken when interpreting results of the subscales themselves.

The results of the patient self-report in this study in general supports previous work, in that radiographic characteristics of the curve or the correction were not significantly related to the overall reported outcome, or to any of the subscales. White et al. did report a relationship between pain and number of vertebrae fused in their follow-up study. This difference in findings could be due, in part, to the longer length of follow-up (seven years) compared to the average two year follow-up in this paper. It should be noted, however, that the correlation between the number of fused vertebrae and pain was 0.15 in the previous, and although statistically significant, represents a very small amount of common variance. The lack of correlation between radiographic changes and SRS scores was the main point of a recent study by d’Andrea et al., who conclude that separate analyses of radiographic and patient-reported outcomes are required to obtain a comprehensive assessment surgical results. This point is supported by findings concerning overall satisfaction with the results of surgery. Both Haher et al. and the current study found that satisfaction is not explained by the size of the curve post-operatively or the amount of correction, or even the need for reoperation, but by pain and self-image. Therefore, as pointed out by D’Andrea et al., researchers need to be wary about substituting patient satisfaction for technical outcomes or vice versa, especially from short-term follow-up studies.

Since introduction of the Harrington instrumentation, there have been several research reports on the surgical treatment of idiopathic scoliosis using methods including the Cotrel-Dubousset, the CD-Horizon, the TSRH or the Isola methods. However, the research on the Moss-Miami System has been scant. In our study, post-surgery follow-up ranged from 2 to 6 years. In light of the recent introduction of this instrumentation (it was introduced only nine years ago), the follow-up period reported in this study can be considered satisfactory. In the present study several outcomes were examined including the Cobb angle correction, coronal and sagittal balance, surgical and medical complications, and personal satisfaction with the surgery. Future studies will establish the long-term outcome of the Moss-Miami system for the surgical treatment of adolescent idiopathic scoliosis.

ACKNOWLEDGMENT
This work was supported by the Sovena Foundation.

REFERENCES


LONG-TERM CLINICAL AND RADIOGRAPHIC RESULTS OF COTREL-DUBOUSSET INSTRUMENTATION OF RIGHT THORACIC ADOLESCENT IDIOPATHIC SCOLIOSIS

Norbert Boos, MD\textsuperscript{2}, Lori A. Dolan, Ph.D, Stuart L. Weinstein, MD\textsuperscript{1}

ABSTRACT

Little substantive data is available in the literature on the long-term clinical and radiological results of Cotrel-Dubousset instrumentation (CDI) for the treatment of adolescent idiopathic scoliosis. We therefore retrospectively investigated the long-term clinical and radiographic outcome of patients who underwent (CDI) for right thoracic adolescent idiopathic scoliosis. 54 consecutive patients (45 females, 9 males) who underwent CDI for right thoracic adolescent idiopathic scoliosis with an average age of 14 years (range 10 - 21 years) at surgery were included in this series. There were 18 King Type II, 19 Type III, 5 Type IV, 3 Type V and 9 double major curves. The average coronal Cobb angle of the primary thoracic curve preoperatively, postoperatively and at latest follow-up was 55°, 17° and 22°, respectively. The lumbar curve (secondary and double major) averaged 40°, 21° and 23°, respectively. Coronal balance (deviation from the central sacral line) was slightly improved from 13 mm to 11 mm. The average shoulder elevation increased from 3° to 5°, presumably as a result of the rod derotation maneuver. Thoracic kyphosis (20° to 22°) and lumbar lordosis (49° to 54°) was preserved or even improved by the instrumentation. All patients were doing well and had no complaints with regard to a substantial limitation of professional or sports activity. There were no apparent non-unions, infections or neurological complications. CDI of adolescent right thoracic idiopathic scoliosis provides encouraging clinical and radiographic results at an average follow-up of 9 years (2 to 16 years). Overall patient satisfaction, functional status and subjective cosmetic improvement is high.

INTRODUCTION

For many years Harrington instrumentation of adolescent idiopathic scoliosis was the gold standard for treatment of this deformity.\textsuperscript{15,18} Although patient satisfaction with Harrington instrumentation was good,\textsuperscript{8,12,15} the drawbacks of this intervention have become apparently clear. While adequate correction in the coronal plane and solid fusion to prevent curve progression was possible, the sagittal contour often deteriorated by the application of this pure distraction implant. Long-term problems with the malalignment in the sagittal contour and imbalance have been reported, particularly if the fusion extended into the lumbar spine.\textsuperscript{9,19}

Since its introduction in 1984, Cotrel-Dubousset instrumentation (CDI) (11) has substantially advanced spinal surgery in terms of modern segmental spinal fixation. Theoretically, this instrumentation should provide a better coronal deformity correction than Harrington instrumentation while at the same time also improving the sagittal contour. However, the axial correction (i.e., derotation potential) has not been as effective as initially hoped. Most studies today have focused on the radiological and technical aspects of CDI.\textsuperscript{3,14,17,20,29,30,32,36,41}

Despite its theoretical advantages, little substantive data is available in the literature with regard to the intermediate and long-term clinical outcome. The objective of this study was, therefore, to investigate the clinical and radiographic outcome of patients who underwent CDI for adolescent idiopathic scoliosis with an average follow-up of ten years.

MATERIAL AND METHODS

This is a retrospective analysis of 54 consecutive patients with adolescent idiopathic scoliosis operated on between 1986 and 1991. The initial inclusion criteria were a right thoracic curve, age at surgery of less than 21 years, Cotrel-Dubousset instrumentation, and a potential minimum follow-up of 10 years. Exclusion criteria
were curves other than right thoracic curve pattern, congenital or neuromuscular curves, or any neurologic or mental abnormality. There were 45 females and 9 males with an average age at operation of 14 years (range 10 to 21 years) who fulfilled these criteria. Menarche had occurred in 24 of 45 girls at the time of surgery. Twelve of the 54 patients preoperatively complained of occasional back pain and 7 patients reported frequent back pain. None of the patients had a functional limitation or a neurological deficit before surgery.

Preoperative and intraoperative data was collected by review of hospital records and radiographs. Preoperative and intraoperative data were available for all but one case where the chart has been lost. The preoperative radiologic work-up consisted of standing posterior-anterior, lateral and side-bending radiographs. The average curve size was 55 degrees (range 40 to 78 degrees). There were 18 King Type II (33%), 19 Type III (35%), 5 Type IV (9%), 3 Type V (5%) and 9 double major curves (17%). The median Risser grade was 2.

The mean interval between discovery of the scoliosis and surgery was 33 months (1-108 months). Twenty-seven patients (50%) had a brace (17 Milwaukee, 10 Boston brace), 4 patients (7%) were treated by electrical stimulation (Scolitron®) and 23 (43%) did not receive any specific treatment. The indications for surgery were curve progression in 32 patients (59%) and the magnitude of curve at initial presentation relative to the Risser grade in 22 patients (61%).

SURGICAL TECHNIQUE

In this study, the CDI was applied exclusively with hooks (i.e., no pedicular fixation). The hook pattern for the different curve types corresponded to the available literature guidelines at that time.10 The senior author (SLW) performed all operations in a single stage through a posterior approach. In 9 patients the surgery was performed using spinal cord monitoring which was not yet routinely available at that time. The average operation time was 212 minutes (130 to 295 min). The estimated average blood loss was 876 cc (200 to 2100 cc). Surgery was performed with a cell saver when available. 20 patients operated on without cell saver required homologous blood transfusion (mean 2.8 units, range 1 – 6 units). The standard post-operative treatment consisted of early mobilization on the first or second postoperative day without the use of a brace. The hospitalization averaged 7 days (5 to 15 days).

QUESTIONNAIRE DATA

A vigorous attempt was made to bring each patient back for physical examination and radiographs. To allow for assessment of the clinical outcome independent of the operating surgeon a self-administered questionnaire used in a previous study38,39 was mailed to all study participants. The questionnaire covered the domains back pain (4 items), function (3 items), self-image (3 items) and patient satisfaction (3 items). When return appointments were not possible, attempts were made to reach each patient by mail in order to obtain follow-up radiographs and responses to the questionnaire. However, the vast majority of patients could not be located or brought back for a 10-year follow-up. Rather than excluding patients because of limited follow-up, we decided to include all available follow-up data of this study population for review including chart notes, questionnaires, and radiographs.

The radiographic analysis consisted of a thorough review of all serial radiographs and was performed by one of us (N.B.) independent of the operating surgeon (SLW). The spine was considered coronally balanced if the deviation of the plumbline from C7 was equal or less than 2 cm off the midsacral line. A coronal imbalance was defined as a deviation more than 2 cm. Loss of coronal curve correction was considered to have occurred if the difference in Cobb angle between the immediate postoperative and follow-up radiographs was more than 10 degrees.

RESULTS

The overall follow-up rate was 96% (52/54 patients) at an average of 79 months (25 to 185 months) after surgery. Two patients were lost to follow-up 2 and 7 months postoperative, respectively. In 49 of the remaining 52 cases a minimum follow-up of 5 years was available, 14 of whom had a follow-up of more than 10 years.

Clinical Results

Two to 15 years after surgery, none of the patients had a neurologic deficit on clinical examination. All patients were doing well and had no complaints with regard to a substantial limitation of daily or recreational activity. At the latest follow-up visit, occasional moderate back pain was reported in 10 patients. None of those patients needed a specific treatment. Three patients complained of prominent hardware.

Questionnaire Results

Thirty-eight of the 54 patients completed a self-administered questionnaire.

Experience of pain. Before surgery, 19 patients (50%) reported that they never had back pain during the past 3 month. Eight patients (21%) reported pain at the iliac crest where the bone graft was taken. None reported leg pain. One patient reported severe but non-specific back pain on a daily basis. Pain medication was never
needed by 56%, rarely by 23%, occasionally by 15% and always by 5% of the patients.

Function. There was no significant difference in the level of daily or recreational activities before and after surgery (paired t-test, p=0.46). None of the individuals reported a substantial limitation in daily or recreational activities but 5 patients (13%) felt that their activity level was worse than before surgery. 21% of the respondents (n=8) stated that their choice of career was not influenced by their condition, while 25 felt the deformity was somewhat of an influential factor. Five patients reported a substantially negative impact on their career choice from their condition.

Self-image. 16 patients (42%) reported that their deformity was not apparent to others while being dressed before surgery, compared to 87% after surgery. Twenty-five patients (64%) believed their appearance had improved due to surgery, while the rest felt the surgery had not made a significant difference. Twenty-four patients (63%) reported that surgery had improved the way they felt about themselves.

Patient satisfaction. 76% of the patients were extremely satisfied with the operation and only one patient was somewhat dissatisfied. The expectations of surgery were exceeded in 23 patients (61%) and only one individual reported that the outcome was less than expected. Out of 39 patients, 33 would definitely and 5 would probably choose the operation again under similar conditions, and one patient was undecided.

Radiographic Results

Five patients had a radiological follow-up of less than 2 years and were not included the radiological analyses. None of those individuals had an implant-related complication or a short-term radiological outcome substantially different than the remaining study sample. The average radiological follow-up of the remaining 49 patients was 94 months (20 to 185 months). Thirty-nine patients had a minimum follow-up of 5 years, 14 of whom had a minimum follow-up of ten years.

We did not detect any non-unions during examination of the postoperative radiographs. The average

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### TABLE 1
Radiographic Results

<table>
<thead>
<tr>
<th>Coronal Measurements</th>
<th>Preoperative</th>
<th>Postoperative</th>
<th>Latest Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cobb-Angle Thoracic Curve (degrees)</td>
<td>55.3 (40-78)</td>
<td>17.3 (0-44)</td>
<td>22.3 (2-55)</td>
</tr>
<tr>
<td>Lumbar Curve (Type II, double major) instrumented</td>
<td>53.0 (33-72)</td>
<td>19.1 (4-30)</td>
<td>26.2 (11-47)</td>
</tr>
<tr>
<td>Lumbar Curve (not instrumented)</td>
<td>40.0 (16-58)</td>
<td>21.5 (0-58)</td>
<td>21.7 (0-48)</td>
</tr>
<tr>
<td>Apical Vertebral Deviation (mm)</td>
<td>42.8 (6-113)</td>
<td>13.1 (0-40)</td>
<td>15.4 (0-75)</td>
</tr>
<tr>
<td>Apical Rotation</td>
<td>2.1 (1-4)</td>
<td>1.7 (0-3)</td>
<td>1.7 (0-3)</td>
</tr>
<tr>
<td>Vertebral tilt of most caudal level with instrumentation (degrees)</td>
<td>21.7 (0-45)</td>
<td>6.8 (0-25)</td>
<td>9.4 (0-32)</td>
</tr>
<tr>
<td>Translation of most caudal level with instrumentation (mm)</td>
<td>13.2 (0-38)</td>
<td>14.6 (0-36)</td>
<td>11.5 (0-32)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Sagittal Measurements</th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Coronal balance (mm)</td>
<td>13.2 (0-45)</td>
<td>15.8 (0-44)</td>
<td>11.3 (0-47)</td>
</tr>
<tr>
<td>Tilt of Free IVD</td>
<td>5.1 (0-15)</td>
<td>3.4 (0-15)</td>
<td>3.5 (0-28)</td>
</tr>
<tr>
<td>Thoracic Kyphosis (T5-T12)</td>
<td>19.7 (0-61)</td>
<td>17.9 (2-45)</td>
<td>22.2 (7-66)</td>
</tr>
<tr>
<td>Thoracolumbar Kyphosis (Th11-L2)</td>
<td>-3.6 (-25–23)</td>
<td>-3.1 (-32-9)</td>
<td>-1.5 (-15–12)</td>
</tr>
<tr>
<td>Thoracolumbar Kyphosis</td>
<td>-4.7 (-35–16)</td>
<td>-5.1 (-33-9)</td>
<td>-2.7 (21-9)</td>
</tr>
<tr>
<td>Th11 to lowest instrumented vertebra Lumbar</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Lordosis (L1-L5)</td>
<td>49.6 (10-82)</td>
<td>47.3 (20-70)</td>
<td>53.5 (34-73)</td>
</tr>
</tbody>
</table>
coronal Cobb angle of the primary thoracic curve was 55 degrees preoperatively, 17 degrees immediately postoperatively, and 22 degrees at latest follow-up (Table 1). In 7 cases there was an unequivocal loss of correction (> 10 degrees) that averaged 17 degrees (range, 11 to 25 degrees). This was assumed to be the result of wrong fusion levels (double major curve mistaken as a thoracolumbar curve) in one patient, a dislocated hook at the apex of the curve in another and related to crankshaft phenomenon in another. In the remaining 4 cases, no readily apparent cause was found. The lumbar curve (secondary or double major) averaged 40 degrees pre-operatively, 21 degrees immediately post-operatively and 23 degrees at latest follow-up. The average correction of the thoracic curve was 38 degrees (range 23 to 55 degrees). The average correction of the secondary lumbar curve or the lumbar component of a double major curve was 15 degrees (range 0 to 32 degrees). The spontaneous correction of the non-instrumented lumbar curve at latest follow-up averaged 34 degrees (range 10 to 50 degrees).

Coronal balance (deviation from the central sacral line) was on average slightly improved from 13 mm to 11 mm. At latest follow-up, 7 patients had a plumbline deviation from the sacral center of more than 20 mm (20-47 mm), 5 of which were due to a secondary truncal decompensation most likely as a result of CDI. In the remaining two cases, a truncal decompensation was already present preoperatively, but could not be sufficiently addressed through the surgery. The average shoulder elevation increased from 3 degrees to 5 degrees, presumably as a result of the rod derotation maneuver.

The mean values for apical deviation were 42 mm preoperatively, 13 mm immediately post-operatively, and 15 mm at latest follow-up for the primary thoracic curve, 27 mm, 27 mm and 23 mm for the secondary lumbar curve, and 13 mm, 15 mm and 11 mm for the lowest instrumented vertebra, respectively. The tilt of intervertebral disc adjacent to the instrumented spine decreased from 5.1 degrees preoperatively to 3.5 degrees at latest follow-up. Thoracic kyphosis and lumbar lordosis was well preserved or even improved by the instrumentation. There was an increase in the average lumbar lordosis (49 degrees to 54 degrees) as well as in the average thoracic kyphosis (20 degrees to 22 degrees) from the pre-operative to the latest follow-up radiograph.

**Complications**

All patients were doing well and had no complaints with regard to a limitation of work or sporting activities. There were no apparent non-unions, infections or neurological complications. The only minor complications were a pneumothorax subsequent to a thoracoplasty, which responded to chest drainage for one day and one urinary tract infection treated successfully with antibiotics. One patient had an irritated bursa over a prominent hardware part and required partial removal of the implant without any intraoperative signs of infection. There were no other revision surgeries.

**DISCUSSION**

The three-dimensional deformity in idiopathic scoliosis is currently well-recognized, as is the need to not only to correct the coronal deformity but also the sagittal misalignment. While a correction of the sagittal contour was not possible with Harrington instrumentation, Luque instrumentation and CDI were the first implants allowing for a contouring of the sagittal profile and segmental derotation. Luque wiring for adolescent idiopathic scoliosis has now been abandoned because of the potential risk for neurologic complications due to sublaminar wire placement.

Preliminary results with CDI for idiopathic scoliosis have demonstrated that this procedure, like any other, has its specific learning curve that resulted in early complications. As has been summarized by Lenke et al., these problems were the result of a failure to identify appropriate placement of the hooks, failure to pay strict attention preoperatively to the deformity in the sagittal plane and early postoperative coronal decompensation. The most significant problem has been coronal decompensation, which was uncommon with Harrington instrumentation. In this series, 7 patients (13%) had a moderate imbalance (20-47 mm) in the coronal plane at latest follow-up. In 4 of the 7 cases, a balanced spine became subsequently imbalanced by CDI. In the remaining three cases, the existing coronal imbalance was not sufficiently addressed or was worsened. However, the moderate imbalance (21-47 mm) did not require any additional treatment in the seven involved individuals.

We were not able to sufficiently address the important question of apical derotation with this retrospective study design because preoperative computed tomography scans were not available. Although the change in apical rotation noted here indicates an improvement, a more sophisticated analysis is impossible using Nash and Moe’s method. We considered using the Pedrielie method, however, this was not possible in cases where implants obscure identification of the pedicles. The importance of the sagittal contour in adolescent scoliosis today is well-recognized. In our study, we have found that CDI allows for a moderate correction of the thoracic hypokyphosis with an improvement of the average kyphosis from 19.7 (range 0-61) degrees preoperatively to 22.2 (range, 0-61) degrees at latest follow-up. Before surgery, 24 patients had a thoracic...
hypo kyphosis (<20 degrees), 10 of whom had a mild thoracic hypo kyphosis (10-20 degrees). Instrumentation by CDI resulted in a substantial improvement of the hypo kyphotic curve in 10 patients. Similarly, average lordosis could be improved from 49.6 (10-82) degrees to 53.5 (range 34 to 73) degrees. At latest follow-up, none of the patients had a hyplordosis (<20 degrees). These results seem to correlate with those reported by de Jonge et al.13

This paper reports a homogeneous series of patients with right thoracic idiopathic scoliosis treated by a posterior approach with CDI by a single surgeon. The main objective of this paper was to analyze the intermediate and long-term radiographic and clinical results with this instrumentation system. As indicated by this study, the clinical results compare favorably with the results obtained using Harrington instrumentation. Two studies reporting 5 to 10 year clinical and radiologic results of CDI have so far been published. Lenke et al.25 evaluated the radiographic results of CDI in seventy-six patients who had adolescent idiopathic scoliosis with an average of six years follow-up (range, five to ten years). Sixty-three of the 74 patients responded to a questionnaire assessing clinical status. The authors reported that the outcome was favorable with regard to function, cosmetic appearance, and general satisfaction with the operative result. Twenty-four (38 %) of the sixty-three patients reported occasional pain in the spine that did not interfere with work or school activities. Sixty-two patients stated that, given the hypothetical situation of reverting to the preoperative status, they would have the operation again.25 Our results are in line with those by Lenke et al.,25 and correspond well with other who have only presented a short-term follow-up.3,14,17,20,25,30,36

Takahashi et al.37 performed a study focusing on postoperative changes in the lumbar spine in 30 patients with adolescent idiopathic scoliosis who had been treated with CDI. They reported that the prevalence of low back pain increased from 3% before surgery to 20% at the final follow-up visit. None of the patients, the pain was so severe that specific treatment was required. They reported that during the follow-up period, seven patients (23%) developed degenerative changes, including mild junctional kyphosis, retrolisthesis, narrowing of disc spaces, or osteophytes. From their data the authors concluded that the overall clinical and radiographic results of CDI for the correction of adolescent idiopathic scoliosis were satisfactory, but that the unfused lumbar segments requires careful surveillance.37 Although the degeneration of the unfused lumbar segments are a valid concern a comparison with the natural history is required for a more conclusive assessment. Furthermore, the vast majority of disc abnormalities occur with a high prevalence in asymptomatic individuals and are not correlated with back pain.4,40

In conclusion, instrumentation of adolescent right thoracic idiopathic scoliosis with CDI provides encouraging clinical and radiographic results at an average follow-up of 7 years. Overall patient satisfaction, functional status and subjective cosmetic improvement is high. Based on the results of this study, CDI is an effective, reliable and safe procedure for the correction of adolescent idiopathic scoliosis.

REFERENCES


RESULTS OF ADJUSTED-DOSE HEPARIN FOR THROMBOEMBOLISM PROPHYLAXIS IN KNEE REPLACEMENT COMPARED TO THOSE FOUND FOR ITS USE IN HIP FRACTURE SURGERY AND ELECTIVE HIP REPLACEMENT

David Yen, MD, FRCSC, William Weiss, MSc

ABSTRACT

The purpose of this study was to compare the results of adjusted-dose heparin (ADH) in the prevention of thromboembolism in knee replacement with those obtained for its use hip fracture surgery and elective hip replacement. Ultrasound was used to diagnose deep vein thrombosis (DVT) and ventilation/perfusion (V/Q) scan to diagnose pulmonary embolus (PE).

Analysis of 438 operations was available. DVT was present after 9.7% of knee replacements, 7.2% of hip fracture operations and 6.8% of elective hip replacements. PE occurred in 1.2% of knee replacements, 0.9% of hip fracture operations and 2.5% of elective hip replacements. Complications of heparin occurred in 4.6% of patients.

Our ADH protocol was equally effective in prophylaxis against thromboembolism in knee replacement, hip fracture surgery and elective hip replacement. Direct comparison with other methods should not be done because ultrasound was used to screen for DVT.

INTRODUCTION

Patients undergoing major elective lower extremity Orthopaedic surgery or hip fracture are in the highest risk category for thromboembolism. In elective hip replacement performed in the absence of anticoagulation prophylaxis, the prevalence of DVT is 40 to 75% while that of PE found clinically and confirmed by V/Q scanning is 0.53 to 3.5%. Similar studies on patients undergoing knee replacement show the prevalence of DVT to be 40 to 84% and PE to be 0.5 to 2%. Patients with hip fractures also have a significant risk of thromboembolism with DVT occurring in 27 to 75% and clinical PE in 4 to 7%. Many methods of anticoagulation prophylaxis have been reported and the number of different methods advocated supports the fact that the ideal means is still being sought.

Fixed-dose heparin prophylaxis is recommended for use in gynecologic surgery, urologic surgery, neurosurgery as well as in moderate and higher risk general surgery patients, but not for hip and knee replacement and hip fracture surgery. One explanation proposed for the lack of consistent effectiveness of heparin in Orthopaedic surgery is that the doses given are insufficient to maintain levels within a “prophylactic” band. It has been shown that 5000 IU of heparin injected subcutaneously into a normal subject reaches peak levels within one to two hours and returns to zero within 10 to 12 hours. Individuals undergoing major surgery exhibit a “prothrombotic state.” Therefore, they have a shorter duration of measurable heparin levels as well as a partial thromboplastin time (PTT), taken to assess heparin action, that is significantly prolonged over pretreatment levels for only five hours after injection. As a result, these patients have periods of insufficient anticoagulation effect during the trough periods of heparin concentration when the drug is given only once every 12 hours.

Attempts have been made to minimize the trough effect by increasing the dose schedule frequency. However, despite a regimen of 5000 IU of subcutaneous heparin every eight hours, the target PTT needed to improve prophylaxis was achieved in only 27% of observations. Therefore, an increase in the dose of heparin is also necessary.

ADH addresses the trough effect by adjusting the heparin dose according to the PTT. One ADH protocol tested has achieved the target PTT value in 40% of observations. Leyvraz and associates conducted a trial involving elective hip replacement in which ADH was compared to fixed-dose heparin with the resultant rate of DVT being 13% compared to 39% respectively. Further studies have confirmed the effectiveness of ADH.
in elective hip replacement and shown it also to be useful in hip fracture.

Several authors believe that ADH may be useful as prophylaxis against thromboembolism in lower limb orthopaedic procedures, others have stated that it is safe and highly effective in elective hip surgery and recommended its use in knee reconstruction. The purpose of this prospective study was to compare ADH in knee replacement with its demonstrated usefulness in elective hip replacement and hip fracture surgery in order to determine if a uniform method of prophylaxis against thromboembolism was possible in these 3 common Orthopaedic procedures associated with high risk for thromboembolism.

MATERIALS AND METHODS

For an 11 month period, patients undergoing knee replacement, hip fracture surgery or elective hip replacement were recruited for entry into the study. The patients were entered consecutively and no attempt was made to match patients in the three groups.

All patients had a baseline PTT measure at routine pre-admission testing, within two weeks of hospital admission, to screen out those with coagulation disorders. Patient use of acetylsalicylic acid containing compounds and non-steroidal anti-inflammatories was discouraged in pre-admission testing, within two weeks of hospital admission. To risk factors for thromboembolism (previous thromboembolism, a deficiency of antithrombin III, protein C or protein S, varicose veins, malignancy, leg edema, obesity) were found in 61, 42 and 63 of the patients undergoing knee replacement, hip fracture surgery and elective hip replacement respectively. There was no statistically significant difference between the 3 groups with respect to risk factors for thromboembolism (P>0.05). Age over 40 was not included as a risk factor as the patients in all groups were over 40 and, therefore, this factor did not serve to distinguish between the groups.

Subcutaneous sodium heparin (Heplean®; Organon Teknika) was started postoperatively, with the first dose given at 0200 hours on the first morning after surgery. This allowed the PTT to be obtained 6 hours after heparin administration from blood drawn at 0800 hours. The first dose of heparin was 5000 IU. Subsequent doses were adjusted to maintain the PTT between 31.5-36 seconds using a fixed sliding scale reported by Leyvraz and colleagues (Table 1), applied to the PTT result of blood drawn at 0800 hours that morning. The PTT was obtained daily for the first seven post-operative days, on alternate days, for the next seven and then twice weekly for the remainder of the hospitalization.

Early mobilization on post-operative day two, elevation of the foot of the bed and encouragement of ankle pumping exercises was encouraged. Ultrasound using a colour flow doppler (QAD.1, Quantum Medical Systems, Issaqu, WA) was obtained for all patients before discharge from hospital. If clinically indicated, V/Q scanning was used to make the diagnosis of PE. Repeat ultrasound was used to diagnose DVT in patients presenting for medical attention due to leg pain or swelling after discharged from hospital. Chart review was used to screen for thromboembolism in patients after hospital discharge. All data generated was analyzed using the chi-square test for independence.

<table>
<thead>
<tr>
<th>TABLE 1</th>
<th>Sliding Scale for Adjustment of Heparin according to PTT</th>
</tr>
</thead>
<tbody>
<tr>
<td>PTT (sec)*</td>
<td>ADJUSTMENT IN HEPARIN DOSE (IU)</td>
</tr>
<tr>
<td>&lt;27.5</td>
<td>+1000</td>
</tr>
<tr>
<td>28-31</td>
<td>+500</td>
</tr>
<tr>
<td>31.5-36</td>
<td>0</td>
</tr>
<tr>
<td>36.5-39</td>
<td>-500</td>
</tr>
<tr>
<td>&gt;39.5</td>
<td>-1000</td>
</tr>
</tbody>
</table>

* range of normal 23-39 seconds in our lab

RESULTS

There were 531 operations eligible for entry into the study. There were 93 excluded from analysis; in 36 there were no color flow doppler results, in 36 the attending physician chose another protocol for thromboembolism prophylaxis, in 21 the treatment was stopped due to complications from heparin and another form of prophylaxis initiated. Therefore, 438 cases were available for analysis: 165 knee replacements (2 bilateral), 111 hip fracture operations and 162 elective hip replacements.

Risk factors for thromboembolism (previous thromboembolism, a deficiency of antithrombin III, protein C or protein S, varicose veins, malignancy, leg edema, obesity) were found in 61, 42 and 63 of the patients undergoing knee replacement, hip fracture surgery and elective hip replacement respectively. There was no statistically significant difference between the 3 groups with respect to risk factors for thromboembolism (P>0.05). Age over 40 was not included as a risk factor as the patients in all groups were over 40 and, therefore, this factor did not serve to distinguish between the groups.

The average duration of patient follow up in knee replacement was 13 weeks (2-43), in hip fracture surgery 10 weeks (2-44) and in elective hip replacement 12 weeks (2-45). In combining the results of screening before discharge and those requiring further investigation during the follow up period, DVT was found after 16 of 165 (9.7%) knee replacements, 8 of 111 (7.2%) hip fracture operations, and 11 of 162 (6.8%) elective hip replacements. The difference between the results in these groups is not statistically significant (P>0.1). PE occurred after 2 of 165 (1.2%) knee replacements, 1 of 111 (0.9%) hip fracture operations and 4 of 162 (2.5%) elective hip replacements (P>0.1). Proximal DVT occurred after 7 of 165 (4.2%) knee replacements, 4 of 111 (3.6%) hip fracture operations and 7 of 162 (4.3%) elective hip replacements (P>0.1).

There was one death related to thromboembolism. This patient had bilateral knee replacements and post-operatively had a myocardial infarct as well as thrombocytopenia with thromboembolism. The terminal event was
heart failure. Post mortem confirmed the diagnosis of acute myocardial infarction and the presence of multiple pulmonary emboli.

The overall mean heparin requirement was 17,620 IU per day. However, the requirement followed a bell curve, increasing throughout the first post-operative week to peak at 19,184 IU and thereafter gradually reducing again (Figure 1). There was variation in the daily requirements for individuals, ranging from high of 45,000 IU to a low of 3,000 IU.

There were 21 complications of heparin treatment: 13 wound hematomas, 5 gastrointestinal bleeds, 2 cases of heparin induced thrombocytopenia and 1 case of hematuria. Heparin was stopped in all of these patients with resolution of the problem except in 1 case. The exception was the patient who had thrombocytopenia with thromboembolism and died with autopsy findings of acute myocardial infarction and multiple pulmonary emboli. None of these patients required reoperation.

**DISCUSSION**

Warfarin and low-molecular–weight heparin (LMWH) are recommended methods of thromboprophylaxis for patients undergoing hip and knee replacement and hip fracture surgery. Gaining tight control of the international normalized ratio (INR) with warfarin is difficult. In studies on hip replacement, Paiement and colleagues reported that 48% of their patients’ INR values were within the target range during their hospitalization and Lieberman and associates found that 67.5% of their patients’ INR values were within the target range at the time of discharge. In line with these authors, in a study of warfarin for thromboprophylaxis in hip and knee arthroplasty, we found that the last inpatient INR value was within our target range in 64% of our patients. Cost comparison between LMWH and adjusted-dose unfractionated heparin has favored the latter at our centre because the expense of laboratory monitoring of anticoagulation is mainly due to labor costs which could not be eliminated by stopping PTT and platelet measurements in our patients undergoing hip and knee replacement and hip fracture surgery. Patient convenience could not be quantified monetarily. As a result of these concerns with warfarin and LMWH we had an opportunity to study ADH in our patients undergoing procedures that put them at high risk for thromboembolism.

The average heparin requirement in this series followed a bell curve with the apex occurring at day 7 post-operatively. This is consistent with the findings of Horback and associates and fits the hypothesis of a post-operative prothrombotic state. A group of patients reached the target PTT value with only 3000 IU of heparin daily. The ADH protocol prevented dangerous levels of anticoagulation in this sub-group.

There was a 4.6% incidence of complications with our ADH protocol. We noted a hematoma rate of 2.8%. Leyvraz and associates reported 7.9% rate of hematoma in 1 study and 24% in another. Dechavanne and his colleagues found a 10% rate of hematoma. We found other complications that others using ADH have not reported (5 cases of gastrointestinal bleeding, 2 cases of heparin induced thrombocytopenia, and 1 case of hematuria). All of our complications of heparin resolved with discontinuation of the heparin except in 1 case. This individual developed a DVT with PE two weeks post-operatively. His platelet count was 22 X 10^9 at this time. Assays to detect heparin-induced thrombocytopenia (HIT) antibodies were not available but his thrombocytopenia, DVT and PE was consistent with the clinical features described by Warkentin in the HIT syndrome. Based on this experience, we now routinely obtain platelet counts on patients on ADH for over one week or patients who develop thromboembolism while on ADH.

The standard ADH protocol is stated to be complicated, labour intensive and impractical for use in routine clinical practice. Efforts to simplify the regimen by decreasing the frequency of heparin adjustment have resulted in a lowering of effectiveness. We modified the standard ADH protocol by starting heparin...
post-operatively to allow same day admission surgery, to reduce the potential for increased intraoperative hemorrhage and to facilitate the use of regional anaesthesia. A drawback of this modification is that it does not allow for intraoperative protection against DVT but postoperatively initiated prophylaxis has been reported to be effective. We also posted the sliding scale for heparin adjustment at the nursing stations to facilitate the writing of any necessary dose change orders after reviewing the daily PTT results attached to the front the patient chart. With our modified protocol, we found the clinical results over an average follow up of 12 weeks to be acceptable and ADH a practical means of prophylaxis.

Our ADH protocol was equally effective in prophylaxis against thromboembolism in knee replacement, hip fracture surgery and elective hip replacement. Direct comparison with other methods should not be done because ultrasound was used to screen for DVT. Although it is reported as the non invasive test of choice, its limitations in sensitivity and specificity as a screening test for post operative DVT is well described. However, since it was being used to compare 3 groups rather to provide an absolute incidence for DVT, we think that it is a valid tool for determining the relative rates of DVT in this study.

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ABSTRACT

It is difficult to study the deformation of articular cartilage because it is an inhomogenous material with depth dependent constituents. In many experimental studies, cartilage is assumed to be homogeneous and is subjected to only static or quasi-static loads. In this study, a thick walled, mechanically active culture device (TRIAX) was used to apply cyclic loading to cartilage explants at physiological stress levels. An arthroscope was fitted into the wall of the TRIAX to monitor and record the cyclic compressive behavior of the cartilage and to measure depth dependent cartilage strains. A common concern with arthroscopy systems is that the images obtained are radially distorted about a central point (“fisheye” view); therefore it is necessary to correct this distortion in order to accurately quantify distances between objects within the images. To do this, an algorithm was developed which used a calibration pattern to create an image transform. Digital video of the cyclic cartilage compression was recorded, and the distortion algorithm was applied to the images to measure the cartilage as it deformed. This technique will provide valuable and practical insight into cartilage mechanics and viability (via calcein AM-stained chondrocytes) during multi-day cyclic loading of living cartilage explants. The implementation of an arthroscopy system provides the advantage of bringing microscope-level resolution into a cartilage compression device to allow for digital visualization of the entire explant at the whole-tissue level.

INTRODUCTION

Articular cartilage is an inhomogeneous soft tissue covering the articulating surfaces of diarthrodial joints, and is subjected to repetitive loading over many years of use. Cartilage often degenerates due to increased contact stress or acute injury, causing pain and/or loss of joint function (osteoarthritis). It is important to understand the mechanical behavior of cartilage so that the onset and changes associated with osteoarthritis can be investigated. Many studies have documented the deformational behavior of cartilage in vitro using...
methods such as indentation, confined, and unconfined compression. However, in these and many other cases, cartilage was assumed to be a homogeneous material. In reality, cartilage deformation is heterogeneous due to the depth dependent distribution of its constituents (collagen, proteoglycans, etc.).

Numerous investigations using various optical methods have been performed to determine the Poisson’s ratio and anisotropic behavior of articular cartilage. Depth dependent intra-tissue strain distributions using methods such as digital image correlation have been quantified. However, previous work using optical methods for these purposes are limited to static or quasi-static analyses, and rarely do they sample continuous cartilage deformation as a result of physiologic cyclic compression. Because intra-tissue strains depend somewhat on the rate of loading, static or quasi-static analyses do not fully represent strain behavior under physiologic conditions.

A thick walled, mechanically active culture device (Triaxial Compression Vessel, or TRIAX) has been developed with the capability to apply cyclic axial and triaxial loading at physiological stress levels (up to 5 MPa) to articular cartilage explants. An arthroscope has been fitted into the wall of the TRIAX so that the cartilage deformation can be monitored and recorded continuously during cyclic compression (Figure 1). This arrangement affords accurate measurements of depth dependent strains under dynamic loading conditions.

A major concern in arthroscopy systems is the barreling effect, or “fisheye” view caused by the lens within the arthroscope. The resulting images are radially distorted about a central point, called the center of distortion. The deformation increases with increasing distance from the distortion center. Therefore, a square grid will appear distorted when viewed through an arthroscope (Figure 2A). To accurately quantify distances between objects within these images, it is necessary to account for potentially large errors resulting from the barreling effect. Therefore, we present a method for accurate correction of this distortion by creating and implementing an image transform to quantify non-homogeneous deformation of axially compressed cylindrical cartilage explants. Numerous methods for lens distortion correction have been published previously. The technique and application here described is unique in its application to continuous, ‘real time’ measurement of the deformation history of articular cartilage explants under cyclic triaxial loading at physiologic levels.

MATERIALS AND METHODS

The image transform was created through the use of a calibration pattern of arrayed dots, where distorted points were mapped back to their undistorted positions. The amount of distortion is a function of the distance between the tip of the arthroscope and the object of interest; therefore, during calibration the arthroscope tip is positioned at a distance from the calibration pattern equivalent to that employed in the experimental setting.

A SMITH+NEPHEW Dyonics® arthroscopy system 4 mm 0° arthroscope and a Dyonics ED-3 Enhanced Digital 3-chip Camera with a 35 mm focal length were used to capture and record real-time streaming video within the TRIAX via a Pinnacle Systems® MovieBox DV analog-to-digital video converter. The image size output by the arthroscope was 640 pixels wide by 480 pixels tall; its resolution was 10 μm when viewing an object at a dis-
tance of 4 mm. In order for the transform to be valid for our application, it was created in a fluid bath to replicate experimental conditions within the TRIAX. Consequently, a device was built which held the arthroscope tip within a small bath filled with fluid (Figure 2B). The known calibration pattern (Zip-a-Tone, 65 lines per inch, 10% gray) was affixed to a microscope slide; the slide and arthroscope were carefully oriented so that the plane of the calibration pattern was perpendicular to the arthroscope lens and the square grid was aligned horizontally and vertically. The arthroscope lens was placed 4 mm from the calibration pattern within the fluid. A still image (one video frame) of the calibration pattern as seen through the fluid was recorded and analyzed in Matlab (version 6.5.1, Release 13). Within Matlab, a threshold and median filter were applied to the image to compute the centroid of each dot within the binary pattern.

To determine the distortion center, a quadratic line was fit to each row and column of dots; if a row or column contained less than three dots, it was excluded from the calculations. Located between adjacent rows and columns with opposite curvatures are two straight lines; one remains horizontal and the other remains vertical after imaging. The intersection of those straight lines is the center of distortion. For each distorted row or column, the maximum of the quadratic function occurs at the x or y value where the derivative is zero. A horizontal line was fit to the maximum of each column and a vertical line was fit to the maximum of each row; the intersection of those two lines was the center of distortion. After determining the distorted dot locations and the center of distortion, the next step was to calculate the corrected location of the dots to deconvolute the distorted image (i.e., where each dot needed to be shifted in order to correct for the distortion). Because the initial dot pattern was a square grid, the distance between each of the corrected dot locations must be equal. Since the amount of distortion increases with the distance from the center of distortion, the edge length nearest the center of distortion was chosen to establish the deconvoluted grid built off the point nearest the center of distortion. The points on this grid were labeled as the “actual” positions (Figure 3). The distorted and actual dot positions were then input into a custom-written code that created a transform to warp the image from its distorted state. The code utilizes the thin-plate spline method to incorporate the distorted and actual points to map a transform for the entire image (Figure 4). Once the transform was calculated for the specific arthroscope and setup conditions,
Arthroscopic Lens Distortion Correction Applied to Dynamic Cartilage Loading

The digital video system has the capability to record (at 30 frames/second) the arthroscope view of the lateral bulging and progressive cartilage deformation as it accumulates over time during cyclic compression. Measurements within the image are made after the transform has been applied to correct for the distortion.

Full thickness articular cartilage explants were excised and subjected to 2 MPa cyclic axial compression within the TRIAX at a frequency of 1 Hz. The load was applied for approximately half of the duty cycle. The load reached its maximum value in 0.05 seconds to simulate physiological loading rates. The mid-plane silhouette of each explant was manually digitized in its undeformed state and during the first cycle of loading at the point of maximum load. The bulge shape was plotted as the distance between the undeformed and deformed circumferential edge of each explant, and the lateral expansion was defined at the location of the maximum bulge.

Figure 5. (A) Shape of the bulge in the first cycle of loading for explants of varying initial heights. The bulge is shown for only one side of the explant silhouette; the bottom of the explant is at a height of zero. (B) Silhouette for Explant 2 in its unloaded state. (C) Silhouette for loaded Explant 2, accompanied by a black line representing the unloaded condition. The center of each white circle indicates the digitized edge location.

An arthroscope was fitted through the TRIAX wall to view and record the cartilage as it is being compressed (Figure 1). The tip of the arthroscope is located 4 mm away from the mid-plane of the platens. Light to the arthroscope is provided by a Dyonics 300XL Xenon light source attached to the arthroscope, in addition to supplemental ambient light. The digital video system has the capability to record (at 30 frames/second) the arthroscope view of the lateral bulging and progressive cartilage deformation as it accumulates over time during cyclic compression. Measurements within the image are made after the transform has been applied to correct for the distortion.

Porous platens provide axial compression, while nutrient medium pressurization is converted to transverse mechanical compression via a thin, impermeable polyester containment sheath surrounding the 4 mm cartilage explant and platens. An arthroscope was fitted through the TRIAX wall to view and record the cartilage as it is being compressed (Figure 1). The tip of the arthroscope is located 4 mm away from the mid-plane of the platens. Light to the arthroscope is provided by a Dyonics 300XL Xenon light source attached to the arthroscope, in addition to supplemental ambient light. The digital video system has the capability to record (at 30 frames/second) the arthroscope view of the lateral bulging and progressive cartilage deformation as it accumulates over time during cyclic compression. Measurements within the image are made after the transform has been applied to correct for the distortion.

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RESULTS
An analysis was performed to determine the error introduced by the warping algorithm itself. The error analysis essentially determined the discrepancy between the corrected dot locations in the output image after the transform was applied and the “actual” dot locations which were input into the algorithm as the desired locations for the corrected points. The maximum disparity between the dot locations after the transform was applied was less than half of a pixel (~5 μm).

The maximum lateral bulge was measured for three articular cartilage explants loaded within the TRIAX under 2 MPa axial compression alone (Table 1). The relative location of the maximum bulge varied between explants due to differences in the properties of the explants (Figure 5). These differences are likely due to the varying proteoglycan and collagen fiber distributions. The shape and magnitude of the lateral expansion of the cartilage during loading indicates depth-dependent transverse isotropy.

DISCUSSION
In this paper, we present a method to correct arthroscopic lens distortion for application in a device (TRIAX) that applies cyclic compression to articular cartilage explants (4 mm diameter) for optical quantification of heterogeneities in the deformational behavior of the explants. A limitation of this method is that it is important to have clear visibility within the TRIAX of the cartilage edge for an accurate digitization of the midplane silhouette and subsequent bulge calculations.

An additional capability of the system and methods described in this paper involves the potential for further investigation into depth dependent strains throughout the cartilage explant during physiological load cycles, similar to the work done using the cartilage deformation by tag registration (CDTR) method. CDTR involves image MRI tagging and image processing in orthogonal planes to determine nonuniform cartilage deformation during cyclic loading. However, in our case, calcein AM-stained living chondrocytes could be used as fluorescent fiducials and viewed through the arthroscope to determine the inhomogeneous cartilage response and the time and locations of chondrocyte death and/or matrix damage due to excessive mechanical loading. This technique and application will provide valuable and practical insight into cartilage mechanics and viability during multi-day cyclic loading of living cartilage explants. The utilization of an arthroscopy system has the advantage of bringing high-resolution (microscope level) digital visualization to a dynamic cartilage loading device, in order to observe the whole tissue at once, rather than using piece-wise static operations.

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ABSTRACT
This paper describes and discusses some of the clinical applications of the posterior interosseous forearm flap in hand reconstruction. It is based on a series of 20 patients in whom a distally based posterior interosseous island flap was used for closure of defects on the dorsum of the hand. Flaps survived completely in 16 patients. Partial necrosis occurred in one flap. Total flap loss occurred in two patients (20%). The flaps adapted well to the recipient site and had excellent color and texture match. The donor site morbidity was minimal.

INTRODUCTION
Soft tissue reconstruction of the hand remains a challenge to the plastic and reconstructive surgeon. This is partly due to the shortage of skin in the hand and partly to the relative broadness of soft tissue defects in that region. The realization that island flaps can be transferred on retrograde perfusion through a distal vascular arcade opened up an entirely new field in the reconstruction of hand defects. The reverse pedicle posterior interosseous artery flap was first described in 1986 by Zancolli and Penteado. The posterior interosseous artery flap is an island fascio-cutaneous flap vascularized by branches of the posterior interosseous artery. The skin, subcutaneous layer and the fascia along the dorsal ulnar aspect of the forearm are nourished by multiple septo-cutaneous perforators that originate segmentally from the posterior interosseous vessels which surface between the muscles and tendons of the extensor carpi ulnaris and extensor digiti quinti. We report our experience with posterior interosseous retrograde island flaps in reconstruction of hand defects.

MATERIALS AND METHODS
From May 2002 to December 2004, 20 patients were scheduled for retrograde posterior interosseous artery forearm flap. There were 18 males and 2 females. Their ages ranged from 9 to 60 years with an average of 34 years. The majority of the cases were due to thresher injuries, with gunshot injuries being the second most common. The sizes of the flap islands varied from 4x4 cm to 10x8 cm. The arc of rotation centered over the distal radio-ulnar joint. Healing of the flaps was assessed clinically at regular intervals. All complications were recorded. In the operative technique, the size and position of the flap were marked on the axis of the flap, according to the size and position of the defect. The flap was raised under tourniquet. Dissection was done at the ulnar border over the proximal two-thirds of the flap by incising the deep fascia over the extensor carpi ulnaris muscle belly. Dissection continued toward the radial border to reveal perforators of the posterior interosseous artery. The flap was raised, with the deep fascia and the intermuscular septum, to the distal third of the forearm by including a cuff of fibro-fatty tissue around the septum. The island flap was lifted and tunneled to its new position. The flap was sutured into the recipient site. The donor area was covered with a graft.

RESULTS
The flaps healed uneventfully in 16 out of 20 cases. Partial ischemic necrosis was noted in one flap (20%) requiring secondary skin grafting. Two flaps were lost due to venous engorgement, and one patient had associated diabetic angiopathy. The majority of the flaps showed excellent adaptation to the recipient site. Color- and texture-match were good. Donor site morbidity was minimal.

DISCUSSION
The advent of radial and ulnar forearm flaps has increased the safety and reliability of local flaps but involves sacrificing a major artery of the hand. The dorso-ulnar flap has a short pedicle and limited rotation. The anterior interosseous flap demands a tedious dissection involving fragile and anatomically variable vessels. Using a distally based pedicled posterior interosseous flap is a suitable means for providing vascularized skin for covering the dorsal region of the hand as far as the proximal phalanx of the hand. The flap depends on the fascial plexus supplying the skin from the posterior interosseous artery for its blood supply and can be used either proximally or distally.

We have used retrograde posterior interosseous flaps on 20 patients. The age of the patients ranged from 20...
to 60 years with a mean of 31 years. In his study, Dap F et al. (1993) transferred 23 posterior interosseous fascio-cutaneous flaps in resurfacing hand surgery cases. Balakrishnan G (2003) used the reverse-flow posterior interosseous flap in 53 patients and reported tip necrosis in four cases and the pin-cushion effect in 10 cases. Buchler and Frey (1991) used this flap on 15 patients with ages ranging from 5 to 68 years (mean 37 years).

The dominant injured hand in our series was the right (13 cases). Most of the patients in our study were manual laborers and were right handed. There were no safety measures for persons working on the threshing machines. Dap et al. reported use of the flap more in the left hand (15) as compared to the right (8). Landi et al. used the flap to cover defects mostly on the left hand.

In our study males outnumbered females. Other authors' series showed similar male dominance like Zancolli & Angrigiani (1988); Buchler & Frey (1991); Costa H et al. (1991); and Dap F et al. (1993).

In our study, the largest flap was 10x8 cm and smallest was 4x4 cm. Buchler and Frey (1991) used flap sizes which varied from 1.5x4 cm to 9x11 cm. Dap et al. (1993) used flap sizes up to 15x9 cm. In Costa's series (2001), the skin flaps varied in size from 4x5 cm to 14x9 cm. L.J. Lu et al. (2004) reported flap dimensions up to 16x10 cm. In Balakrishnan's series (2003), the smallest flap measured 5x2.5 cm and the largest flap measured 21x10 cm (210 cm²). Buchler and Frey used flaps whose size varied from 1.5x4 cm to 9x11 cm. In Landi's series, flap islands varied from 4x7 cm to 11.5x5.5 cm. Costa H, Gracia et al. reported flap islands which varied in size from 4x5 cm to 14x9 cm.

In our study, partial flap loss was seen in one patient with a defect over the palm. The secondary defect was covered with a split skin graft. Dap(1991) noted partial ischemic necrosis in seven (21%) of the 34 flaps, as flaps showed a tendency toward venous congestion. This was localized at the site of the relevant septocutaneous perforator. Shibata M et al. (1997) noted partial necrosis in two flaps and two flaps had epidermal necrosis. Chen et al. (1998) reported partial flap loss of 18.2% (two out of 11 flaps) in their series. Buchler and Frey reported four cases (25%) of infection which caused partial necrosis of the flap.

In our study only two patients (10%) had total loss of flaps. One patient with a known case of diabetes suffered venous congestion and the flap was lost despite conservative treatment. The other was lost due to arterial insufficiency because there was no perforating branch. Landi et al. also were not able to identify pedicle vessels in two patients. Buchler and Frey found the vessels to be missing in two cases. Angrigiani et al noted absence of continuity of the posterior interosseous artery in two cases. L.J. Lu et al. (2004) reported loss of one flap out of 90 cases because of vascular deficiency. Chen H et al. (1998) reported a failure rate of 21.3% (three out of 14 flaps).

Jones reported acute ischemia of the ipsilateral hand after harvest of a free radial forearm flap. No vascular ischemia was seen in our series. In our study, all donor areas were covered with split skin graft. The secondary defect was more acceptable than that of the radial forearm flap since skin grafts survive well on exposed muscle and the final appearance was satisfactory. Postoperative immobilization was not necessary. L.J. Lu et al. reported no adverse loss of function of the hand and forearm. Dap et al. performed primary closure of the donor area in four cases and in the other 19 patients split-thickness skin grafts were placed on the donor area. Costa, Gracia et al. reported direct closure of donor sites smaller than 7x6 cm, whereas larger defects were covered with a skin graft.

At the time of final evaluation, all recipient flap sites had healed. The final appearance of the hands with dorsal defects was found to be acceptable because the flap was thin, reliable and versatile. Zancolli et al. also reported the same. Donor site morbidity was insignificant from a cosmetic and functional point of view in smaller flaps. In larger flaps, there was some cosmetic disfiguration but this improved with time and the forearm was quite acceptable.

The flap is thin, hairless, and has an excellent texture for skin resurfacing. This flap contours easily. It offers the benefits of being single-stage, reliable, versatile with minimal donor site morbidity. This flap is composed of less fat as compared to the groin, lateral arm and radial artery forearm flap. In conclusion, we find that the distally-based posterior interosseous flap has proven to be reliable and very useful for reconstruction of the first web space as well as the dorsal aspect of hand.

REFERENCES


MODIFICATIONS OF THE FULKERSON OSTEOTOMY: A PILOT STUDY ASSESSMENT OF A NOVEL TECHNIQUE OF DYNAMIC INTRAOPERATIVE DETERMINATION OF THE ADEQUACY OF TUBERCLE TRANSFER

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ABSTRACT

Medial transfer of the tibial tubercle is commonly implemented to correct patellar alignment in patients with patellar instability. However, the extent of transfer needed is difficult to determine. This article reports a pilot-study experience with a novel technique employing intraoperative femoral nerve stimulation to better determine the distance of tubercle transfer required for proper patellar tracking. This pilot study is a case series involving seven knees, all with a clinical history of dislocation, evidence of maltracking, and excessive medial patellofemoral ligament (MPFL) laxity to the point of producing a positive apprehension sign. All seven knees received femoral nerve stimulation for patellar tracking assessment as part of a modified Fulkerson osteotomy. All knees received clinical follow-up for a minimum of 24 months. Six of the seven cases in this series remained stable during two years of follow-up. Through these findings we conclude that the use of femoral nerve stimulation for patellar tracking assessment may be associated with a sufficiently high rate of success to warrant more extensive investigation.

INTRODUCTION

Patellar instability is a common and complex problem that is at times both difficult to characterize and difficult to treat. There are many underlying predisposing factors that may lead to patellar instability. These factors include abnormalities in muscle origin and insertion and the dynamic forces created by them as well as significant anatomic malalignment of the femur, tibia, or patella. Additionally, deficiencies of passive restraints (e.g., medial patellofemoral ligament), patella alta, trochlear dysplasia, soft tissue dysplasia, abnormal foot/ankle alignment, genu valgum, and excessive lateral tibial torsion are thought to contribute to patellar instability. Due to its complexity, over 100 surgical methods have been described to treat this condition. It is clear that no single surgical option is appropriate for every patient with patellar instability and treatment decisions must be made based on the underlying pathology.

It is the senior author’s experience that a lateral retinacular release along with direct medialization of the tibial tubercle and a “sliding dovetail” procedure (described by Southwick et al.) are very successful in stopping dislocation episodes. However, it became apparent that this procedure resulted in a mechanical displacement of the tubercle attachment posteriorly in the sagittal plane. Although the sliding dovetail procedure had been highly successful in relieving instability, it did little to relieve the fairly common symptom of patellofemoral pain. Many surgeons sought ways to improve the procedure by providing more symptomatic pain relief.

Anteromedialization of the tubercle provides stabilization of the patella without mechanical displacement as seen in dovetail direct medialization. This realignment procedure has also been previously demonstrated to produce a high percentage of positive outcomes in correcting instability. For these reasons, the anteromedialization procedure has gained popularity in recent decades. The Fulkerson osteotomy is a realignment procedure combining an anteromedial transfer of the tibial tuberosity with or without release of the lateral retinaculum.

The Fulkerson procedure theoretically reduces patellofemoral pain by anteriorizing the tibial tubercle, thereby reducing joint contact forces, while at the same time medializing the extensor mechanism. However, in the senior author’s experience, the success rate for achieving stability with the Fulkerson procedure was lower than the rates reported in the literature, and lower than the success achieved with the dovetail procedure. It was also noted in the dovetail follow-up study that roughly one in five patients, while not experiencing dislocations, did exhibit a positive apprehension sign. These observations led to modifications in the existing operative techniques.
First, in order to eliminate the positive postoperative apprehension sign, an addition to the traditional Fulkerson was needed. Recent emphasis on the existence and role of the MPFL as a stabilizing structure for the patella made it a repair an attractive addition to the Fulkerson procedure. Therefore, the senior author employed reefing and reattachment of the medial capsule and MPFL as a routine method of improving results. Second, a more exacting method was sought to see how far the tibial tubercle should be transferred to achieve proper alignment. With the availability of regional anesthesia at our institution, we developed a method of intraoperative quadriceps activation by femoral nerve stimulation.8

The purpose of this pilot study is to assess the results of modifying the Fulkerson osteotomy with the use of intraoperative femoral nerve stimulation. We hypothesized that adding dynamic assessment to the more traditional passive manipulations would lead to a stable extensor mechanism with optimal alignment of dynamic forces.

**METHODOLOGY**

There were seven knees in six patients (one bilateral) in this series that had a history of recurrent lateral patellar dislocation. Patient age range was 15-37 with an average age of 22. All patients were in good general health. Patients with Dejour type III trochlear dysplasia,3 the most severe type, or those with obvious “miserable malalignment” (excessive femoral anteversion compensated by external tibial torsion) were not included. All of the patients in this study were indicated for treatment based on a clinical history of recurrent dislocations and correlating examination findings. They all demonstrated excessive lateral patellofemoral laxity and a positive apprehension sign. All were also found to have signs of lateral maltracking including combinations of lateralization of the patella on merchant view, abnormal Q-angle, and/or a J-sign with active extension. Patients who received reconstruction of the medial patellofemoral ligament (MPFL) or lateral patellofemoral ligament (LPFL) were eliminated from this study. All patients in the study were followed for a minimum of two years postoperatively at The University of Iowa Sports Medicine Clinic. Follow-up range was from 25 months to 148 months with an average follow up time of 47 months. At clinical follow-up visits, these patients were assessed for recurrent dislocation, apprehension sign, and J-sign for maltracking. Patients who had one or more of these three criteria were considered outcome failures. Patients who had none of these symptoms were considered to have a successful outcome.

These cases were gathered between March of 1994 and November of 2005 at the University of Iowa Hospitals and Clinics. The novel surgical procedure performed on these knees is described below.

**Technique: Fulkerson with limited lateral release, medial capsular repair/plication, and femoral nerve stimulation**

Patients in this case series underwent a modification of the traditional Fulkerson osteotomy with the addition of plication of the medial capsule for repair. The technique initially described elsewhere9 is detailed here. Preoperatively, the patient had a “stimulating” femoral catheter placed on the femoral nerve. After induction of general anesthesia (or spinal anesthesia) but before surgery, the settings of the stimulating catheter were calibrated so the quadriceps muscle contraction elicited was of physiologic speed and force. At that time, a first assessment of patellar tracking was made by contraction of the quadriceps muscle with the patient under anesthesia. That assessment was compared to the preoperative assessment in the conscious patient.

Following this calibration, a single long incision lateral to the patella was made and the patellar tendon was identified and dissected so its distal attachment had full exposure medially and laterally. Moving then to the tibia, the bone was cut beneath the tibial tuberosity at an oblique angle posteriorly, from medial to lateral. The proximal end of the tubercle was then cut in a “dowel” shape while the distal end remained attached. A tamp was then used to slide the tubial tubercle medially and anteriorly along the oblique cut to a desired distance that was measured with a ruler at the proximal end of the cut. Once it was in the desired position, the tubercle was temporarily fixed in place. After this initial bone transfer was complete, lateral patellofemoral structures were evaluated. Lateral retinacular release was then performed when required, based on the tightness of the lateral structures, to achieve soft tissue balancing. The buttress effect of the proximal dowetail along with the temporary fixation of the tubercle, with temporary pin fixation and thumb pressure on the proximal tibia, allowed the surgeon to assess tracking in a variety of ways.

Assessments of patellar tracking in these cases were done in two ways. First, using passive intraoperative manipulation of the knee, and second, by observing active tracking with femoral nerve stimulation.9 For the passive intraoperative manipulation, the first maneuver to observe patellar tracking was moving the knee from full extension to full flexion without any rotation and watching the patella for maltracking medially or laterally. Next, to check for under-correction of tubercle placement, the patella was manually displaced laterally in the fully extended knee and then released at the moment passive knee flexion was initiated, with the coupled forces of external rotation and valgus applied to the knee. Lateral dislocation with this motion indicated under-correction.
Finally, to assess over-correction, the patella was displaced medially with the knee extended, then the knee was flexed and varus and internal rotation forces were applied. Medial dislocation in this maneuver indicated over-correction and/or excessive lateral release.

After passive assessment of patellar tracking, intraoperative femoral nerve stimulation was performed. Through the calibrated femoral nerve catheter the quadriceps muscles were stimulated to actively move the knee from flexion to extension against gravity. Special care was taken to note the presence or absence of a J-sign in active extension of the knee. Based on observation of the active patellar tracking, adjustments were then made in the distance of tubercle transfer. A final assessment of tracking was done before the tubercle was secured with two bicortical screws at the position that demonstrated the best tracking.

Following tibial tubercle transfer, all patients in this pilot study received plication of the medial capsule to reduce redundancy of the medial structures created by the tubercle transfer. The integrity of the medial capsules in all seven cases was judged to be sufficient to avoid restretching postoperatively. The medial patellofemoral ligament was assessed manually with the medial structures under tension. In all seven cases, the MPFL was believed to be both present and of adequate integrity to allow for repair as opposed to reconstruction. The repair was performed in each case after assessing patellar tracking. The capsule and MPFL were attached to the patella with bone suture anchors.

Patellar tracking was always assessed prior to plication of medial structures and after release of the lateral retinaculum. This was done so that tracking could be assessed without any medial or lateral restraints. This allowed for adjustments to be made in both the extent of the lateral retinaculum release, and the tightness of medial reefing, in order to fine-tune patellar tracking.

RESULTS

There were seven total cases in this pilot study. Of these seven cases, one patient had a postoperative positive J-sign and recurrent dislocation at the two-year follow-up visit. The six positive outcomes in this group had no apprehension or J-sign, and no recurrent dislocation.

DISCUSSION

In the years prior to the development of this technique, patients at our institution received only the modified Fulkerson osteotomy without dynamic assessment of patellar tracking. During that time period it was noted that nearly all of the few patients who continued to show patellar instability following the Fulkerson without femoral nerve stimulation had a positive J-sign on follow-up. Our theory is that by using femoral nerve stimulation to dynamically assess patellar tracking and appropriate placement of the tubial tubercle, intraoperative maltracking should be eliminated as well as any postoperative J-sign.

This first series of patients has shown promising results. We have been impressed with the ability of the femoral nerve stimulation technique to reproduce the same pattern of quadriceps firing demonstrated by the conscious patient. Although we have not yet validated this observation of reproducibility between conscious and anesthetized patients, that validation study is currently under way.

In looking at our novel technique in relation to other possible ways for achieving patellar stability, the most important difference to note is this technique’s ability to balance dynamic structures. Other authors, such as Teitge, have shown that MPFL reconstruction alone can eliminate instability in patients with or without an adequate trochlear groove. However, those studies do not directly address the additional problem of dynamic and bony malalignment. It is our contention that principles learned during the polio era still hold true; static repairs will not last forever if the dynamics involved are not also correct. It is speculated that with the passage of time, MPFL repairs with persistent pathologic dynamics will eventually loosen again. Additionally, simple correction of bony malalignment, such as the traditional Fulkerson, without consideration of pathologic dynamics may end in the same poor result in the long term. Certainly this short-term pilot study cannot illuminate that issue.

Implementation of this technique has been successful as shown in the results of this case series. There was a high success rate (six of seven) in cases where femoral nerve stimulation was implemented. During these cases the transfer distance of the tibial tubercle was changed anywhere from zero to six millimeters because of the observations made during the femoral nerve stimulation. When the tubercle transfer distance was changed based on the femoral nerve stimulation it was always transferred further medially. This often resulted in a medial transfer distance that was larger than originally thought necessary. There were no medial dislocations postoperatively despite this, suggesting another possible advantage to using this technique. As muscle strength improves postoperatively there may be a tendency for a stronger VMO to pull the patella medially. Assessing forceful contraction of the VMO intraoperatively may predict this change at the time of surgery and allow for adjustments to be made. To determine the significance of using dynamic assessment as part of the Fulkerson
ostectomy, a large prospective study that evaluates outcomes with and without use of femoral nerve stimulation for dynamic balancing is needed.

The experiments in the above paper comply with current laws and ethical standards set by the United States Government as well as this institution’s Internal Review Board.

REFERENCES
GREATER TUBEROUS OSTEOTOMY AND TERES MINOR TRANSFER FOR IRREPARABLE SUPERIOR ROTATOR CUFF TEARS

Brian R. Wolf, MD*, Andrew D. Bries, BS*, James V. Nepola, MD*

ABSTRACT
The purpose of this study was to evaluate the mid- to long-term objective, subjective and radiographic results of patients who underwent antero-superior transfer of remaining infraspinatus tendon and teres minor tendon for irreparable superior rotator cuff tears. Thirteen patients were identified who underwent infraspinatus tendon transfer to a more superior position on the humeral head between January 1, 1990 and December 31, 2001. Nine shoulders in eight patients were available for clinical examination, radiographs and questionnaire follow-up at an average of 83.5 ± 31.4 months. Radiographic examination revealed 1 fibrous union and 6 united tuberosity osteotomies. Samilson-Prieto grading of radiographs revealed 4 shoulders with mild, and 4 shoulders with moderate, OA. Seven of the patients were satisfied with their shoulder. There were two poor outcomes. Local antero-superior teres minor and residual infraspinatus transfer provides a viable option for irreparable rotator cuff defects. Mid- to long-term satisfactory outcome was achieved in 7 out of 9 shoulders.

INTRODUCTION
Treatment for massive irreparable rotator cuff tears is challenging. There have been published reports of local tendon transposition and distant tendon transfer to solve this problem. Subscapularis tendon transfer has been attempted using the upper third of the tendon to cover lesions involving the supraspinatus and infraspinatus. This technique is not commonly used as it has increased risk of internal rotation weakness or internal rotation contracture. Advancement of the supraspinatus as described by Debeyre et al has offered some promise. However, supraspinatus advancement can require extensive dissection and care must be taken to avoid neurovascular damage thus making this technique less than attractive in cases of severe supraspinatus degeneration. Latissimus dorsi transfer has been used frequently for loss of function of infraspinatus and supraspinatus thus providing an external rotation and humeral head depression moment arm. Latissimus dorsi transfer does, however, require extensive dissection of the shoulder and back, and has significantly increased morbidity. Despite this, it can provide good results for massive irreparable tears. Use of a deltoide muscle flap and trapezius transfer have also been described.

The purpose of this study is to present series of patients who underwent a modification of a previously described technique using local greater tuberosity osteotomy and transfer of the teres minor and any residual infraspinatus tendons for treatment of shoulder pseudoparalysis and irreparable superior rotator cuff tears, and to report the mid- to long-term outcome of these patients. Despite its previous description, the authors could not find any clinical follow-up of such surgical management in the peer-reviewed literature.

MATERIALS AND METHODS
After obtaining institutional review board approval, patients were identified for the study via a chart review of the operative notes of one of the authors (JN) from January 1, 1990 to December 31, 2001. Criteria for inclusion were a patient with an irreparable superior rotator cuff tear involving at least the supraspinatus, who underwent teres minor and residual infraspinatus transfer greater than 2 years prior to review.

All patients underwent antero-superior transfer of the teres minor and any residual infraspinatus to close
an irreparable superior rotator cuff defect. Seven of the 8 shoulders had undergone osteotomy of the greater tuberosity with fixation during the transfer. One additional patient had undergone soft tissue transfer without bony osteotomy.

Preoperative range of motion, strength and operative indication were obtained from the patients' charts. At the follow-up exam the patients were asked to complete the Western Ontario Rotator Cuff Index (WORC) and the L'Insalata questionnaires. An independent examiner performed passive and active range of motion and manual rotator cuff strength testing. Radiographic evaluation included AP, axillary, and outlet views and the radiographs were assessed for osteoarthritic changes utilizing the Samilson-Prieto grading system, again by an independent examiner. Acromio-humeral distance and healing of the greater tuberosity were evaluated as well. Acromio-humeral distance, measured on AP view, was used to assess superior humeral head migration. Available pre-operative radiographs were evaluated in similar fashion and compared to the follow-up radiographs.

**Surgical technique**

An arthroscopy may be performed and subacromial decompression may be carried out if deemed necessary. An open approach to the shoulder is then carried out beginning at the acromioclavicular joint and extending over the anterolateral corner of the acromion. The deltoid raphe is identified and the deltoid is split in line with its fibers. The split is carried superiorly and the delto-trapezial fascia is elevated off the acromion anteriorly and posteriorly until adequate exposure of the subacromial space and greater tuberosity is achieved. Traction sutures are placed in the edge of the residual rotator cuff tendon. Blunt dissection is used to mobilize this residual tendon in an attempt to perform a primary repair back to the tuberosity. If the superior cuff defect is irreparable (Figure 1) and residual intact posterior cuff consisting of teres minor and perhaps some infraspinatus tendon is present, then advancement is begun.

The intact insertions of the teres minor and any residual infraspinatus are clearly marked on the tuberosity. A small oscillating saw or osteotome is then used to osteotomize a portion of the greater tuberosity of approximately 2 x 2 cm (or 2.5 cm diameter) where the intact posterior cuff is attached (Figures 2 and 3). The osteotomized fragment is ideally at least 5 mm in thickness. The superior anterior and inferior cortical borders of the fragment are cut with a micro sagittal saw. Then a thin osteotome of appropriate size is used to complete the osteotomy posteriorly, prying the frag-
ment up to carefully include the tendon insertion on the fragment. A small amount of bone is resected from the recipient site of the irreparable defect anteriorly on the greater tuberosity matching the shape of the fragment to be transferred so the transferred tuberosity will not be prominent and will not cause impingement on the acromion.

Fixation of the fragment is most typically achieved with one 3.5 mm cortical screw. The fragment and tendon are transferred antero-superiorly to the desired location and temporarily fixed in place with two 0.45 mm or smaller Kirschner wires (Figure 4). Fixation is then finalized with a centrally placed 3.5 mm fully threaded cortical screw with a washer (Figures 5 and 6). Bi-cortical purchase is achieved by carefully directing the screw toward the infero-medial surgical neck, taking care not to over compress and split the fragment. The screw should be of appropriate length and should not penetrate the neck of the humerus medially by more than 4 mm. Fluoroscopy or plain AP radiograph are used to confirm appropriate hardware placement. Once fixation to the tuberosity is complete the transferred tissue can be sutured to the subscapularis tendon to close any residual antero-superior defect (Figure 7).

Alternatively, fixation can be accomplished with suture anchors. When using suture anchors usually one or two Bionx pins (ConMed-Linvatec, Utica, NY) will be utilized to stabilize the fragment. Then 2 to 3 anchors are placed at the edges of the trough deep to the transferred fragment. The sutures from the anchors are passed through the thin fragment using a needle. These holes through the fragment need to be placed carefully, according to the desired final location of the tendon-tuberosity fragment.
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Post-operatively patients are immobilized in a shoulder immobilizer with a small abduction pillow. The patient is allowed early pendulum activities and gradual institution of passive range of motion over the first month. Active-assisted range of motion or resistive exercises should not be started until there is radiographic evidence of bony union.

RESULTS

Thirteen patients were identified and contacted via phone and mail. Of the 13 patients and 14 cases, 7 patients with 8 shoulders returned to clinic to undergo clinical and radiographic examination and questionnaire evaluation. Of the six patients who did not return, two refused to participate, two could not return for health reasons, one patient was deceased and one patient’s procedure failed and had progressed on to a total shoulder arthroplasty.

The mean follow-up was 83.5 ± 31.4 months. The mean age at surgery was 56.6 ± 12.3 years. Six of seven patients were men. Five of the eight cases were revisions of failed previous rotator cuff repair. One patient had failed 2 prior repairs and one patient had failed three prior repairs.

Pre-operative values for active flexion were available by chart review for all eight shoulders. The mean pre-operative value was 70.6 ± 21.3 degrees.

Clinical evaluation at follow-up revealed a mean active flexion of 110.6 ± 52.9 degrees and active abduction of 98.8 ± 51.9 degrees. The mean increase in active flexion post-operatively was 40.0 ± 42.4 degrees. Table 1 summarizes basic demographic information and results for each case.

External rotation strength was graded as 5 out of 5 manually on 2 shoulders, 4/5 on 4 shoulders and 3/5 on 1 shoulder. Manual forward flexion strength in the scapular plane was 5/5 in 3, 4/5 in 2 and 3/5 in 2 shoulders.

Samelson-Prieto grading of radiographs revealed 4 shoulders with mild osteoarthritis and 4 shoulders with moderate osteoarthritis. Radiographic examination revealed 1 fibrous union and 6 united tuberosity osteotomies. Despite single screw fixation of the tuberosity fragment in most cases, no fragments appear to have rotated.

Questionnaire follow-up showed a mean WORC score of 76.3% (range, 44-85.6%) and a mean L’Insalata score of 76.7 (range, 52.0-97.7). Seven of the patients were satisfied with their shoulder. There was 1 poor outcome. The one poor result had undergone 3 prior failed repairs by the age of 41 and was a worker’s compensation case with debilitating shoulder pain. This patient later went on to have a hardware removal and then later failed a latissimus dorsi tendon transfer. He has subsequently been offered shoulder fusion for chronic pain but has refused. None of the other patients had undergone any further surgery.

### TABLE 1

<table>
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<th>Case</th>
<th>Sex</th>
<th>Age at Surgery (mos.)</th>
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<th>Active Flexion (degrees)</th>
<th>Acromio-humeral distance (mm)</th>
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<td>(12.3)</td>
<td>(31.4)</td>
<td>(21.3)</td>
<td>(52.9)</td>
</tr>
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DISCUSSION

Neviaser and Neviaser originally reported on 17 patients who underwent superior transfer of the teres minor tendon with or without superior transfer of the subscapularis as well for irreparable tears. The transferred tendons were repaired primarily to the greater tuberosity and relied on tendon to bone healing. This series resulted in 12 good/excellent results and 5 poor results. The number of patients that needed subscapularis transfer in addition to the teres minor transfer was not reported. This procedure was used for salvage of otherwise irreparable superior rotator cuff tears. Paavolainen modified the original technique of teres minor transfer described by Neviaser et al. by incorporating a greater tuberosity osteotomy in an effort to enhance healing. Performing an osteotomy of the greater tuberosity and using rigid fixation allows bone to bone healing, as opposed to relying on tendon to bone healing of transferred tissue, which in a patient with a torn rotator cuff may be compromised. He reported a series of 31 patients at a minimum of 1 year after surgery. Relief of pain was seen in 93% of patients and functional improvement was seen in 90%. Average post-operative abduction was 135 degrees. The currently described modification of the technique allows for rotation of the tuberosity fragment to maximize superior defect closure and, in our opinion, is less technically demanding. Six of seven osteotomies in this series showed bony healing and one patient had a fibrous union that was asymptomatic. The current series is very similar in terms of post-operative pain relief, function and motion.

Another frequent method of treating irreparable cuff defects is latissimus dorsi transfer. Recent series report 82-94% relief of pain during rest and with exertion and results are much better with an intact subscapularis. These series also report restoration of approximately 80% of function in the presence of an intact subscapularis. The functional outcomes in the current series and the series reported by Paavolainen appear comparable to those achieved with latissimus transfer. An additional benefit is that greater tuberosity transfer is performed through a standard approach traditionally used for open rotator cuff repair. This allows attempted primary repair, and progression to local transfer if primary repair is not feasible, through the same approach.

The intended functional impact of superior transfer of the teres minor and residual infraspinatus is to increase the abduction moment arm of the remaining rotator cuff. The posterior cuff musculature has been found to contribute to abduction and flexion in the scapular plane in its native position. Theoretically, with superior transfer of these muscle tendon units this function would be advanced. This has been shown with superior advancement of the subscapularis for irreparable cuff defects. To our knowledge, it has not been studied for transfer of the residual posterior cuff.

This study is limited by its small size. Of the 12 patients still living, we had one poor result and another patient who has subsequently undergone arthroplasty. In addition, two patients refused to return. However, in those patients evaluated, the results in this series provided good improvement in seven of eight shoulders that included pain relief and improvement of motion at a mean of nearly seven years. Despite these good clinical results, radiographic exam revealed a prevalence of narrowed acromio-humeral distance and high riding humeral head suggesting that proximal migration of the humeral head is not reversed and may, in fact, get worse. Lastly, half of the cases had moderate arthritic changes on follow-up.

As evidenced by our small series size, the indications for this operation are limited as other techniques of repair are often possible. Cases potentially treated with this technique would include chronic retracted L-shaped tears and two tendon tears that are not amenable to standard medial-to-lateral or side-to-side repairs. Superior transfer of the posterior cuff offers an alternative when other means of repair are not feasible. In conclusion, local teres minor and residual infraspinatus transfer has proven to be a viable option in this small selected case series of salvage rotator cuff repairs.

REFERENCES


ABSTRACT
Anterior femoroacetabular impingement (FAI) is a major etiologic factor in the pathogenesis of hip arthritis. In this condition, mechanical abnormalities of the hip joint lead to early hip dysfunction, inflammation, cartilage injury, and eventual joint degradation. FAI is now more commonly diagnosed and there is an increasing need for a thorough understanding of the broad spectrum of clinical presentation for the disease as well as more precise definition of the possible surgical options.

INTRODUCTION
There is increasing evidence that anterior femoroacetabular impingement (FAI) is a major etiologic factor in the pathogenesis of hip osteoarthritis. In this condition, deformities of the femoral head-neck junction, acetabulum, or both, produce abnormal contact around the periphery of the joint. Articular pathomechanics result in clinical signs of hip dysfunction, and can mediate articular cartilage delamination, acetabular labral disease and progressive secondary joint deterioration. FAI is now being diagnosed more commonly, and there is an increasing need for joint preservation surgical treatment. As the diagnosis and treatment of this condition becomes more commonplace, it is critical to emphasize that this disorder encompasses a spectrum of disease patterns, and deformity severity as well as varying degenerative stages. Accordingly, surgical techniques continue to evolve. The optimal surgical solutions must be defined, and may vary according to the underlying disease type, patient-related factors and the technical expertise of the surgeon. In this article, we review the clinical and radiographic characteristics of hip impingement disorders, summarize the contemporary treatment options and present our surgical preferences for distinct disease patterns.

IMPINGEMENT DISEASE PATTERNS
Murray first introduced the concept of hip impingement as an underlying cause of degenerative joint disease, and Stuhlberg subsequently described the “pistol-grip” deformity noting its presence in 40% of all patients who developed hip osteoarthritis. Harris linked osteoarthritis to residual structural abnormalities of the joint resultant from childhood hip diseases such as acetabular dysplasia, Legg-Calve Perthes (LCP), and slipped capital femoral epiphysis (SCFE). More recently, Ganz and colleagues have popularized the concept of impingement as a cause of pre-arthritic and early arthritic hip symptoms and secondary osteoarthritis. Two types of FAI, namely cam and pincer, have been described as having distinct joint pathomechanics and unique intraarticular disease characteristics. It should be emphasized that these two disease patterns can occur alone, yet commonly, components of both cam and pincer impingement coexist (Figure 1).

Cam Impingement
Structural impingement deformities of the proximal femur are referred to as “cam” impingement abnormalities (Figure 1). The defining characteristic of these disorders is an abnormal osteocartilaginous prominence located at the anterolateral femoral head-neck junction. The deformity decreases the head-neck offset, increases the femoral head radius of curvature and results in relative retroversion of the femoral head. The normal smooth arc of motion between the femoral head and acetabulum is disrupted due to the loss of femoral head-neck junction concavity. As the aspherical femoral head is forced into the acetabulum during hip flexion, compressive and shear stresses at the labral-articular cartilage junction push the labrum towards the capsule and the adjacent articular cartilage into the joint. Hip chondral injury may range from fissuring to complete delamination and usually precedes acetabular labral tear. Labral tears often occur through the weaker 1-2 mm transition zone between articular hyaline cartilage and labral fibrocartilage. With cam impingement, labral tears are usually associated with articular cartilage delamination, and articular-sided labral tears are more common than...
capsular-sided tears. Acetabular cartilage and labral injuries in cam impingement primarily occur along the anterior and superolateral acetabular rims. Cam impingement has been reported in conjunction with SCFE deformity, Legg-Calve Perthes, posttraumatic femoral neck fracture malunion, elliptical femoral heads, and decreased femoral anteversion. While linked to numerous anatomical anomalies, the underlying etiology of cam impingement in many cases remains unknown. Siebenrock has suggested an abnormal separation of the proximal femoral epiphysis into the capital epiphysis and the trochanteric apophysis as a potential cause of a non-spherical femoral head.

Pincer Impingement

Pincer impingement results from excessive anterolateral femoral head coverage due to abnormal structural anatomy of the acetabulum (Figure 1). Acetabular overcoverage is observed in coxa profunda, protrusio acetabuli, coxa vara and acetabular retroversion. Acetabular retroversion may be seen in isolation, in association with acetabular dysplasia, as a result of childhood trauma, or after overcorrection with a reconstructive ostectomy (Figure 2). In these clinical situations, the femoral neck abuts the prominent acetabular rim during joint motion leading to repetitive abutment and cartilage damage.

Figure 1. Femoroacetabular disease patterns. The reduced clearance during joint motion leads to repetitive abutment between the proximal femur and the anterior acetabular rim. (A) Normal clearance of the hip, (B) reduced femoral head and neck offset (cam impingement), (C) excessive over coverage of the femoral head by the acetabulum (pincer impingement), and (D) combination of reduced head and neck offset and excessive anterior over coverage can be seen (combined impingement).


Figure 2A

Figure 2B

Figure 2C

Figure 2. Anteversion PAO for the Treatment of Acetabular Retroversion. This 28-year-old female had a history of bilateral periacetabular osteotomies in adolescence. She presented 10 years after the left sided PAO with symptomatic femoroacetabular impingement due to retroversion of the acetabulum. This anteroposterior radiograph (A) demonstrates major retroversion of the left acetabulum (B) when compared to the right. The patient was treated with anteversion PAO and has an excellent result two years after that procedure (C).
hip flexion creating impingement mechanics. Repetitive microtrauma between the femoral neck and acetabular rim may cause multiple cleavage planes within the labrum leading to labral hypertrophy, intrasubstance cyst formation, calcification and labral degeneration. Acetabular labral ossification further deepens the acetabular socket and increases acetabular overcoverage. In contrast to cam impingement, the acetabular labral disease may not be associated with adjacent acetabular cartilage degeneration. In pincer impingement, acetabular chondral injury occurs in a narrow circumferential band at the anterior and superolateral acetabular rim where force transmission is greatest, and as a “contre-coup” lesion in the posterior capsular-labral junction by a distraction force due to femoral head levering out of the hip socket with continued hip flexion. The “contre-coup” injuries tend to be larger in pincer impingement than cam impingement disorders. Femoral head cartilage damage occurs with cam and pincer impingement, but in early disease is less prevalent than acetabular cartilage damage.

PATIENT EVALUATION
Clinical Presentation
Femoroacetabular impingement typically presents in active, young or middle-aged adults. Cam impingement is most common in young male patients, while pincer impingement is a more likely diagnosis in middle-aged female patients. Additionally, FAI is now being diagnosed more frequently in adolescent patients because of increased awareness of impingement disease as a sequelae of common pediatric hip conditions, and as an etiology of hip pain in young adult persons. Patient activity level seems to be an important factor in the pathogenesis of this disorder, and athletes who participate in sports that require repetitive hip flexion and abduction, such as soccer and hockey, are at increased risk for symptomatic FAI.

Symptom onset, duration, frequency, quality and progression are elicited with a thorough history. Hip symptoms are often activity-related and insidious in nature. Any history of previous hip disease, trauma or hip surgery is important and may be the inciting event. Hip symptoms may initially be intermittent but become more frequent as labral disease and articular degeneration progress. Activities such as putting on shoes and socks, getting in and out of a car, or prolonged sitting may cause hip pain and discomfort. Groin pain is most common, but femoroacetabular impingement may also be associated with buttock or lower lumbar symptoms. Patients commonly complain of mechanical symptoms (locking, catching, and giving way) suggestive of an acetabular labral tear or articular cartilage delamination with an unstable articular flap.

Physical Examination
In early hip impingement disease, gait is usually normal, but an antalgic gait may occur with disease progression. Hip abductor strength is variably weak as indicated by a positive Trendelenburg test. Restricted range of motion is noted on exam with passive flexion often limited to less than 105°. Internal rotation in flexion is limited to 0 – 15°. These range of motion abnormalities are commonly bilateral. Many patients have passive hip flexion of only 90-100°. Assessment of internal rotation at 90° of flexion is an important aspect of the exam, and is usually limited to 0°-15°. The impingement test is positive in the majority of patients. This test is performed with the patient lying supine, the hip is gradually flexed to 90°, adducted and internally rotated. This maneuver reproduces groin pain from the shearing force at the site of chondral or labral disease. Posterior impingement of the hip is assessed with the patient in a prone position. The hip is extended and externally rotated to reproduce posterior impingement between the femoral head-neck junction and the posteroinferior acetabular rim. Posterior impingement occasionally results from development of a posteroinferior traction osteophyte secondary to progression of FAI disease. Posterior impingement may also be associated with previous hip trauma or secondary to reconstructive osteotomy of the hip, both of which can lead to residual, posterior impingement.

Radiographic Evaluation
For evaluating impingement conditions, an AP pelvis, true cross-table lateral, and supine frog-lateral or Dunn view provide comprehensive information regarding the acetabular and proximal femoral anatomy. A diagnostic AP pelvis radiograph requires neutral pelvic rotation as confirmed by the tip of the coccyx being centered on pubic symphysis, and symmetric appearing teardrops, obturator foramina and iliac wings. Pelvic inclination is standardized by measuring the vertical distance between the sacrococcygeal junction and superior border of pubic symphysis. This distance should measure 4.0 to 5.5 cm in women and 2.5 to 4.0 cm in men.

The AP pelvis radiograph is used to assess multiple parameters of structural hip anatomy. Most relevant to impingement disease are acetabular version, femoral head coverage, acetabular depth, integrity of the cartilage space and proximal femoral anatomy. Acetabular version is estimated by the relationship of the anterior and posterior lips of the acetabulum (Figure 2). A “cross-over sign” is present when the anterior rim of the acetabulum (normally more medial and horizontal) crosses over the posterior rim of the acetabulum (normally more lateral and vertical). This radiographic finding suggests excessive anterosuperior coverage due to

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acetabular retroversion. Similarly, exaggerated femoral head coverage or acetabular depth (coxa profunda or acetabular protrusio) can be consistent with a diagnosis of pincer impingement.

On the femoral side specific attention is directed to observe femoral head sphericity and the head-neck region for loss of the normal concave transition between the femoral head and neck (a pistol-grip deformity). Radiolucent fibrocystic changes (“impingement cysts”) at the femoral head-neck junction should be noted, as these lesions are associated with femoroacetabular impingement. Specifically, Leunig et al. identified head-neck junction fibrocystic changes on AP radiographs in 33% of impingement hips and speculated that impingement-induced pressure causes these lesions.

The cross-table and frog-lateral views may be used to quantify the amount of femoral head-neck offset. The head-neck junction should display a concave transition from spherical head to neck. A reduced concavity, flat transition or prominence may variably produce cam impingement. Eijer described a method of measuring offset on a cross-table lateral radiograph and defined offset less than 9 mm as potentially abnormal. The offset ratio accounts for the size of the femoral head and is determined by the ratio of the head-neck offset distance relative to the diameter of the femoral head. An offset ratio less than 0.17 is judged to be abnormal. In patients with subtle radiographic findings and questionable femoroacetabular impingement, a diagnostic hip exam with fluoroscopy can determine the presence or absence of osseous impingement through direct visualization of the articulation between the femoral head and acetabulum.

Associated hip dysplasia may also be present. The lateral center-edge angle of Wilberg and acetabular index (Tonnis angle) are measured on the AP radiographs of the pelvis. On the false profile x-ray, anterior acetabular coverage is assessed with the vertical-center-anterior angle of Lequesne and d’Sèze. Reduced offset and aspherical femoral head changes commonly coexist with acetabular dysplasia. In planning treatment of impingement and/or dysplasia, care must be taken to determine the dominant pathomechanic deformity (structural instability versus impingement).

Imaging

Additional imaging modalities may be obtained to further define femoroacetabular impingement abnormalities and to characterize associated intra-articular disease. Magnetic resonance arthrography provides detailed information regarding the anatomy of the femoral head-neck junction, presence of labral disease and integrity of the articular cartilage. It should be noted that MR arthrography can underestimate intra-articular disease. An MR arthrography diagnosed acetabular labral tear should prompt a full examination for an underlying osseous abnormality, since labral tears are commonly a manifestation of underlying structural hip disease. CT scans offer more detail of bony anatomy and may assist in analyzing acetabular version, head-neck junction offset, head sphericity, joint congruency and subchondral degenerative cystic changes. The role of CT scan analysis will likely expand to better characterize osseous impingement lesions preoperatively.

TREATMENT OPTIONS

The treatment options for FAI varied, and there remains controversy regarding optimal surgical techniques. Successful surgical intervention is most dependent upon careful patient selection, an accurate preoperative diagnosis and selection of an appropriate surgical solution. The surgical solution for a given patient may reflect the underlying disease pattern and/or the technical preferences of the treating surgeon. It must be emphasized that effective treatment and good clinical results may be accomplished with distinct surgical techniques. Potential treatment options include activity modification, non-surgical measures (nonsteroidal anti-inflammatories, physical therapy, corticosteroid injections), joint preservation surgical techniques and prosthetic replacement procedures for advanced disease.

Non-surgical Modalities

Non-operative treatment encompasses rest, activity modification, anti-inflammatory medications, physical therapy and corticosteroid injections. The success of these treatment modalities is variable and often depends on the severity of structural impingement, stage of secondary osteoarthritis, patient age and patient activity level. The length of time needed to optimize non-surgical care has not been clarified. There remains concern about the efficacy of these treatments because they do not address the underlying pathomechanics of FAI and disease progression is common.

SURGICAL OPTIONS

Goals of Surgery

Surgical treatment for FAI is dictated by the anatomic location of the offending lesion, associated intra-articular problems and the extent of joint degeneration. In advanced degenerative joint disease, prosthetic replacement surgery is the treatment of choice, and will not be emphasized in this review. In contrast, the goal of joint preservation surgery for symptomatic disease (prior to advanced osteoarthritis) is to improve the clearance of the head-neck junction within the acetabulum and to
eliminate the abnormal contact between the proximal femur and acetabular rim. The technical goals are to correct the underlying pathomechanics of the joint, and to treat the diseased elements of the acetabular labrum and articular cartilage. In our practice, we utilize a spectrum of surgical procedures to manage a corresponding wide spectrum of disease patterns. For “nonfocal” disease patterns we tend to employ open procedures that enable major osseous reconstructions of the joint. In more focal disease, we prefer less invasive procedures that adequately address the disease characteristics, yet carry the potential advantage of a less invasive intervention. The surgical options include open surgical procedures (surgical dislocation), periacetabular osteotomy, hip arthroscopy and combined limited open osteochondroplasty and complete arthroscopic techniques. An understanding of the indications, contraindications, advantages and disadvantages of these different surgical procedures is evolving, and will require continued clinical investigation.

**Open Impingement Procedures**

The published, clinical results of impingement surgery have primarily analyzed the efficacy of open surgical treatments. Most notably, Ganz et al. introduced the surgical hip dislocation as an operative procedure for the treatment of hip impingement. By protecting the obturator externus muscle, the main blood supply to the femoral head from the medial femoral circumflex artery is not disrupted. This approach enables a 360º view of the femoral head and acetabulum allowing the surgeon to address structural disease in a comprehensive fashion (Figure 3). A reshaping osteochondroplasty of the femoral head-neck junction is performed to restore a more normal concave transition zone and improve femoral head-neck offset. Acetabular and femoral head chondromalacia may be addressed with chondroplasty, drilling or microfracture to stimulate a fibrocartilaginous response. Excessive acetabular coverage can be reduced by acetabular “rim trimming.” Extra-articular impingement of the femoral neck can be corrected by relative neck lengthening (Figure 3), and trochanteric advancement can be achieved at the time of trochanteric fixation. Acetabular labral tears may be debrided or reattached if healthy tissue remains. Since the acetabular labrum is thought to have an important role in load-bearing and hip joint stabilization, preservation of healthy labral tissue may improve the overall integrity of the hip joint and prevent or delay the onset of hip osteoarthritis. In contrast, aggressive labral resection may alter important physiologic functions such as maintaining the sealing mechanism, enhancing joint stability and decreasing cartilage consolidation.
Early and mid-term results of surgical hip dislocation for femoroacetabular impingement have been encouraging. Beck et al. reported the initial results in 19 patients with a mean follow-up of 4.7 years. Fourteen of 19 patients had an excellent or good outcome, but five patients required subsequent total hip arthroplasty at an average of 3.1 years postoperatively. Murphy et al. reported on 23 patients (22 surgical dislocations: 6 surgical dislocations with trochanteric flip osteotomy; 14 direct lateral exposures; 2 iliofemoral exposure; and 1 combined iliofemoral and direct lateral exposure) with 2 to 12 year follow-up. While only 6 patients underwent the current standard of surgical hip dislocation with trochanteric flip osteotomy, 15 patients had significant improvement in the Merle d'Aubigne scores and did not require any further surgeries at last follow-up. Three patients with unaddressed hip instability had early surgical failure and four patients with advanced preoperative osteoarthritis had late failure requiring total hip replacement. These data suggest that surgical dislocation for impingement disease is a safe and effective procedure in appropriately selected patients, but it may fail in hips with preoperative advanced osteoarthritis, instability or dysplasia. More recently, Espinosa showed that surgical hip dislocation with acetabular labrum reattachment has superior clinical and radiographic results compared to surgical hip dislocation and labral resection. At two years, 28% of labral resection patients reported excellent results compared to 80% of labral reattachment patients. Significant improvement in overall Merle d'Aubigne scores (15% vs. 30%) and pain scores (59% vs. 73%) were reported at two years. In our practice, surgical dislocation is utilized to
address “nonfocal” or circumferential disease patterns (Figures 3 and 4). Hips with severe deformity involving both sides of the joint and in need of relative femoral neck lengthening or trochanteric advancement are managed with this very versatile surgical technique.

**Periacetabular Osteotomy**

The periacetabular osteotomy (PAO) was introduced and popularized as a reconstructive option for the management of “classic” acetabular dysplasia, which is characterized by insufficient anterolateral femoral head coverage, superolateral inclination of the acetabular joint surface and associated structural instability. The PAO reduction maneuver was subsequently modified to correct the acetabular deformity associated with acetabular retroversion. Presently, severe acetabular retroversion associated with major posterior wall deficiency is the main indication for a PAO in the treatment of impingement disease. This relatively uncommon combination of structural deformities can be encountered as a primary problem or secondary to overcorrection with acetabular reorientation (Figure 2). The anterolateral overcoverage by the acetabulum is corrected with internal rotation and flexion of the acetabular fragment. Siebenrock et al.
have reported the clinical outcomes of this osteotomy in 29 patients with symptomatic FAI due to acetabular retroversion. Twenty-six of 29 patients had a good or excellent outcome at an average follow-up of 30 months. Hip flexion, adduction and internal rotation improved significantly. They concluded that when correcting anterolateral overcoverage, the acetabulum should be repositioned to achieve a residual lateral center-edge angle of 20-25 degrees. Importantly, posterior impingement may occur from overcorrection or application of this technique to an acetabulum without posterior wall deficiency. This surgical technique is utilized in our practice on a very selective basis, and is combined with an anterior arthroscopy to assess and treat the acetabular labrum and to perform a osteochondroplasty of the anterolateral femoral head-neck junction if necessary.

**Hip Arthroscopy and Combined Limited Open Osteochondroplasty**

For “focal” impingement disease, less invasive procedures are attractive because they provide the opportunity to address the disease elements effectively without a large open procedure. Nevertheless, it must be acknowledged that the published clinical results of less invasive procedures are limited, and there exists a need for early and midterm results associated with these procedures. One alternative technique combines hip arthroscopy with a limited open anterior osteochondroplasty of the head-neck junction or acetabulum (Figure 5). Hip arthroscopy is performed first to treat associated acetabular labral disease and articular cartilage lesions. A limited anterior incision (8-12 cm) is then made and the Smith-Peterson interval is utilized to access the anterior hip joint, anterolateral head-neck junction and acetabular rim. As in surgical hip dislocations, an anterolateral femoral head-neck osteochondroplasty is performed to recreate femoral head-neck offset. Rotation of the extremity allows access to an approximate 200 degree arc of the anterior and lateral head-neck junction. Fluoroscopic radiographs and dynamic motion examination confirm the location and adequacy of the resection. The clinical outcomes of hip arthroscopy and limited open osteochondroplasty are limited, yet we have recently reported on the early results of our learning curve experience with this technique. At an average 1.5 year follow-up, we demonstrated good to excellent clinical results in 23 of the 24 patients. Radiographically, femoral head-neck offset was significantly improved from preoperative measurements. There were no major complications. These data suggest that at early follow-up this technique can provide adequate deformity correction and clinical improvement in the majority of patients with “focal” disease patterns. We presently prefer this technique in the management of cam impingement disease, as the arthroscopic component of the procedure addresses intra-articular problems and the limited open component provides efficient and accurate recontouring of the head-neck junction.

**Hip Arthroscopy**

The techniques for arthroscopic treatment for FAI continue to evolve, yet the clinical outcome reports of these techniques are scarce. Arthroscopy allows access to both the central (labrum and all structures located centrally) and peripheral (all structures peripheral to the labrum but within the joint capsule) compartments for treatment of labral and articular cartilage damage and femoral head-neck recontouring. First, acetabular labral disease and articular cartilage problems are addressed. The labrum may be treated with partial resection or repair, while articular cartilage disease may be treated with chondroplasty or microfracture. The femoral head-neck offset is examined and recontoured by shaving off the head-neck osteocartilagenous prominence. Additionally, arthroscopic treatment of pincer impingement has been described. The excessive acetabular coverage is reduced by “rim trimming” with a motorized burr and/or an arthroscopic osteotome. After resection of the acetabular rim, reattachment of the labrum can be performed. Guanche and Bare have reported 16 month clinical results on ten consecutive patients treated arthroscopically for cam impingement disorders. At last follow-up, average nonarthritic hip scores improved from 75 to 95 points. Eight patients without intra-articular cartilage degeneration did substantially better than the 2 patients with articular cartilage degeneration. These early clinical results suggest that in carefully screened patients, hip arthroscopy may be a valuable surgical option to treat FAI. This technique is most suitable for focal disease like isolated cam impingement, but the spectrum of impingement disease treated arthroscopically continues to expand.

**Complications**

Complications of surgical treatment for FAI vary depending upon the specific surgical technique. All procedures carry a small risk of infection, thromboembolic disease, heterotopic ossification and neurovascular damage. Anterior approaches can be associated with lateral femoral cutaneous nerve injury, while transtrochanteric approaches have a small risk of trochanteric nonunion. Osteonecrosis of the femoral head is a theoretical complication of surgical dislocation, but is extremely uncommon with sound surgical technique. The lateral retinacular vessels are branches from the medial femoral circumflex artery and are thought to provide the major
blood supply to the superior femoral head. These vessels perforate the hip joint capsule and run along the postero-lateral femoral neck.\textsuperscript{40} Caution should be exercised when performing osteoplasty (open or arthroscopic) to avoid damaging these vessels and potentially causing femoral head osteonecrosis. Femoral neck fractures associated with head-neck osteochondroplasty are quite uncommon, but can occur.\textsuperscript{51} In a cadaveric study, Mardones et al showed that the risk of femoral neck fracture is lower if less than 30\% of the neck diameter is resected from the anterolateral quadrant.\textsuperscript{50} Preoperative assessment of femoral head-neck diameter, intraoperative fluoroscopic assessment and surgical experience provide guidance for a safe bone resection. Appropriate resection depth and protected weight bearing postoperatively seem to minimize the risk of femoral neck fracture. It is also critical to realize that the degree of secondary osteoarthritis clearly effects the prognosis of all impingement procedures. The presence of extensive full-thickness articular damage is a poor prognostic factor, and the patient should be counseled regarding the risk of early failure due to progression of the underlying arthritic disease.

CONCLUSION

Femoroacetabular impingement has been recognized as an underlying cause of hip pain and secondary osteoarthritis. While isolated cam or pincer impingement occurs, a combination of both mechanisms is common. Multiple disease etiologies have been proposed, and many of these present clinically in the adolescent and young adult patients. Hip pain and restricted hip motion are common initial findings. Several surgical options are available for treatment, and are guided by the severity and location of the disease, as well as the expertise and technical preferences of the surgeon. Accurate diagnosis of the impingement disease pattern and precise surgical technique are the basis of successful surgical care. In general, early outcomes of surgical treatment for impingement are favorable, and suggest that these interventions help maintain function and preserve the natural hip over time. Clearly, long-term outcomes are needed to fully understand the impact of surgical intervention on disease progression associated with FAI.

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ABSTRACT
Following correction with the Ponseti method some idiopathic clubfeet still will relapse even after six years of age. A better understanding of the cause for these late relapses will greatly help in the management of this condition. We evaluated a consecutive case-series from 1948 through December 1984 including 209 patients (321 clubfeet). Patients were treated following the Ponseti method. Initial number of casts, age at relapse, neurological evaluation, and final treatment for the late-relapses were recorded. There were 12 patients (6%) having a relapse after the seventh birthday. In 4 of these patients (6 clubfeet) a neuromuscular disease was diagnosed, representing 33% of the late relapses. These patients were initially treated with an average of 4 casts (range: 2-6) with 2 requiring an Achilles tenotomy. Patients used the brace for an average of 4 years. The average age at the relapse prior to the suspicion of neuromuscular disease was 9 years (range: 8-11 years). Two patients had family history of neuromuscular disease (myotonic dystrophy and multiple core disease). In the other two cases (Charcot-Marie-Tooth Disease type IA and myasthenia gravis) neuromuscular disease was not suspected. All four patients required an anterior tibialis transfer, three had a plantar fasciotomy, and two had peroneus longus to brevis transfers. One patient required a subsequent posterior tibialis transfer and another patient a triple arthrodesis (myotonic dystrophy). In conclusion, late relapses in patients with idiopathic clubfoot may represent the onset of a previously undiagnosed neuromuscular disease, and should be thoroughly evaluated.

NEUROMUSCULAR DISEASE AS THE CAUSE OF LATE CLUBFOOT RELAPSES: REPORT OF 4 CASES
Matthew E. Lovell, BS, and Jose A. Morcuende, MD, PhD

INTRODUCTION
A small number of clubfeet treated at our institution relapsed after six years of age following correction with the Ponseti method. Interestingly, several neuromuscular diseases may imitate a clubfoot relapse by causing cavus and equinus deformities. A late relapse may represent the onset of a previously undiagnosed neuromuscular disease as suggested by the following four cases.

METHODS
Consecutive case-series from 1948 through December 1984 including 209 patients (321 clubfeet). Patients were all treated following the Ponseti method. Of these patients there were 13 patients (19 feet) noted to have a clubfoot relapse after the age of six years. Four of these patients (6 clubfeet) were diagnosed with a neuromuscular disease after their clubfoot relapse. No other cases have had a neuromuscular disease diagnosis. Initial number of casts, age at relapse, neurological evaluation, and final treatment for the relapse were recorded (Table 1).

CASE 1
Patient presented as a 3.5 month old white female, product of normal delivery. Pregnancy was uneventful except for a cold during 2-3 month. Baby was otherwise normal at birth except for a right clubfoot deformity. Subsequently, the child was found to have a ventricular septal defect at 3 months of age. Neurologic and neuromuscular exams were normal at this time. Clubfoot casting at an outside institution was started shortly after birth and involved 7 casts before referral to our institution at the age of 3.6 months. She was treated with 4 casts and a tenotomy resulting in 10 degrees of dorsiflexion. The brace was consistently used until the age of six and a half years. A year after discontinuing the brace she had her first recurrence consisting of a tight heelcord and mild supination during gait which was treated with a plantar/Achilles lengthening and an anterior tibialis tendon transfer to the 3rd cuneiform. Four months later it was felt that she had an excellent result, with the exception of tight heelcord. A year later, she had begun to slump forward, was unable to sit with her legs outstretched due to tight hamstrings, and developed a progressive equinus deformity on the right foot. She has a history of chronic constipation, occasionally soiling herself, but
had no urinary incontinence. She had a rectal sphincter with 50% normal tone. Motor strength was symmetrical at 4+. There were no muscle fasciculations or wasting. No sensory deficit was observed. Deep tendon reflexes demonstrated: biceps 0, triceps 2+, brachioradialis 2+, patellar 0, ankle 2+, with no clonus. She was diagnosed with Dejerine-Sottas Disease (Hereditary Hypertrophic Polyneuritis / Charcot-Marie-Tooth Disease Type IA).

At the age of 11 years she had a right Achilles tendon lengthening with a percutaneous plantar fasciotomy for right equinocavus foot. At 24 years of age she still had 5 degrees of equinus, weakness of her dorsiflexors, and marked clawing of all toes. She underwent a left total hip replacement at the age of 26 due to early onset degenerative joint disease of unknown etiology.

CASE 2

Patient with a unilateral left clubfoot and a positive family history of clubfoot was initially treated elsewhere with casts and a tenotomy followed later by an Achilles tendon lengthening at the age of two. He presented to our institution at nearly five years of age for clubfoot deformity. He required two casts to obtain correction and had a left anterior tibialis transfer to the 3rd cuneiform, and the foot abduction brace was started. He wore the brace faithfully, but had a recurrence at the age of 6, involving a varus foot, which was corrected with a left posterior tibialis slide. A year later he had a left plantar fasciotomy, peroneus longus to brevis transfer, transfer of the large toe extensor to the metatarsal neck for pes cavus deformity. Nine months after this surgery he was diagnosed with the same type muscular dystrophy as his mother. On examination of strength, he has grade 3/5 anterior tibialis, 2/5 peronei, 4/5 extensor digitorum communis, and 0/5 extensor hallucis longus activity.

He had marked, fixed heel varus. At the age of 8.5 years he underwent posterior tibialis transfer through the interosseous membrane to the cuboid. At 20 years of age, it was determined he had multiple core disease by muscle biopsy. Several of his relatives had biopsy confirmed multiple core disease as well. In his last follow up at age of 21, he was working as a security guard and walking approximately 7 miles per day, without evidence of further progression of his neuromuscular disease.

CASE 3

Female patient with bilateral clubfoot initially seen at our institution at the age of 8 weeks. Clubfeet were corrected with 6 casts and an Achilles tenotomy, after which she began to use the Mitchell brace. Brace wear was not consistent and she had a relapse at the age of 4 years, which was corrected with 3 additional casts.
The brace was started again and worn until she was 8.5 years of age. One year after stopping the brace she had another relapse with forefoot supination and tight heel cords treated with left tendo Achilles lengthening and bilateral anterior tibialis tendon transfer to 3rd cuneiform. Two and a half years later she was diagnosed with myasthenia gravis. No further clubfoot treatment has been required.

CASE 4

Patient with bilateral clubfeet and a medical history significant for mitral valve prolapse, a large left parietal cephalohematoma, and mental retardation. Treatment was started at 16 weeks of age and consisted of 3 casts, followed by bracing until she was four years old. She developed a recurrence at the age of 11 years consisting of bilateral cavovarus feet without significant heel cord tightness. She was diagnosed with myotonic dystrophy by electromyography. At the age of 12 years she had surgery consisting of bilateral peroneus longus to brevis transfers, plantar fascia releases, 1st metatarsal osteotomies with recession of the extensor hallucis longus, and anterior tibialis tendon transfer to the 3rd cuneiform. At 13 years of age she had a right triple arthrodesis to correct persistent, progressive varus and cavus.

DISCUSSION

For the first few years after full correction of a clubfoot during childhood, there is a tendency for relapse. However, it is very rare to observe a relapse after age 6. Unfortunately, there are no clinical criteria that will help in the differentiation of these cases.

In this study we evaluated all patients treated under Dr. Ponseti until 1984, and we found that 13 cases (6%) had a relapse after 7 years of age. Evaluating these patients it was observed that 4 (31%) had a diagnosis of a neuromuscular disease, suggesting that this may have caused the relapse.

Initial correction of the deformity was easily accomplished in all cases with a few casts and the patients were mostly compliant with the bracing protocol. Interestingly, patients wore the brace until 4 years of age or older; a time that has usually been considered enough to prevent relapses.\(^1,2\) In 3 cases the neuromuscular disease was suspected early after the relapse and the patient was referred for a neuromuscular examination immediately. However, in the 4th case (myasthenia gravis) neuromuscular disease was not suspected until 2.5 years later. The relapse that immediately preceded the diagnosis of neuromuscular disease occurred at an average age of 9.2 years (range: 7.6 to 10.9 years). Neuromuscular disease was diagnosed an average age of 9.9 years (range: 7.6 to 12.1 years).

The neuromuscular diseases represented were myotonic dystrophy, myasthenia gravis, multiple core disease and Charcot-Marie-Tooth Type IA. Myotonic dystrophy has been associated with clubfoot deformities and Charcot-Marie-Tooth disease has been associated with foot drop and inversion.\(^3,5\) Myasthenia gravis, while classically affecting the bulbar muscles and proximal limbs, is occasionally associated with distal limb weakness and fatigability.\(^6\) Clubfoot deformity has been associated with multiple core disease, scoliosis and joint contractures.\(^7\)

In conclusion, late relapses in patients with idiopathic clubfoot may represent the onset of a previously undiagnosed neuromuscular disease, and should be thoroughly evaluated.

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UNUSUAL ASSOCIATION OF CONGENITAL KYPHOSIS AND CONUS
LIPOMA PRESENTING AS A DOUBLE SPINAL CORD TETHER

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Arnold Menezes, M.D., and Stuart L. Weinstein, M.D.

ABSTRACT

The case of a four-year-old child is described who presented to our institution with cervicothoracic deformity and a two-year history of progressive paraparesis. His past medical history was significant for meningocele which was closed at age two months. Imaging studies revealed severe congenital kyphosis with a hypoplastic T3 vertebra, as well as a tethered filum terminale with a conus lipoma. The spinal cord was found to be severely compressed at the apex of the kyphotic deformity. Discussion is focused on the diagnosis of tethered cord syndrome, and treatment options. In particular, this case required careful thought on the order of events, which followed initial tethered cord release and removal of the conus lipoma, and subsequent kyphectomy and fusion of the upper thoracic spine. A favorable clinical outcome was obtained with complete reversal of the paraparesis.

INTRODUCTION

Type I congenital kyphoses—in particular, upper thoracic—are the most deforming and have the highest neurologic risk. Rigid deformities with neurological impairment are indicated for spinal cord decompression followed by anterior and posterior fusion. Congenital malformations of the spinal axis may be associated also with occult forms of dysraphism and potentially may be a second source of spinal cord tether. The object of this paper is to report an unusual presentation of a double spinal cord tether caused by congenital kyphosis and a conus lipoma.

CASE REPORT

A four-year, ten-month-old Hispanic male child presented to us with a two-year history of unstable gait, frequent falls and diminished strength in both lower extremities, accompanied by progressive kyphotic deformity in the cervicothoracic junction. At two months of age he had been subject to surgical excision of a lumbar meningocele. His parents were unaware of any bowel or bladder dysfunction.

On physical examination the child had a midline scar in the lumbar region and a prominent cervicothoracic kyphosis. He presented with a wide-based gait and poor balance, with bilateral atrophy of the gluteal, thigh and calf muscles. Radiographs showed a thoracic kyphosis of 90° with T3 apex, thoracolumbar scoliosis and widened lumbar pedicles (Figure 1).

During the following six weeks, he became unable to stand without assistance, and developed a positive Babinski sign. Urologic evaluation revealed a hypotonic bladder with incomplete voiding. CT and MRI further demonstrated severe kyphoscoliosis of 138° with cord compression at T3 and abnormal cord signal (Figure 2), and a conus lipoma at T11-T12 with cord tethering (Figure 3).

A two-stage procedure was indicated with: (1) Tethered cord release and resection of the conus lipoma, which was found to be anchored to the dorsal dural sac (Figure 4); (2) Cord decompression at T2-T4 two weeks later, with kyphectomy and fusion of the cervicothoracic junction. The spine was exposed through a T-shaped incision and costo-transversectomy of the second through fourth ribs (Figure 5). Following subperiosteal exposure...
of T2-T5, the T2-T4 pedicles were removed, and the
intervening nerve roots were dissected and preserved.
Three hemivertebrae were excised with rongeurs and
curette back to the posterior cortex. The spinal canal
was entered at the T2-T3 disc space and bone was
removed from convex to concave, toward the apex. An
anterior rib strut was placed between T1-T5. Posterior
\textit{in situ} fusion from T1-T5 was then carried out through
the same incision, with rib autograft and morselized al-
lograft. The patient was placed in a Minerva brace for
six months until fusion was certain (Figure 6).

At the two-month follow-up visit neurological recovery
was already evident, and at six months his neurological
assessment was normal. At his last visit three-and-a-half
years after surgery, he continued to be neurologically
intact and fully active in school, including soccer and
other sports.

\section*{DISCUSSION}
Congenital kyphosis secondary to formation and/or
segmentation defects progresses, especially during
the adolescent growth spurt, and may result in severe
deformity and spinal cord compression.\footnote{16} McMaster\footnote{2} de-
scribed that 7 of 68 patients developed spontaneous neu-
rological deterioration. Kyphosis was the most prevalent
deformity (42 of 43 patients) causing neurological deficit
in the review of Lonstein et al.\footnote{22} Congenital kyphosis
is classified based on anatomical characteristics: Type
I—Anterior failure of vertebral-body formation; Type
II—Anterior failure of vertebral-body segmentation,
or; Type III—a combination of both. Type I deformi-
ties— in particular, upper thoracic deformities like the
present case—are the most deforming and have higher
neurologic risks.\footnote{23}
Successful management depends on recognizing poor prognosis at an early stage (age <5 years), and on balancing spinal growth by means of a posterior arthrodesis with a kyphosis of <45°. Anterior and posterior fusions are indicated in Type I deformities when kyphosis exceeds 50° in children >3 years old. Early treatment of the deformity is relatively straightforward and provides excellent results, whereas late treatment is difficult and the results are usually less than ideal. Once neurologic symptoms develop, treatment depends on the onset of symptoms and flexibility of the curve. If the onset of symptoms is <3 years of age and the apex is flexible, the deformity may be progressively improved with distraction followed by fusion if neurological symptoms improve satisfactorily. In rigid deformities and/or if immobilization and rest do not result in neurological recovery, then decompression of the spinal cord, followed by anterior and posterior fusions, is indicated. Decompression of the spinal cord in kyphotic deformities is risky because of the potential for anterior migration of the cord, which is significantly increased in the presence of a distal tether. For this reason, tethered cord release was chosen as the first procedure.

Congenital malformations of the spinal axis may also be associated with occult dysraphism and malformations of other systems or organs. Myelodysplasia is often associated with tethered cord syndrome (TCS), a broadly used term for progressive neurological deterioration localized to lower spinal abnormalities (fibrous bands, adhesions, thickened filum terminale, diastematomyelia, or intradural lipomas) resulting in traction of the conus medullaris.

Symptoms of TCS develop gradually as a product of disproportionate longitudinal growth between the vertebrae and the tethered cord, resulting in stretching of the conus medullaris and nerve roots. Clinical manifestations include back pain, sensory disturbance, gait deterioration, contractures of lower extremity muscles and increasing foot deformities (pes cavus, pes adduc-
Progressive scoliosis, neurogenic bowel and/or bladder, and frequent urinary tract infections (impaired bladder compliance) are also common manifestations. Hyperreflexia associated with motor dysfunction or the Babinski sign can be found in 10-17% of patients with TCS. The initial evaluation includes a thorough neurological examination and appropriate imaging. Spina bifida occulta is found in 90% of patients with tethered cord syndrome. Alternatively, few children with incidental radiologic findings of spina bifida occulta have cord tethering. An elongated spinal cord caudal to L2 and thickened filum terminale (>2 mm) are the most common findings. TCS occurs in 3-15% of patients with history of a repaired meningo(myelo)cele.

Management of patients with TCS is controversial and there are two different approaches: (1) prophylactic release of the tethered cord; or (2) observation until neurological signs develop. Reversal of upper motor neuron symptoms may be poor once neurological signs and/or orthopedic deformities are detected, and prophylactic surgery has shown better results. On the other hand, not all patients with tethered cord develop clinical symptoms, and low-lying conus does not necessarily translate into TCS. For this reason, some authors advocate close surveillance to determine the need and timing for surgical untethering. Prolonged or accentuated neuronal dysfunction may lead to structural damage to the neuronal perikarya and axons. Untethering improves the oxidative metabolism of the cord and gait has a greater chance for improvement than bladder function. In the present case, bladder function normalized, as did the gait disturbance.

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ABSTRACT

We report six children with pigmented villonodular synovitis. They ranged in age from seven to fifteen years. In four patients, the knee was involved. One patient had involvement of the ankle, and one had diffuse involvement along a metacarpal. In five cases, the diagnosis was not suspected clinically or radiographically, and the delay in making the correct diagnosis was as long as two years. Clinical diagnosis in these five patients was usually bacterial synovitis or juvenile rheumatoid arthritis. We feel that the diagnoses of pigmented villonodular synovitis should be considered in any child with chronic joint effusion.

Pigmented villonodular synovitis (PVNS) is a disease of synovial membrane characterized by a proliferation of mononuclear cells, probably of histiocytic origin, deep to the synovial lining cells. In addition to mononuclear cells, multinucleated giant cells, foam cells, and hemosiderophages are present in varying amounts. As a result of these cells, the synovial membrane, either intraarticular or extraarticular, is transformed into thickened brownish nodules and greatly elongated villi. Pigmented villonodular synovitis occurs in one of two growth patterns—a localized nodule or a diffuse villous hyperplasia of large portions of synovial membrane. The localized form, when arising in tenosynovium, is sometimes called giant cell tumor of tendon sheath.

The etiology of pigmented villonodular synovitis is uncertain. Because many of the mononuclear cells show trisomy 7, a feature which suggests clonality, a neoplastic origin is possible.¹ A neoplastic origin is further supported by the observation that some cells in this process are aneuploid.² Another observation has confirmed the histiocytic differentiation of the mononuclear cells—most contain the histiocyte markers Leu-M3 and Leu-3.³

Pigmented villonodular synovitis most commonly affects adult patients in the third of fourth decades of life. Patients present with joint pain, swelling, and stiffness. Although any joint may be affected by PVNS, the knee is the most common site, involved in 80 percent of cases. Other joints frequently involved are the hip, shoulder, and ankle. Usually only one joint is affected. Pigmented villonodular synovitis is rare in children and may present diagnostic problems. We are presenting six children with PVNS. We studied the clinical history, histologic features, and radiographic images of their lesions (Table 1).

CASE HISTORIES

Patient One

A seven-year-old girl had a swelling on the small finger of her right hand for three months. The swelling was confined to the volar aspect of the PIP and MCP joints. There was no history of an injury, and laboratory studies were normal. An open biopsy demonstrated pigmented villonodular synovitis. The lesion was excised. Over the next five years, the patient developed four recurrences of this lesion with diffuse involvement of the flexor tendon sheath. Each time the lesion was excised in its entirety. Since the last excision, the patient has not had a recurrence in two years. Despite multiple surgeries, she has full range of motion in the small finger with slight stiffness in the metacarpophalangeal joint.

Patient Two

An eight-year-old boy had a two-month history of swelling in his right knee. He was in otherwise good physical health but had shown some developmental delay of uncertain origin. An MRI of the knee showed a joint effusion and thickened synovial membrane in

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²Department of Pathology

Each author certifies that he or she has no commercial associations (e.g., consultancies, stock ownership, equity interest, patent/licensing arrangements, etc.) that might pose a conflict of interest in connection with the submitted article.

Each author certifies that his or her institution approved the reporting of this case series, that all investigations were conducted in conformity with ethical principles of research, and that informed consent was obtained.

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Pigmented Villonodular Synovitis in Children

The anterior compartment, suggestive of a synovial inflammatory process (Figure 1). Juvenile rheumatoid arthritis was suspected, but laboratory studies were normal. After following the patient clinically for a year, he underwent an arthroscopic synovectomy, with the specimen showing pigmented villonodular synovitis. Over the next two years, he required two additional arthroscopic synovectomies to completely remove the affected synovial membrane. Five years after the last surgery, there was no evidence of recurrence, and he had full range of motion.

Patient Three

An 11-year-old girl developed a sudden painful swelling of her right knee. In the emergency room, she was felt to have bacterial synovitis, so she was hospitalized and treated with IV antibiotics. Because bacterial cultures were negative, the working diagnosis was changed to juvenile rheumatoid arthritis. However, workup for this disease was negative. She was followed for a year with intermittent increases in the swelling. Plain radiographs showed normal bone structure, but an MRI showed a synovial mass in the posterior aspect of her knee joint (Figure 2). She underwent a needle biopsy, which showed pigmented villonodular synovitis. Subsequently, she had an arthroscopic synovectomy of the knee. One year post-op she is doing well with no recurrence.

### TABLE 1

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age/Sex</th>
<th>Location</th>
<th>Presentation</th>
<th>Radiology</th>
<th>Pre Op Dx</th>
<th>Treatment</th>
<th>Follow-Up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Patient 1</td>
<td>7/F</td>
<td>Right hand, fifth finger</td>
<td>Painful swelling</td>
<td>Non-specific</td>
<td>Tumor</td>
<td>Synovectomy x5</td>
<td>Four recurrences, two years disease free</td>
</tr>
<tr>
<td>Patient 2</td>
<td>8/M</td>
<td>Right knee</td>
<td>Two months pain and swelling</td>
<td>Synovial inflammatory process</td>
<td>Juvenile rheumatoid arthritis</td>
<td>Arthroscopic Synovectomy x3</td>
<td>Two recurrences, five years disease free</td>
</tr>
<tr>
<td>Patient 3</td>
<td>11/F</td>
<td>Right knee</td>
<td>Acute painful swelling</td>
<td>Synovial mass</td>
<td>Bacterial synovitis/juvenile rheumatoid arthritis</td>
<td>Arthroscopic synovectomy</td>
<td>One year disease free</td>
</tr>
<tr>
<td>Patient 4</td>
<td>11/F</td>
<td>Right knee</td>
<td>Six months swelling and pain</td>
<td>Synovial mass</td>
<td>Bacterial synovitis/juvenile rheumatoid arthritis</td>
<td>Arthroscopic synovectomy</td>
<td>Two years disease free</td>
</tr>
<tr>
<td>Patient 5</td>
<td>15/M</td>
<td>Right knee</td>
<td>One year pain and swelling</td>
<td>Pathologic synovial mass</td>
<td>Synovitis</td>
<td>Arthroscopic synovectomy</td>
<td>Three years disease free</td>
</tr>
<tr>
<td>Patient 6</td>
<td>13/M</td>
<td>Right ankle</td>
<td>Two years pain and swelling</td>
<td>PVNS</td>
<td>Rheumatologic process</td>
<td>Arthroscopic synovectomy</td>
<td>Two years disease free</td>
</tr>
</tbody>
</table>

Figure 1: A T2-weighted MRI of knee of patient two. There is an effusion and synovial thickening in the anterior portion of the joint.

Figure 2: A T2-weighted MRI of knee of patient two. There is an effusion and synovial thickening in the anterior portion of the joint.
Patient Four

An 11-year-old girl had a six-month history of a swollen and tender right knee. There was no history of trauma, and laboratory values were within normal limits. Aspiration showed no evidence of an organism, although septic synovitis was the principal diagnosis. The differential diagnosis also included juvenile rheumatoid arthritis. An MRI showed a joint effusion with synovial hyperplasia (Figure 3). The patient was then felt to have juvenile rheumatoid arthritis. The patient underwent an arthroscopic synovectomy for diagnostic purposes. During the synovectomy, the synovial membrane was notably hypertrophic throughout the entire knee. The tissue diagnosis was pigmented villonodular synovitis. She is doing well two years post-operatively and continues to be asymptomatic.

Patient Five

A 15-year-old male had a one year history of increasing right knee pain and swelling following a car accident. A mass gradually developed over the lateral aspect of the right knee. The patient had no history of an infectious illness or a rheumatologic disorder, and his laboratory values were normal. An MRI of the knee showed a poorly defined 5 cm mass in the intrapatellar fat pad and was interpreted as a “pathologic synovial process.” A diagnostic arthrotomy showed the knee to be filled with brown hyperplastic synovial membrane. Biopsy of the synovium revealed PVNS. The synovium was extensively debrided. Three years post-operatively there is no evidence of recurrence, and the patient is asymptomatic.

Patient Six

A 13-year-old boy had pain and swelling in the anterior aspect of his ankle for two years. His pediatrician suspected a rheumatologic disorder and referred the patient to an orthopaedic surgeon. Clinically there was a 3-4 centimeter mass in the anterior aspect of this ankle joint (Figure 4). An MRI was highly suggestive of pigmented villonodular synovitis. A needle biopsy demonstrated PVNS, and the patient subsequently underwent an arthroscopic synovectomy. There were osteochondral defects of the dome of the talus, and these were treated by chondroplasty. Two years post-operatively the patient is pain free, with no evidence of recurrence.

DISCUSSION

These six cases highlight the frequent delay in diagnosing PVNS in children. In fact, in only one case (patient six) was the diagnosis considered prior to surgery. Most cases were followed for at least a year with varying diagnoses, the most common being juvenile rheumatoid...
Pigmented Villonodular Synovitis in Children

arthritides or bacterial synovitis. Eventually, each patient underwent a tissue sampling, with the histologic features of the lesions diagnostic of PVNS. The histologic features of these lesions were no different than those in adults (Figure 5). The radiographic features showed a spectrum of involvement varying from diffuse synovial hyperplasia with extensive effusion to an isolated discrete intra-articular mass. In adults, bone erosion secondary to the synovial proliferation occurs in almost 50% of cases. However, in only one of these six children was there evidence of bone erosion (patient six).

There are 25 cases of unifocal pigmented villonodular synovitis in children in the literature. The youngest patient was eight months. In addition to these 25 cases, three cases of presumed PVNS were associated with hemangiomas of the skin overlying the affected joints. These cases may represent examples of vascular proliferation in synovial membrane, a process known to mimic PVNS. As in our cases, the diagnosis of pigmented villonodular synovitis was not suspected clinically in most reported cases. Pre-operative diagnosis included ganglion cysts, septic arthritis, juvenile rheumatoid arthritis, soft-tissue sarcoma, and bone neoplasms.

Adults may rarely develop pigmented villonodular synovitis in multiple joints. Children may also develop polyarticular disease. There are twelve reported cases of pigmented villonodular synovitis in children which involve several joints either synchronously or metachronously. The youngest of these patients was four years of age. Most of the children with polyarticular disease have other congenital problems suggesting a possible genetic basis for this syndrome. Skin lesions have been reported, as well as pulmonary stenosis with or without mental retardation, Noonan syndrome and fetal hydantoin syndrome. In these children, the histologic and radiographic features of the synovial lesions are similar to lesions in other children.

These six new cases confirm the findings of previously reported cases that pigmented villonodular synovitis occurs in children and that it is often not diagnosed promptly. Pigmented villonodular synovitis should be considered when a child presents with a painful swollen knee and laboratory and clinical studies do not support a diagnosis of bacterial synovitis or juvenile rheumatoid arthritis. These six cases suggest that arthroscopic synovectomy is the treatment of choice.

REFERENCES


SCOLIOSIS CAUSED BY RIB FUSION FOLLOWING THORACOTOMY FOR TRACHEOESOPHAGEAL FISTULA: CASE REPORT

Ryan P. Dunlay, Kevin B. Jones, Stuart L. Weinstein

ABSTRACT
Scoliosis as a late complication of thoracotomy has been described previously, but reports are rare. We present the case of a 22 year-old female referred for symptomatic scoliosis. Radiographs demonstrated a severe, structural, upper-thoracic scoliosis with associated right-sided rib fusions. Her medical history was noteworthy for a right posterolateral thoracotomy for repair of a tracheoesophageal fistula during infancy. Radiographs from her general surgical treatments during infancy demonstrated no congenital rib or vertebral anomalies. This report reviews her case in detail as well as the scarce literature available regarding scoliosis secondary to thoracotomy at a young age.

INTRODUCTION
Scoliosis of a severity requiring surgery is extremely rare, however scoliosis rates following neonatal surgery for tracheoesophageal fistula or esophageal atresia have been reported to range from 6% to 50%.3,4,6,7

CASE REPORT
A 22 year-old female was referred from a local orthopaedic surgeon with a diagnosis of scoliosis. Her chief complaints were progressive fatigue-type back pain and cosmetic rib deformity. She had no family history of scoliosis.

Past medical history was significant for a right-sided posterolateral thoracotomy for tracheoesophageal fistula repair shortly after birth. This procedure had been performed at the University of Iowa. Records were retrieved with the patient’s permission.

On the fifth day of life she had undergone surgery for repair of a tracheoesophageal fistula via a right posterolateral thoracotomy, through the bed of the fourth rib, which was not removed in the approach. Coverage of the fistula was accomplished by incorporating some loose areolar tissue from the mediastinum around the fistula. Her postoperative course was uncomplicated. She was discharged 11 days later. An anteroposterior chest radiograph from the neonatal hospitalization was retrieved, and demonstrated no scoliosis, no vertebral anomalies and no rib anomalies (Figure 1).

On presentation to our clinic, physical examination revealed an otherwise healthy 22 year old female with a large left thoracic prominence on Adam’s forward bend test, a 1 cm left shoulder elevation, and a well-healed right posterior thoracotomy scar. She had no neurologic deficits.

Conventional radiographs of her spine demonstrated a 50 degree left convex scoliosis from T1-T8 with the apex of the curve at T5 (Figure 2). Right-sided rib anomalies with decreased intercostal space were also noted. A computed tomography scan with three-dimensional reconstruction revealed fusions between the right third and fourth ribs and right fourth and fifth ribs posteriorly (Figure 3). Degenerative discs were also noted at T2-3, T3-4, and T4-5. There were no vertebral anomalies other than the typical slight lateral wedging of vertebral bodies, expected with severe scoliosis, and the notable absence of severe rotational vertebral deformities characteristic of idiopathic scoliosis.
Due to her curve progression documented prior to referral to our clinic, and the likelihood of further progression, we discussed surgical options and she elected to proceed. Posterior spinal fusion and segmental instrumentation from T2-L1 with iliac crest autograft was performed. The rib fusions were taken down only so far as could be safely accomplished via an extra-thoracic approach.

The patient had an uncomplicated postoperative course. Postoperative standing posteroanterior and lateral films demonstrated a significant correction of the profound levoscoliosis deformity with a Cobb angle measuring 17 degrees (Figure 4).

DISCUSSION

In 1934, Bisgard reported the development of spinal deformity following surgical treatment of various thoracic conditions—including tuberculosis, bronchiectasis, and chronic empyema. He divided what he called thoracogenic scoliosis into two separate groups—that following rib resection and that following scarring of the thoracic cavity secondary to infection, or “pleural” scoliosis.

In a more recent study of 61 children who had thoracotomies for a variety of non-infectious reasons, Westfelt et al. reported a 30% incidence of scoliosis measuring greater than 10 degrees. Only two patients (3%) had curves greater than 20 degrees. The curves were all convex toward the side of the thoracotomy except in the four patients who had undergone repairs.

In 1983, Gilsanz et al. reviewed 82 children who had undergone surgery for esophageal atresia. Fourteen of the 82 children required reoperation for breakdown in the anastomosis. Eight of these 14 went on to develop scoliosis of 20 degrees or more—all eight having had severe mediastinitis and empyema in the wake of breakdown. Further, the degree of deformity was proportional to the number of thoracotomies performed. The authors hypothesized that the scarring and rib fusion that followed infection and re-operation pulled the trunk toward the involved side. This would fit the Bisgard definition of “pleural” scoliosis. However, this would not apply to our case, as our patient had experienced no complications or re-operations.

More applicable to our patient, Durning et al. specifically studied scoliosis developing after tracheoesophageal fistula repairs. Nine of 18 patients followed 10 years or more had developed spinal curves of 10 degrees or greater. Only two of these patients had curves greater...
than 40 degrees—one a 13 year-old with a 42 degree left thoracic curve and the other a 14 year-old with a 55 degree left thoracic curve. All of the patients in their study had a rib resection. The authors estimated that most curves appeared five or more years after the thoracotomy.

In conclusion, we present a case of severe scoliosis secondary to rib fusions that occurred following neonatal tracheoesophageal fistula repair. These rib fusions acted like a congenital unsegmented bar causing severe scoliosis. Although the severe degree of deformity in this case is exceedingly rare, it nonetheless illustrates the need for scoliosis screening in patients who have undergone thoracotomies during infancy.
REFERENCES
ABSTRACT
A 46 year old male developed spontaneous acute carpal tunnel syndrome of the right wrist without any antecedent trauma. Surgical exploration revealed hemorrhage secondary to diffuse giant cell tumor of tendon sheath as the underlying cause.

INTRODUCTION
Acute carpal tunnel syndrome is a rare entity, most commonly resulting from trauma to the upper extremity. Spontaneous acute onset of symptoms has been reported in patients with hemophilia, a history of anticoagulation therapy, or other bleeding diathesis. Acute carpal tunnel syndrome due to diffuse giant cell tumor of tendon sheath (GCTTS) arising from the flexor tendons has not been reported previously in the literature, though there is one previous account of acute carpal tunnel syndrome secondary to hemorrhage from pigmented villonodular synovitis (PVNS) involving the intercarpal joints.¹

CASE REPORT
A 46 year old right hand dominant male presented to the emergency room with a one hour history of severe right hand and wrist pain. The patient had been eating lunch with his wife when he experienced the sudden onset of severe pain in his wrist. He denied any trauma to the wrist or hand. Over the next hour, he also developed paresthesias in a median nerve distribution in his hand. His pain was not relieved by ice, elevation, or 15 mg of morphine administered in the emergency room.

Of note, the patient had experienced similar symptoms, though less severe, six months prior to his presentation to the emergency room for this episode. At that time, he had moderate wrist pain with mild paresthesias, again without any associated trauma. He was evaluated by his primary care physician, who recommended ice, elevation, and anti-inflammatory medication. The patient’s symptoms resolved over the next few days, though he noted that extensive ecchymosis developed several days after the onset of symptoms.

He had a history of known right scaphoid nonunion dating back to junior high school (Figure 1). He had no history of bleeding disorder or anticoagulation therapy, and was not taking any medications at the time of presentation.

On physical exam, he had firm swelling in the area of the wrist and proximal palm. There was no ecchymosis or skin lesion. He had 5 mm two point discrimination in his ring and small fingers, with greater than 10 mm two point discrimination in his thumb, index, and long fingers. He was able to flex and extend all digits, including his thumb. He had a normal radial pulse.

An MRI was obtained to evaluate for a space occupying lesion in the wrist. The MRI revealed heterogeneous fluid surrounding the flexor tendons with associated fluid-fluid levels consistent with hemorrhage (Figure 2). At this point, the patient’s symptoms had not improved despite strict elevation, and open carpal tunnel release and flexor tenosynovectomy was performed.

An approximately 10 cm volar longitudinal incision was made along the FCR tendon sheath that extended past the distal palmar crease. Dissection was taken down to the transverse carpal ligament, which was released under direct vision. An infiltrative soft tissue hemorrhagic mass was identified and excised. The mass had encompassed all of the flexor tendons as well as the median nerve. The mass was fusiform in shape, approximately 12 cm long and 3 cm wide, and appeared to arise from the flexor tendon sheaths themselves and not the carpal bones. The mass was carefully dissected free from the underlying tissues and a flexor tenosynovectomy was performed.
performed. The wound was closed over a drain which was removed on the first postoperative day.

Pathologic examination of the specimen revealed fibroadipose tissue with a synovial lining containing abundant hemosiderin deposition. The specimen’s appearance was consistent with diffuse giant cell tumor of tendon sheath, as it contained numerous synovial clefts and relatively few giant cells (Figure 3).

The patient had immediate pain relief following surgery and required only acetaminophen for analgesia during the postoperative period. Two weeks following surgery, his wound was well healed and he had 5 mm two point discrimination in all digits, except his long finger, which had 6 mm two point discrimination. Approximately six months after his surgery, the patient reported intermittent bouts of swelling and pain in his wrist, usually following activity. Each episode resolved with rest and over the counter anti-inflammatory medication, but these symptoms interfered with his daily activities.

A repeat MRI was performed which showed a small amount of heterogeneous material at the base of the carpal tunnel. A repeat flexor tenosynovectomy was performed through the previous incision. Some hemorrhagic tissue adherent to the flexor tendons was excised. Microscopic examination of this tissue revealed chronic synovitis with mild chronic inflammation. Two months following the repeat surgery, the patient reported resolution of his previous aching and swelling symptoms, though he continued to have slightly diminished sensation in his long finger.
Acute Carpal Tunnel Syndrome Caused by Diffuse Giant Cell Tumor of Tendon Sheath: A Case Report

Figure 2. Axial T2-weighted MRI just proximal to the carpal tunnel reveals fluid-fluid levels consistent with hemorrhage. Axial T2-weighted MRI of the carpal tunnel at the level of the tunnel itself reveals a heterogeneous mass surrounding the flexor tendons.
DISCUSSION

Spontaneous onset of carpal tunnel syndrome without antecedent trauma is very unusual. Bauman et al previously reported five cases of acute carpal tunnel syndrome, all of which were associated with previous distal radius fracture or surgery. Spontaneous carpal tunnel syndrome has been reported in patients with hemophilia and von Willebrand’s disease, patients on anticoagulation therapy, and as a result of a thrombosed persistent median artery. Chidgey published the only previously reported case of acute carpal tunnel syndrome as a result of pigmented villonodular synovitis. He described an 89 year old female who developed symptoms following aspiration of her wrist.

PVNS and giant cell tumor of tendon sheath are the result of the same pathologic process and have similar histologic appearance. The term PVNS is reserved for intraarticular lesions, and is most commonly found in the knee and hip. Giant cell tumor of tendon sheath applies to extraarticular lesions, and can be localized (nodular) or diffuse. Localized giant cell tumor of tendon sheath is the second most common neoplasm identified in the hand, and 85% of cases occur in the digits. Localized GCTTS often presents as well circumscribed, firm, slow growing nodules in the hand. Diffuse GCTTS, also sometimes referred to as extraarticular PVNS, is much less common than the nodular form, and most commonly occurs around the knee. Complete surgical excision is the treatment of choice for both localized and diffuse GCTTS, though recurrence is common and occurs in 9-44% of cases. Though these lesions may be locally aggressive, they do not metastasize.

Though localized GCTTS may be suspected from a characteristic clinical history and physical exam, diagnosis of diffuse GCTTS often requires more advanced imaging and biopsy. MRI can be helpful both for identification and surgical planning. MRI often shows hypertrophied synovium, sometimes with fatty infiltration due to the presence of lipid laden macrophages. Foci of low signal intensity on both T1 and T2 weighted images suggest the presence of hemosiderin. In our case, hemorrhage resulted in fluid-fluid levels within the tissue as well.

Regardless of etiology, early recognition and decompression is the key to successful treatment of acute carpal tunnel syndrome. Review of the literature reveals that patients who underwent surgery 12 or more hours after presentation were more likely to have residual neurologic deficits. Bauman described five cases of acute carpal tunnel syndrome in five patients following trauma or surgery. Surgical decompression was undertaken 36 to 96 hours following trauma in four patients when conservative measures failed to relieve symptoms. Only one patient recovered full median nerve function. Ford treated four patients with acute carpal tunnel syndrome following trauma. One patient underwent surgical release four hours after presentation and had full recovery. Of the remaining four patients who underwent decompression four to 96 hours after presentation, only one patient recovered normal median nerve function. The remaining patients suffered dysesthesias and diminished sensation.

Even patients with an atraumatic origin of their symptoms benefit from early decompression. Rahimtoola operated within 12 hours of admission in two cases of hemophiliacs with acute carpal tunnel syndrome and both patients experienced complete resolution of their neurologic deficit. Likewise, Chidgey performed carpal tunnel release within 24 hours of admission in the patient with PVNS of the wrist, and the patient had full recovery.
REFERENCES

1. **Chidgey LK, Szabo RM, Wiese DA.** Acute carpal tunnel syndrome caused by pigmented villonodular synovitis of the wrist. *CORR* 1988; 228: 254-257.


SUBPERIOSTEAL CHONDROMYXOID FIBROMA: A REPORT OF TWO CASES

Ryan K. Takenaga, MAa, Frank J. Frassica, MDc,d, Edward F. McCarthy, MDb,c

ABSTRACT
Chondromyxoid fibroma is a rare cartilage tumor that represents less than 1% of all bone tumors. When in a long bone, it is usually an intramedullary lesion that is eccentrically located in the metaphyseal region. Chondromyxoid fibroma may also have unusual presentations. These include intracortical lesions and subperiosteal lesions. There have been 14 reported cases of intracortical chondromyxoid fibroma, but there have been only four reports of subperiosteal lesions. A subperiosteal location, therefore, is extremely rare for a chondromyxoid fibroma. We present two new cases of subperiosteal chondromyxoid fibroma. Given its rarity, chondromyxoid fibroma is often not in the differential diagnosis of a painful, subperiosteal scalloped lesion in a long bone. Other entities such as periosteal chondroma, periosteal myxoma, subperiosteal ganglion cyst, or subperiosteal osteoid osteoma are more likely to be considered. Our cases illustrate that subperiosteal chondromyxoid fibroma, although rare, should be included in the differential diagnosis of a painful, radiographically inactive lytic lesion on the surface of a long bone.

INTRODUCTION
Chondromyxoid fibroma is a rare, benign cartilage tumor characterized by incomplete cartilage differentiation. It is the least common benign tumor of cartilaginous origin and represents less than 0.5% of all bone tumors.1 It usually presents in patients during their second and third decade and has a predilection for the metaphyseal region of the distal femur and proximal tibia. Chondromyxoid fibroma may occasionally have an intracortical or subperiosteal presentation. Intracortical involvement is the more common of these presentations with 14 cases reported.2,12 By contrast, a subperiosteal location is extremely rare. Only four confirmed cases are reported (Table 1). We present 2 new cases of subperiosteal chondromyxoid fibroma.

Case 1
A 36-year-old man presented with a two year history of intermittent right leg pain without any precipitating trauma or injury. The pain was along the medial border of the distal one third of the tibia and was mainly present on palpation. He denied pain on weightbearing or night pain. When present, the pain was an eight to nine out of 10. On physical examination, there was no visible or palpable soft tissue mass.

Plain radiographs of the tibia showed a 1.0cm subperiosteal blister on the surface of the medial cortex of the distal tibia (Figure 1). There was an overlying rim of periosteal new bone. There was no obvious soft tissue mass.

TABLE 1
Summary of Cases of Subperiosteal Chondromyxoid Fibroma

<table>
<thead>
<tr>
<th>Authors</th>
<th>Year</th>
<th>Sex</th>
<th>Age</th>
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<td>Andrew et al.26</td>
<td>1982</td>
<td>M</td>
<td>33</td>
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<td>M</td>
<td>45</td>
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<td>25</td>
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<td>Estrada-Villasenor et al.22</td>
<td>2005</td>
<td>M</td>
<td>4</td>
<td>Distal tibia</td>
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<td>Current authors</td>
<td>2006</td>
<td>M</td>
<td>36</td>
<td>Distal tibia</td>
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<tr>
<td>Current authors</td>
<td>2006</td>
<td>M</td>
<td>45</td>
<td>Distal fibula</td>
</tr>
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</table>

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Each author certifies that his institution has approved the reporting of this case series, that all investigations were conducted in conformity with ethical principles of research, and that informed consent was obtained.

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d Department of Oncology
A millimeter subperiosteal lesion on the medial surface of the tibia (Figure 2). It had a heterogeneous $T_1$ signal, increased $T_2$ signal, and it enhanced with intravenous contrast.

The patient underwent excisional biopsy of the lesion. Histologic examination of the biopsy tissue showed a chondromyxoid fibroma (Figure 3). Due to the possibility of local recurrence, the patient had a completion of the excision. There has been no recurrence after one year.

Case 2

A 45-year-old man presented with a history of chronic right ankle pain that he attributed to multiple ankle sprains and trauma from playing soccer. Plain films of the ankle (Figure 4) demonstrated chronic synovitis in the ankle region, as well as a small lytic lesion in the lateral distal fibula. MRI revealed an 8mm long lesion in the lateral cortex of the distal fibula. The lesion did not extend to the marrow cavity and the overlying periosteum was elevated. $T_1$-weighted images revealed an intermediate signal, while $T_2$-weighted images showed some areas of intermediate signal and some areas of increased intensity similar to bone marrow.

The patient was thought to have an osteoid osteoma or fibrous cortical defect. During an excisional biopsy, a prominent, palpable distal fibular lesion was found. A 0.5cm soft area considered to be the nidus of an osteoid osteoma was excised. Wide curettage was then performed on the rest of the lesion. Histologic examination of the lesional tissue showed a chondromyxoid fibroma. There has been no recurrence after one year.
DISCUSSION

Jaffe and Lichtenstein gave the first account of chondromyxoid fibroma in 1948. Fifty years later Wu et al. reviewed 278 cases of chondromyxoid fibroma from the Mayo Clinic files and summarized the following general features of the lesion. It has a slight predilection for males over females (1.1:1) and its peak incidence is in the second and third decades of life. It usually is found in the long bones (46.9%), particularly the distal femur and proximal tibia, but a large number can also occur in flat bones (30.3%). Radiographically, it is usually an intraosseous lesion that is most often found in the metaphyseal region of long bones (88%). The lesion is typically purely lytic, with occasional instances of intralesional calcifications (35.3%). It is usually a well-circumscribed lesion, usually with a sclerotic rim. Cortical thinning and expansion are also commonly seen on plain films. Periosteal new bone formation, however, is rare. Histologically, the classic chondromyxoid fibroma has stellate or spindle-shaped cells arranged in lobules on a chondromyxoid background. Within the lobules, the neoplastic cells tend to be localized more toward the periphery. The lobules are separated by fibrous bands that contain blood vessels and sometimes multinucleated giant cells (56.8%).

A subperiosteal presentation is extremely rare for chondromyxoid fibromas. A small subperiosteal lesion on a long bone, therefore, is not likely to raise suspicion of a chondromyxoid fibroma. More likely, periosteal chondroma, periosteal myxoma, subperiosteal ganglion cyst, or subperiosteal osteoid osteomas are considered.

Periosteal chondroma is a benign cartilage tumor that develops under the periosteum of long or short tubular bones. The most frequently affected site is the proximal humerus. This rare tumor, which accounts for less than 1% of all chondromas, typically affects males in the second and third decades of life. Since they are frequently found at tendon insertion sites, a palpable mass and pain at the site during activity are often the presenting symptoms. Radiographically, they present as a well-circumscribed lesion on the bone surface with punctuate calcifications; cortical saucerization is also often present. Histologically, they exhibit lobules of hyaline cartilage. A feature which helps distinguishing it from a periosteal chondromyxoid fibroma which lacks mature hyaline cartilage.

Periosteal myxoma is another lesion that can present on the bone surface. Myxomas are rare connective tissue tumors. When in the long bones, they tend to affect children more frequently, and often present with rapid onset pain and swelling. Radiographically, they can present with a profuse periosteal reaction, which increases the thickness of the underlying cortex. Histologically, they are characterized by stellate cells within a loose mucoid stroma. Chondroid tissue is noticeably absent, which a key feature that distinguishes it from a periosteal chondromyxoid fibroma.

Subperiosteal ganglion cysts are coalescing cavities of jelly-like material that are thought to be the result of mucoid degeneration and cyst formation of the periosteum. Radiographically, they are surface lesions that produce external cortical scalloping and a periosteal new bone reaction. They usually present as palpable, but painless, masses. Cross-sectional imaging provides the definitive diagnosis by revealing the purely cystic nature of the lesion.

Osteoid osteoma is a benign osteoid forming tumor that usually arises in the cortex or adjacent medullary cavity of the femur or tibia. A subperiosteal origin is less likely, but not uncommon. Like the conventional version, subperiosteal osteoid osteomas also present as a localized bone pain in a young person. This pain is often dramatically relieved by aspirin. Radiographically, it is a lytic lesion on the surface of the bone that causes cortical erosion and an adjacent periosteal reaction.
is diagnostic as it reveals the nidus at the center of the lesion. Histologically, there is a varied pattern of fibrovascular stroma, reactive bone, and varying amounts of mineralization of the nidus, which tends to be smaller than those of conventional osteoid osteomas.

When confronted with a small subperiosteal lytic lesion on a long bone, orthopaedic surgeons, pathologists, and radiologists are more likely to consider periosteal chondroma, periosteal myxoma, subperiosteal ganglion cyst, or subperiosteal osteoid osteoma. However, our two cases and the four previously reported cases illustrate that chondromyxoid fibroma should also be included in the differential diagnoses of a small, scalloped surface lesion.

REFERENCES

CHRONIC EXPANDING HAEMATOMAS WITH INTERESTING PRESENTATIONS

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ABSTRACT
The unusual presentation of our two cases posed a diagnostic dilemma between a chronic expanding haematoma and soft tissue sarcoma even after all investigations and biopsy reports were complete. Salient points to differentiate between the two are discussed along with literature review.

INTRODUCTION
Chronic expanding haematomas are tumour like lesions with a vague history of trauma and has been mentioned infrequently in literatures with a variety of presentations.¹,²,³ They usually lay dormant for many months before suddenly starting to expand in a mode very similar to that of a chronic subdural haematoma. The importance of these swellings lies in its mimicry of soft tissue sarcomas. We present two such cases where the aggressive nature of the swelling and unusual presentation posed a diagnostic dilemma to the treating surgeon in spite of all investigations. Both patients underwent complete excision of the swelling with excellent results. Salient points to differentiate between the two are discussed along with literature review.

CASE REPORT
Case 1
A 61-year-old gentleman presented with a sudden increase in size of an already existing swelling over the mid third of right leg associated with throbbing pain of 4 months duration. He had noticed a mild swelling at the above mentioned site for the past 11 years following an RTA when he fractured his tibia but chose to ignore it until now. There was no recent history of trauma or any bleeding abnormalities. Clinically the swelling was tense, shiny and tender with engorged veins and diffuse margins associated with complete foot drop but involving only the deep peroneal component of the common peroneal nerve (Figure 1). X-rays showed some calcifications within the substance of the swelling and an urgent MRI scan revealed a well encapsulated mass between the peroneal muscles mechanically compressing the deep peroneal nerve (Figure 2). A trucut biopsy showed cholesterol clefts along with RBC's (Figure 3), haemosiderin pigmentation and areas of dystrophic calcification suggestive of a chronic haematoma. The patient successfully underwent enucleation of the swelling (Figures 4A, 4B) along with cutaneofascial suture to obliterate the dead space. Cut section revealed thick chocolate coloured fluid characteristic of the fluid found in an expanding chronic subdural haematoma. His foot drop started to recover within 2 weeks of surgery and had completely recovered by the 4th month of follow up.

Figure 1. Swelling with engorged veins and right sided foot drop.
Figure 2. Coronal and axial MRI sections showing the well encapsulated swelling.

Figure 3. Biopsy findings showing the characteristic picture of chronic haematoma with cholesterol clefts and RBCs.

Figures 4A, 4B. Swelling excised in toto.
tumours in haemophiliacs, and rectus sheath haematomas\textsuperscript{1,3} in that they all have the potential to expand at anytime during an individual’s life.

Various theories have been put forth to explain the cause of sudden expansion. One such theory is that a high osmotic pressure gradient is produced by the breakdown products of the haematoma, resulting in a localized inflammatory reaction.\textsuperscript{9,10,11} Factors in the coagulation cascade along with release of vasoactive substances are believed to be associated with an inflammatory reaction which may cause additional bleeding from fragile capillaries. This additional bleeding contributes to a perpetual cycle of inflammation.\textsuperscript{2,9}

Macroscopically chronic haematomas have a peripheral wall of dense fibrous tissue and a central space containing fresh and altered blood. Histologically\textsuperscript{1,3} there is an outer zone of dense collagenous tissue with deposits of hemosiderin, a middle zone of eosinophilic amorphous material, and an inner zone of granulation tissue overlain by RBCs and fibrin into which new capillaries grow. Areas of dystrophic calcification associated with reactive inflammatory changes are also seen.

In 1997, Mentzel et al.\textsuperscript{7,12} noted that the above mentioned histological features are similar to conditions previously described as calcific myonecrosis, chronic expanding haematoma, or post traumatic cyst of soft tissues. He grouped these processes together, calling them ancient haematoma. The importance of ancient haematomas lies in their clinical, and sometimes histologic mimicry of soft tissue sarcomas.

An X-ray and biochemical tests are always essential to rule out any fracture or bony mass, but MRI\textsuperscript{13,14} is the gold standard when evaluating these swellings. On MRI, chronic expanding haematomas exhibit heterogeneous signal intensity. They display high-signal central zones of fluid collection and a peripheral low-signal rim of fibrocollagenous tissue. A blood pooling study can sometimes demonstrate the “Halo Sign” of an encapsulated haematoma.\textsuperscript{15} Biopsy is the only way to rule out a malignant tumour.

Included within the differential diagnosis are myositis ossificans and tumoral calcinosis.\textsuperscript{16} Myositis does not

Case 2

A 22-year-old gentleman presented with pain and sudden increase in size of an already existing swelling over the dorsum of right foot of 3 months duration. He had a cystic swelling measuring 0.5 by 0.5 cm at the same site since 1999 following a football injury which did not bother him until now when the pain worsened and the swelling suddenly increased in size with no apparent trauma. Clinical and MRI findings (Figure 5) revealed a tender 4 by 4 cm densely cystic swelling in the subcutaneous tissue on the dorsolateral hind foot with engorged veins and well defined margins. The MRI showed a mass of high signal intensity on the T\textsubscript{1} weighted sequence with appearances consistent with haematoma and showed no post contrast enhancement. Trucut biopsy confirmed the swelling as a haematoma. The patient successfully underwent complete excision of the swelling with no recurrence at 6 months follow up.

DISCUSSION

Reid et al.\textsuperscript{1} first used the term chronic expanding haematoma for haematomas that persisted and increased in size more than a month after the initiating haemorrhage. The cause of initial haemorrhage\textsuperscript{4,5} is most commonly trauma which results in displacement of skin and subcutaneous fatty tissue from more deeply located fixed fascia with formation of blood filled cysts. Normally the haematoma gets completely absorbed but if it is large, the blood may not be completely resorbed in which case it becomes encapsulated by a fibrous wall forming a chronic swelling.

Rarely, these swellings slowly expand\textsuperscript{3,6} and can be mistaken for a soft tissue malignancy. Most expanding haematomas have been reported in the thigh,\textsuperscript{5,7} while the calf is a less common location.\textsuperscript{1,8} These swellings have marked similarities with subdural haematomas, pseudo

Figure 5. MRI images of the foot haematoma.
Chronic Expanding Haematomas with Interesting Presentations

manifest as a chronic expanding mass and histology shows a characteristic zoning phenomenon. Tumoral calcinosis is seen frequently in the vicinity of large joints and shows florid histiocytic proliferation with osteoclast like giant cells and lymphocytes on histology.

Management of chronic haematomas includes simple analgesics and anti inflammatories, aspiration, or incision and drainage. Surgical excision (including the fibrous pseudocapsule) along with cutaneofascial suture to obliterate the dead space is the treatment of choice in cases of large haematomas. Aspiration of the fluid or incomplete excision could lead to recurrence, continued growth, or a chronic draining sinus with or without infection. Both of our cases underwent complete excision with excellent results. The first patient had complete recovery from foot drop by the 4th month of follow-up, while the second case had no recurrence at the 6th month of follow up.

In summary, it is difficult to differentiate between a chronic expanding haematoma and soft tissue sarcoma based upon clinical findings alone. X-ray and biochemical tests are frequently indicated to rule out a fracture or bony mass, but MRI is the gold standard of non-invasive evaluation. A biopsy is the only way to definitively rule out malignancy, while the treatment of choice is surgical excision and cutaneofascial suture.

REFERENCES
ABSTRACT
Recurrent hemorrhagic shoulder is a rare entity that is scarcely addressed in the literature. We describe a case of recurrent hemorrhagic shoulder and review treatment options for this unusual disorder.

INTRODUCTION
Recurrent hemorrhagic shoulder is a rare entity first described in the sixties. In some cases it is related to hemophilia or to another bleeding dyscrasia. A few cases were described in which the etiology has been massive rotator cuff tear with resultant cuff-arthropathy. McCarty in 1994 described two and reviewed five cases in whom rotator cuff tear was associated with recurrent bleeding. The authors suggested that conservative management is the first line of therapy, and if unsuccessful, then replacement arthroplasty should be considered. Recently arthroscopic cauterization of a bleeding vessel in such a case was reported. We report a case in which repeated bleeding caused massive synovitis that eroded the humeral head and led to eventual pathological fracture.

CASE REPORT
An 80-year-old male presented four times to the emergency room with recurrent spontaneous hemarthroses of the right shoulder. There was no history of trauma or excessive activity prior to the episodes. Previous medical history included hypertension and type II diabetes mellitus but was otherwise unremarkable. In particular there was no evidence of any bleeding diathesis. During admission the patient appeared to be in severe pain and tachypneic. Physical examination revealed subcutaneous ecchymoses on the lateral aspect of the arm. The range of motion of the right shoulder was greatly limited with a positive shoulder hike sign. Active abduction and flexion were nil. Passive range of motion was up to 120 degrees of abduction and flexion and 40 degrees of external rotation though causing extreme patient discomfort. The shoulder was swollen and 60 ml of bloody fluid were aspirated from the shoulder. Microbial cultures were negative and microscopy did not reveal any crystals (possibly due to numerous red blood cells). Radiographs were compatible with erosive cuff-tear arthropathy with superior humeral head migration (Figure 1A). An ultrasonographic examination of the shoulder indicated a massive rotator cuff tear with synovitis mimicking pigmented villo-nodular synovitis. Computerized tomography demonstrated gleno-humeral arthrosis with a large effusion and synovitis of the gleno-humeral space (Figures 1B, 1C). Hemoglobin level was 8.5 grams per deciliter. The patient was managed conservatively by blood transfusion and physiotherapy with some symptom resolution but without improvement of the range of motion. Four months later the patient sustained a pathological fracture through the humeral head without trauma. CT scan (Figure 2A) demonstrated a split-head four-part fracture with extensive synovitis eroding the head. The patient was treated with a cuff-tear arthropathy hemiarthroplasty (Cuff-tear arthropathy head, The Global Total Shoulder Arthroplasty System [Depuy Inc, a Johnson & Johnson Company, Warsaw, Indiana]) (Figure 2B). During operation the glenoid appeared severely eroded. The split humeral head was filled with synovial tissue that appeared hemorrhagic. Histological examination revealed hemorrhagic synovitis without evidence of crystal deposition or pigmented villo-nodular changes.

The post-operative course was uneventful. Three months after the operation the active range of motion was 60 degrees of abduction and 70 degrees of flexion (Figure 3).

DISCUSSION
Recurrent hemorrhagic shoulder is a rare entity. Since its description in 1967 only 9 cases were reported in the English-language literature, with several other cases reported in French. The underlying pathology appears to be massive rotator cuff tear with recurrent bleeding episodes. The hemorrhagic synovitis slowly
Figure 1. (A) shoulder AP radiograph demonstrating erosive cuff arthropathy with superior humeral head migration. (B, C) Computerized axial tomography (B) and coronal reconstruction (C) demonstrating gleno-humeral degenerative changes with a large effusion and synovitis (arrows on C) of the glenohumeral joint.

Figure 2. (A) CT scan showing a head-splitting four-part fracture with synovitis eroding the head. (B) A cuff-tear arthropathy hemiarthroplasty.
invades the joint structures including the biceps tendon, humeral head and glenoid. The eventual outcome appears to be massive joint disruption. A somewhat similar entity is the Milwaukee shoulder. This entity is typified by the presence of relatively large concentrations of calcium phosphate crystals that were not seen in the above presented case. However, it should be considered possible that the blood in the aspirate masked the presence of crystals. Sano suggested arthroscopic treatment of the bleeding source. However in our case this type of treatment was deemed impossible due to the severity of the joint destruction even prior to the pathological fracture. Replacing the glenoid was not feasible due to the severe erosion of the bone. Furthermore in cases of rotator-cuff deficient shoulders it has been suggested that abrasion of the glenoid allows medialization of the humeral component and better coverage of the prostheses by the remnants of the rotator cuff. Thus, we chose to implant a cuff-substituting hemiarthroplasty component that compensates for the lack of the coraco-acromial ligament. This ligament is essential during arthroplasty in rotator-cuff deficient shoulders to prevent superior subluxation of the head of the prosthesis. The range of motion achieved appears to be adequate for an elderly patient’s demands.

REFERENCES
ABSTRACT

The moment of decision to proceed with surgical intervention is charged with some of the deepest uncertainties in medicine, but has long been cloaked under the confidence asserted by the traditionally custodial surgeon. This paper reviews the history and ethical basis for informed surgical consent. Beginning with theoretical foundations and the changing ethics of medical decision making since the ancient Greeks, it then reviews how the stage was set for informed consent by technological breakthroughs that made surgical interventions tolerable and acceptably safe. Finally, the legal generation of the doctrine of informed consent is reviewed and the current state of disclosure, shared decision-making, and uncertainty explored.

INTRODUCTION

Medical and surgical therapies arise from a series of judgments. These judgments include a number of physician-intrinsic judgments such as what the leading diagnosis appears to be, what additional tests—be they additional verbal questions, specific physical examination maneuvers, imaging studies, serological tests, etc.—may confirm or refine the diagnosis, or whether a case requires input from a consulting service or colleague. However, most judgments arise through the reciprocity of the doctor-patient relationship. This relationship is plagued with complexities.

Surgical decisions are somewhat unique in the world of medicine in that part of the decision to undertake surgery usually includes a corollary decision that a patient will experience some period of time during which they will not be capable of making decisions from the effects of analgesic and sedative drugs or frank anesthesia. This adds further complexity to the surgeon-patient relationship. Also, while all medical interventions have risks and benefits, the morbidity and mortality that complicate surgical procedures and the sometimes dramatic benefits surgery can accomplish are usually so readily attributable to the surgical event itself, that the decision to proceed with surgery is easily isolated as a distinct decision.

In the period from 1957 to 1972, the concept of informed consent became legally codified. While this punctuated some of the flux of the surgeon-patient relationship, it did not simplify that relationship whatsoever. New challenges such as direct-to-patient advertising, the medical liability crisis, and the problematic relationship that many surgeons have with the implant and pharmaceutical industries can give the suggestion that this relationship is actually spiraling out of control. Nonetheless, every hour of every day surgeries are indicated and performed. In fact, the sheer volume of care yields little time to consider where we are in this sacrosanct relationship, let alone how we got here. While we may be legally documenting more, are we truly sharing the burden of decision making more with our patients?

THEORETICAL FOUNDATIONS

The technological imperative, or the decision to provide a given treatment simply because it is technologically feasible, is dangerous in surgery. From the early Aesculapian medical tradition came the doctrinal recognition that some technically feasible treatments should, indeed, not be implemented. In the myth of Aesculapius, he applied his surgical arts to a patient whom Zeus had decided was marked for death.1 When Aesculapius’ treatments were successful, Zeus was furious and struck Aesculapius down with a thunderbolt. Clearly, a finite decision to proceed with an available treatment was recognized, but in this myth the surgeon shouldered all the responsibility for this decision.

Underlying the surgeon’s heavy decision making burden is the basic assumption that a surgeon’s will is unified with a patient’s. This assumption has had varied underpinnings at various moments in history, but it has really only been questioned recently.

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The Ancients

The Hippocratic medical tradition is frequently cited as the philosophical basis for the beneficent, custodial medical care of patients. The Hippocratic Oath in its varied forms usually includes some mention of "be of benefit and do no harm." Hippocrates did not entirely deny the concept of a physician-patient relationship, but this relationship was only a utilitarian one. "The physician must be ready . . . to secure the cooperation of the patient, the attendants, and of externals." Conversation with a patient was a tool in the physicians' armamentarium to promote cooperation or compliance with treatments rendered.

Plato is even quoted as writing that "physicians employ lies for good and noble purposes." His and Hippocrates' foundations for the basic assumption that the physician's will and patient's will are unified came from the concept of philanthropia, or the physician's love of humanity, and philotechnia, the patient's love of medicine. It was felt that patients intrinsically trusted physicians as the source of relief from what ailed them. Physicians, of course, had only altruistic and noble desires and would only choose what was best for a patient. In the doctor-patient relationship, trust and obedience, which were critical to the generation of both philanthropia and philotechnia, might be compromised by full disclosure of uncertainty or poor prognosis.

Medieval Medicine

In medieval times, divine authority replaced philanthropia as the doctor's contribution to the physician-patient unity of will. Faith replaced philotechnia as the patient's contribution. The typical routine prior to surgery included the laying out of instruments, prayer with the patient, and finally the technical process of care.

This worked smoothly as the Catholic church had a significant monopoly on medicine in Europe during this period. Most physicians were monks. It was felt that faith itself was the key to healing and that the technical treatments administered were merely adjuncts to this faith. Doctor-patient conversations during the time period were intended to stimulate hope, and provide comfort and reassurance—faith, but not necessarily truth.

A medieval French surgeon, Henri de Mondeville, had much to say about what conditions promoted successful surgery:

The surgeon . . . should promise that if the patient . . . will obey the surgeon for a short time he will soon be cured and will escape all the dangers which have been pointed out to him. . . . If the patient is defiant, seldom will the result be successful.2

While this quotation does note that some "dangers" have been "pointed out" to the patient, there is no sense that any decision has been made on the patient's part. It was felt that when patients exerted will with defiance, it was truly dangerous for them. Faith was their safest focus, even if that faith was based on a promise that no surgeon could routinely keep.

Another physician, Samuel de Sorbiere, wrote a book in 1672 entitled Advice to a Young Physician Respecting the Way in Which He is to Conduct Himself in the Practice of Medicine. In one passage, Dr. de Sorbiere conjures a fictitious doctor-patient conversation that surprisingly approximates the documentation found on modern surgical consent forms in most hospitals. However, he considered this conversation a satire, reducing a young doctor's introduction to a patient to what he thought was absurdity.

I must tell you that medicine is a very imperfect science, that it is quite full of guesswork, that it scarcely understands its subject matter; nor is it familiar with the things employed to maintain it; that the more enlightened only feel their way in it groping amidst a thick gloom.3

Dr. de Sobiere concluded, not satirically, that a wise physician should always have "a few doses of nonsense to bestow." He felt honesty with patients was a danger to be avoided. What is most interesting about Dr. de Sobiere's advice to young physicians is his reason for dishonesty, which remains a powerful influence on consent discussions today: That if a physician is honest with patients, they will simply seek another physician.

Age of Enlightenment

As the age of enlightenment closed in upon the Western world, the thought that faith and obedience to physicians was paramount (with resultant divine authority of physicians) began to crumble. No longer were physicians directly associated with the church. Reason and superior knowledge became the only physician claim to authority in the physician-patient unity of will. Patients were thought to defer to superior knowledge as their contribution. Again, the assumption remained that the patient and physician wills were unified, but that assumption stood on increasingly contestable ground. Unity of will was acceptable only as long as physicians had answers that promoted benefit for patients. Unfortunately, as treatments were increasingly brought under scrutiny, it was recognized that many of them had no benefit to offer. Not surprisingly, it was at this point that courts began to get involved.

SETTING THE TECHNOLOGICAL STAGE

A number of technological advances that were part of the creation of modern surgery were also critical in
creating the opportunity, necessity, and even urgency for informed consent. Four characteristics of surgical practice prior to the mid-1800s limited both the need and possibility for informed consent. These included the fact that most surgical treatments were wholly intolerable, unsafe, and based upon poor diagnostic and prognostic information.

**Intolerable Treatments**

Most surgical interventions, even as late as the early eighteen hundreds, were intolerable except in the most dire of circumstances. There was much emphasis on the intolerability of treatments in the surgical literature. An editorial in *The Lancet* in 1836, noted the following:

> Much also depends on the steadiness, and mild, though confident manner, in which the surgeon himself sets about his painful duty, inspiring his victim with reliance on a certain tee of relief at the expense of the temporary amount of suffering.

There were major efforts to create faster surgeries to minimize the duration of extreme discomfort in patients. Surgeons’ reputations were largely based on their speed, perhaps even more than their efficacy. Such was the case with Dr. Robert Liston, a British surgeon, famous in his time for his masterfully quick amputations, but infamous in historical perspective for his high mortality rates.

Herman Melville, in his novel, *Whitejacket*, published in 1850, was keenly aware of the grotesque absurdity of some surgical scenarios of the pre-anesthetic era. In one scene, he depicts a ship surgeon invoking informed consent prior to an amputation of a lower extremity of a seaman who sustained a gunshot to the thigh. In grand display before the entire ship crew, the surgeon tries to inspire the patient with confidence:

> I would advise perfect repose of your every limb, my man. The precision of an operation is often impaired by the inconsiderate restlessness of the patient. It is better to live with three limbs then to die with four.

Herman Melville next describes a lengthy lecture to the crew about the gory details of the surgery that is about to take place. In the process, the patient faints, in a sense, proving Melville’s thought that what awaited him was too intolerable for a conscious discussion of it ahead of time. After the amputation is performed, the patient is taken to a back room. The surgeon continues to lecture, perhaps emphasizing that the consent discussion was more with the crew than the patient himself. After a few minutes, attendants announce that the patient has died, despite the amputation.

As Melville illustrates, intolerable treatments made the possibility of informed consent almost laughable.

Only patients in the worst state could even consider undergoing an operation, and then likely could not tolerate discussion of its details ahead of time.

In 1846, this situation began to change as ether anesthetics were first tested in the United States. Ether brought a broad new tolerability to surgical intervention. Dr. Robert Liston, a British surgeon who tried ether anesthetics a little over a year after their use in the United States commented, “this Yankee dodge beats mesmerism hollow.” His comments are not far off. Certainly ether anesthetics made treatments much more tolerable than any form of hypnotism, mesmerism, or even drunkenness. However, the use of ether anesthetics also created a situation in which a patient was wholly unable to communicate or exert any will after the initiation of treatment. This brought into close focus the necessity for pre-operative unity of will between patient and surgeon.

**Unsafe Treatments**

In addition to being intolerable, most surgical interventions without anesthesia were so unsafe that their application was limited to clearly moribund patients. It is recorded that Dr. Robert Liston’s most notorious surgery yielded 300% mortality. The patient, the surgical assistant, and a family member bystander, each of whom felt the blade of Dr. Liston’s slashing amputation knife, died of gangrene in the days following.

The safety of surgical intervention had its greatest improvement through the efforts of Joseph Lister. In 1867 he used carbolic acid asepsis to surgically debride—rather than amputate—an open fracture in an eleven-year-old boy, James Greenlees. The case was successful and the boy’s limb was saved. Three years later, Dr. Lister published his “aseptic technique” in the *Lancet*. He noted that only six of 40 patients with open fractures died using carbolic acid-cleansed surgical debridement, compared to 16 deaths out of 35 patients treated prior to carbolic acid. Lister’s spray became widely used almost immediately.

Suddenly, surgery was not a deadly undertaking. Just a few years after Lister’s famous report, Scottish surgeon William MacEwen published a series of 1800 osteotomies for limb deformity corrections in children. No deaths occurred in the series. With asepsis, surgeons could address problems of function and deformity, rather than only life-threatening conditions.

In 1894, Jules Emile Pean even performed a semi-elective arthroplasty. He replaced the proximal humerus with a metallic and rubber endoprosthesis after tuberculosis had destroyed a man’s shoulder. While no long-term follow-up of the arthroplasty was ever reported, the mere performance of the case and subsequent short-
term survival were proof that the era of elective surgery had arrived.

With the advent of elective surgery came an increased recognition that in some fashion a decision was made prior to proceeding, even if surgeons were yet primarily making these decisions alone during this period. Patients were deciding to seek surgical treatment, at least.

**Diagnostic Confidence**

Every surgeon can remember obtaining consent from a patient early in training and explaining—often from poor knowledge of what the surgical plan actual was—that details would have to be sorted out after the surgeons “get in there,” but such stark uncertainty was the rule, not the exception, early in surgical history. For other than open fractures or frankly draining wounds, diagnosis was frequently not known prior to embarking upon surgery. It is not surprising that most early orthopaedic operations corrected deformities which were obviously visible from outside the body. Even if the source of the deformity may not have been known, there was an identifiable goal for intervention.

In 1895, Wilhelm Konrad Roentgen began to report experimentation with radiographs. Like ether anesthetics and aseptic technique, roentgenograms became popular very quickly. They provided a method by which diagnosis might be known prior to surgery. This not only created the opportunity to explain to a patient what the diagnosis and interventional plan might be prior to an operation, but it also provided the opportunity to follow surgeries that had been performed.

It is interesting that not three months had passed from the initial report of the invention of radiographs before editorials began to note medicolegal concerns with the use of x-rays. An editorial in the *British Journal of Photography* stated “the surgeons’ new ally may become a tacit witness against them.” Thus, while pre-operative conversations with patients gained additional substance with radiographic diagnostic methods available, post-operative conversations began to be informed as well.

Surgery, especially orthopaedic surgery, became slightly less mystical with radiographs. There was somewhat less uncertainty for surgeons to hide; there was also less uncertainty to hide behind.

**Prognostic Confidence**

A final technological advance that was required prior to any useful informed consent conversation could take place was the awareness of the outcomes of treatment. The importance of outcomes was a new concept to many physicians and surgeons in the early twentieth century.

An Orthopaedic surgeon, Ernest Amory Codman, was a pioneer of what he called the end-result concept. However, the extremely forward-thinking nature of his concept was most clearly evidenced by the fact that it was not supported by his fellow Harvard faculty. Unswayed, he decided to start a hospital on the other side of Boston based on the end-result concept and follow-up of patients after treatments, including surgeries. At a medical meeting shortly after his move, he went so far as to poke fun at his colleagues remaining at the Massachusetts General Hospital, much to his societal discredit. He depicted the rich patients of Boston’s Back Bay, who supported the Harvard hospitals, as an ostrich with its head stuck in the sand. The resultant public outrage at Codman’s suggestion that the lack of end results might be the source of such a blind devotion to the Harvard hospitals certainly is not so difficult to understand. Not all patients want to know the true results of the treatments that they seek. Some simply want treatment and want to trust the deliverers of that treatment. Very few, if any, want to admit that such trust is necessarily blind.

Codman’s perseverance eventually paid off, with others (including his former colleagues at Massachusetts General Hospital) putting results-based surgery into practice. Outcomes became the language of comparison within the medical community. However, in the adoption of the concept, the public transparency of end results that Codman initially intended was lost until only very recently.

Whether publicized or discussed privately within the medical community, the drive to study outcomes greatly increased the knowledge of what patients can expect in the course of surgical treatment. Not only could risks be better predicted, but benefits could as well. Such prognostic information is critical not only to an informative consent conversation, but to the decision-making process itself.

**LEGAL ESTABLISHMENT OF INFORMED CONSENT**

With tolerable treatments, sufficiently safe treatments, planable treatments, and at least partly predictable treatments, elective surgery flourished. With it came an increased legal focus on the decisions made to get a patient into the surgical suite.

With the age of enlightenment, when modern medical treatment came under the scrutiny of reason, there was opportunity for more of a discussion between the patient and surgeon. No longer was the blind faith of the medieval patient expected. As these conversations became increasingly frequent, not only patients, but the courts began to question the unity of will between patient and surgeon. While this did not begin with a questioning of a surgeon’s autonomy in treatment selection, it eventually arrived there.
Duty to Inform

In 1767 in England, a patient, Slater, sued his surgeons, Baker and Staplton, for intentional refracture of his healing tibia rather than the agreed-upon dressing change.\textsuperscript{2} Apparently, the two surgeons applied an iron device to the leg hoping to remove a protuberance from the union site by realignment. The details of how they managed to restrain the patient in this pre-anesthetic, delayed-fracture manipulation are not clear. In fact, the court did not rule that the surgeons should have obtained the willingness of the patient at all. The court did rule that there is a “requirement to inform” the patient of an intended treatment.

With language hearkening back to the philotechnia of the ancients, the faith of the medieval patients, and the deference to superior intellect and knowledge in the age of enlightenment, the court felt that information given to the patient was critical so that the patient “may take courage, and put himself in such a situation as to enable him to undergo the operation.” Thus, even at the legal birth of consent, the justification had everything to do with more effective custodial and beneficent care on the part of the surgeon, rather than the patient’s right to autonomy. Nonetheless, it was codified that the patient’s will and the physician’s will may not be unified, and that this should at least be noted prior to proceeding.

Right to Self-Determination

In 1914, Schloendorff sued the society of New York Hospitals after a uterine fibroid was surgically removed rather than a manual examination performed under anesthesia.\textsuperscript{3} The court convicted the surgeon of assault. What was especially astonishing from this case was that a conversation had taken place between the patient and the surgeon prior to anesthetic; the patient expressed clear determination that no surgery was to be performed beyond the simple examination. The court’s ruling of assault became the first documentation of the overriding power of the patient’s will over the surgeon’s will in a medical treatment. The judge concluded that “every human being . . . has a right to determine what shall be done with his own body.”\textsuperscript{3}

Not only were patient and physician wills not unified, but priority was for the first time granted to the will of the patient.

Combining Knowledge and Consent

The first mention of both knowledge and consent in a single case happened in an orthopaedics-related case in 1935.\textsuperscript{2} A sixty-year-old man, Mr. Fortner, developed syphilitic distal femur osteomyelitis. His treating surgeon, Dr. Koch, felt that the patient had developed a large sarcoma in his distal femur. Oncologic treatments failed miserably. The diagnosis and treatments had been inappropriate. While this court ruling did discuss the importance of both knowledge and consent to treatment, the case was really a question of the standard of care. The ruling failed to combine knowledge and consent with any right to autonomy on the patient’s part.

The patient’s right to information and necessary subsequent consent to a procedure was not technically ruled until 1957. In one case, consent for research and consent for surgical therapeutic interventions finally met. Martin Salgo won his case against the Leland Stanford, Jr. University Board of Trustees after he was left paralyzed following experimental translumbar aortography.\textsuperscript{2} The court ruled that the duty to disclose “any facts which are necessary to form the basis of an intelligent consent by the patient to a proposed treatment” was evident.

The next twenty years brought further refinements to the legal definition of the informed consent conversation. Another orthopaedic case during this period continues to receive a lot of attention in the informed consent literature. In 1972, Canterbury versus Spence was decided in favor of the patient.\textsuperscript{2} What is most intriguing about this ruling is that it was regarding paralysis that developed after a patient fell out of a hospital bed the day after a laminectomy. The patient had been neurologically intact after the laminectomy and prior to the fall. Nonetheless, in the discussions that followed it was noted that paralysis is a potential outcome of a laminectomy that was not specifically discussed with the patient. The court ruled for the necessity of risk disclosure. Their statement included the comment, “the context in which the duty of risk disclosure arises is invariable on the occasion for decision as to whether a particular treatment procedure is to be undertaken.” This case is noted as one of the first times that a patient’s right to decide for or against a treatment or procedure is truly codified. Serendipitously, it may have flowed from what was no more than a freak accident, rather than a complication clearly related to surgical morbidity.

THE FUTURE OF UNCERTAINTY

While the necessity of an informed consent conversation is now legally clear and the absence of its documentation legally indefensible, the real doctor-patient conversation is only getting more complex. Consent for an operation may no longer be the issue at hand. It is clear, at least, that the ancient assumptions of unified patient and physician will are no longer accepted by the public. We are in an era of increasing desire for self-determination. This is closely connected with an increasing demand for surgical interventions that may or may not be of benefit to patients. Surgeons, especially, find themselves torn between roles as beneficent
custodians of a specific knowledge base and the unique providers of highly sought after services. Are surgeons primarily service providers, or first advisors and then service providers. How much physician judgment can and should be applied to cases in which patients want something done?

It is arguable that one judgment surgeons have made poorly is in the communication both to patients individually and to society as a whole of the uncertainties involved in medical decisions. It remains psychologically and philosophically difficult to hedge surgical decision-making with the acknowledgment that uncertainty, even if slight, always remains in diagnosis, in the selection of treatment plans, and in the outcomes to be expected. Nonetheless, the reluctance of surgeons to clearly communicate the uncertainties they silently shoulder complicates the surgeon-patient relationship more now than ever.

First, the failure of surgeons to adequately communicate uncertainty to patients has brought much focus from society on the potential biases surgeons may have in decisions they make. The lack of transparency in the judgments made within these uncertainties has fueled distrust of the decisions made. In March 2005, The United States Department of Justice began investigating orthopaedic surgeons’ relationships with implant companies. If the federal government has decided to involve itself, it is clear that the public is no longer necessarily assuming that surgeons have only the patients’ best interests in mind.

Second, implant and pharmaceutical companies have begun advertising to patients directly, usually with the notation that patients should ask their doctors about the implant or drug. The opacity of the physician-patient relationship has prompted a desire for others to influence it from the patient’s side. Even the press and the politicians in Washington could not remove themselves from a recent family’s individual debate about a physician-family discussion concerning the right to determine a loved one’s fate after cessation of demonstrable brain function. If physicians and surgeons could better train themselves to communicate the uncertainty in medical decision-making, perhaps the public would not be so angry and surprised when such uncertainties are blatantly paraded in the media.

The most tragic result of the prior failure to communicate uncertainty is that the ability to diminish uncertainty in the future is increasingly constricted. One of the principle mechanisms by which uncertainty decreases in medical and surgical diagnosis and treatment is randomized controlled trials. Surgeons have found the concept of consenting patients for randomized controlled trials nearly intolerable. Surgeons may correctly feel that patients expect them to have a confident surgical plan. This expectation, however, is based on a long tradition of communicating confidence even in the frank absence of fact-based certainty. While surgeons remain comfortable in effectively randomizing patients by their own decisions (despite the lack of hard evidence for the varied treatments between which they choose) they are often unwilling to communicate the uncertainty necessary for a patient to understand and agree to be (ostensibly) randomized.

The crisis of uncommunicated uncertainty is quickly coming to the public eye. The Institute of Medicine’s report on errors in medicine re-punctuated the fact, long-known among physicians and surgeons, that errors in judgment are real and frequent. Surgeons’ sluggish entry into the discussion may have left the courts, the politicians, and the media with an open invitation to criticize.

Perhaps the key to regaining the trust of patients and the public will come from honesty about what we can and cannot accomplish surgically and about what we do and do not know. Patients want to be involved in decisions about their care, but judgment about how they are informed and what they are/are not told during the surgeon-patient conversation will always rest on the shoulders of surgeons. Increased transparency is now demanded by the public. Hopefully, as orthopaedic surgeons we will think more about how we will answer these demands, rather than having our answers written for us by external sociopolitical forces.

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FUNCTIONAL CAPACITY EVALUATION & DISABILITY

Joseph J. Chen, MD

ABSTRACT
Function, Impairment, and Disability are words in which many physicians have little interest. Most physicians are trained to deal with structure and physiology and not function and disability. The purpose of this article is to address some of the common questions that many physicians have with the use of functional capacity evaluation and disability and also to provide a unifying model that can explain the medical and societal variables in predicting disability. We will first define the functional capacity evaluation (FCE) and explore the different types available as well as their uses. We will review several studies exploring the validity and reliability of the FCE on healthy and chronic pain patients. We will examine the few studies that look into whether an FCE is predictive of return to work and whether an FCE is predictive of disability. In the second half of this article, we will focus on the Assessment of Disability from the origins of the United States Social Security Administration to a bold new concept, the World Health Organization’s International Classification of Function, Disability and Health.

FUNCTIONAL CAPACITY EVALUATION: WHAT IS IT?
In order for us to assess function, ideally we would like an instrument that can reliably measure the functional physical ability of a person to perform a work-related series of tasks. Terms used to judge reliability include intra-rater reliability, test-retest reliability, and inter-rater reliability. Additionally, an FCE instrument should be valid. A valid instrument should measure what it intends. Face validity implies that the test appears to measure what it intends to measure and is plausible. Content validity implies the test seems related to the construct which it is intended to measure. Concurrent validity, or criterion-related validity, implies the test is well correlated with an established “gold standard.” Although we would ideally like all tests to be measured against such a “gold standard,” much of medicine cannot be measured as such. The function of an individual is definitely not something that has a universal “gold standard.” Construct or convergent validity implies that the test is well correlated with a theoretical expectation, something researchers should be able to elucidate.

Over the past twenty years, many researchers have tried to develop functional capacity evaluation instruments. Matheson provided one of the earliest examples in 1984. Isernhagen followed in 1988 with the suggestion that a multidisciplinary team should assist in determining a person’s functional capacity. Hart in 1994 also advocated a physician and physical therapist working in conjunction to assess a patient’s resulting impairment. There are approximately 10 different types of commonly used functional capacity evaluations. These include the Blankenship, Ergos Work Simulator and Ergo-Kit variation, the Isernhagen Work System, Hanoun Medical, Physical Work Performance Evaluation (Ergoscience), WEST-EPIC, Key, Ergos, ARCON, and AssessAbility.

WHY WOULD ONE USE A FUNCTIONAL CAPACITY EVALUATION?
Functional Capacity Evaluations are used for a variety of reasons. One can use an FCE to develop a treatment program, to measure the physical abilities of patients before and after a rehabilitation program, to modify a rehabilitation treatment, to evaluate whether an injured worker can work, and to determine when he/she can return to work.

SO ARE FUNCTIONAL CAPACITY EVALUATIONS RELIABLE AND VALID?
Gottebarge and Wind et al. studied 4 of the more common functional capacity evaluation instruments and identified 12 papers which assessed either the reliability or validity of these instruments. They found that the Isernhagen Work System had consistent inter-rater reliability and predictive validity, but the intra-rater reliability was not rigorous enough for conclusion. Without a gold standard with which to compare, neither the Ergo Work System nor the Ergo Kit system demonstrated concurrent validity. There was no study found that documented the reliability and validity of the Blankenship System. The authors concluded that more rigorous studies are needed to demonstrate the reliability and validity of Functional Capacity Evaluations especially the Blankenship, Ergo Work System, and Ergo-Kit systems. They did find that the reliability of the Isernhagen Work System was good. Another study by Reneman in 2004 studied

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28 adults with the Isernhagen Work System, during a 2-day evaluation. However, a review of that article demonstrates two patients who developed acute low back pain “unrelated” to the first testing session. Therefore, two subjects returned for the second test 2-3 weeks later. However, 1 developed acute low back pain after the first session and performed only half of the items on the second session. Although this study demonstrated reliability and validity of the Isernhagen Work System, a 10% minor complication rate of the development of back pain can be troublesome when testing patients who already have back pain.

So, naturally our next question is how reliable and valid are functional capacity evaluations in patients with chronic back pain? Brouwer and Reneman investigated the test-retest reliability of the Isernhagen Work Systems Functional Capacity Evaluation in patients with chronic low back pain. They studied 30 adults with chronic back pain and asked them to complete two Functional Capacity Evaluation sessions within 2 weeks. Twenty-seven patients completed both sessions, and there was partial data for 2 patients. They defined some statistical variables as reliable and found that most of their variables were indeed reliable. However, there were 4 subtests that did not achieve their agreed upon standard of reliability.

If we have evidence that a Functional Capacity Evaluation is indeed somewhat reliable and valid, we next should focus on this instrument’s ability to predict a patient’s return to work. Gross, Battle, and Cassidy evaluated the prognostic value of functional capacity evaluations in patients with chronic low back pain in a 2-part study in 2004. They found that the floor to waist lift test was predictive of the number of tasks failed. Some other researchers found disagreements in how the authors defined their endpoint in return to work and until the claim was closed. It is common for a completed functional capacity evaluation to result in the closure of a claim, though the actual performance on the FCE may not be predictive of return to work. In essence, by having a functional capacity evaluation, a patient is likely to be put in a position of deciding whether he or she is willing to return to work. The closure of a claim often results in a proximate suspension of disability benefits. This suspension of disability benefits was observed in many studies an average of 32 days after the completion of a functional capacity evaluation. Some other flaws include that there were some unemployed patients who were tested who had no specific return to work opportunities. They concluded that the influence of psychosocial and contextual factors on return to work are significant. They recommended that further studies of return to work would preferably use cohorts of clients who have a realistic option of returning to work within the same company. In addition, they found that time off work may actually be a stronger predictor of return to work. In summary, they concluded that functional capacity evaluations are most accurately considered behavioral tests influenced by many factors, including physical ability, beliefs, and perceptions. Therefore, these results should be interpreted within the subject’s broad personal and environmental context.

So, can a functional capacity evaluation predict sustained recovery? In the second part of the Gross and Battle study, the authors tried to evaluate the prognostic value of functional capacity evaluations in patients with chronic back pain. They defined sustained recovery as no new claim of total temporary disability within the time period studied. Several researchers had trouble with this definition because some employees may have a new injury in a different part of the body with the resumption of total temporary disability benefits. They also defined sustained recovery as no new claim opened and no old claim reopened. They found that 46 of the 226 patients or 20% had recurrent low back pain following their functional capacity evaluation. Surprisingly, those who had the lower number of failed tests were actually associated with a higher risk of recurrence. Perhaps these were the patients that were physically doing well, but had some motivational or psychosocial barriers that were not adequately addressed. Apparently, the clinicians felt it was easier to get them back to work than to address those difficult issues. Therefore, the authors concluded that the ability of an FCE to identify claimants who are safe to return to work is suspect. Perhaps the FCE process and its administration are only as good as the examiner. In a follow-up of these studies, the authors concluded “performance on functional capacity evaluations is influenced by physical factors, perceptions of disability, and pain intensity. Therefore, FCEs should be considered behavioral tests influenced by multiple factors including physical ability, beliefs, and perceptions.”

So, if in fact an FCE is primarily a behavioral test without a true gold standard, we must reexamine our concepts of disability. The Social Security Administration was developed in 1954 in an effort to ensure that those individuals in our society who could not function without state or federal assistance had the ability to live within our society. Similarly, the American Medical Association was asked to provide some type of objective measurement of disability. Perhaps the oldest system of disability was the McBride system, which was based upon the workers compensation boards in 1936. The American Academy of Orthopaedic Surgeons developed a manual in 1962 to incorporate common orthopaedic injuries and define the associated disability with these
Kessler’s Disability: Determination and Evaluation was published in 1970.

So we now get back to one of our original questions—what is the difference between impairment and disability? Impairment is defined by the AMA Guides as an alteration of an individual’s health status that has been assessed by medical means. The Florida Impairment Schedule defines an impairment as an anatomic or functional abnormality or loss after MMI has been achieved. The common thread with impairment is that the problem is typically with the organ or body part. Impairments can be minor (e.g., a finger amputation), or devastating such as a cervical spinal cord injury resulting in quadriplegia. None of these definitions include the effect of the impairment onto the individual’s ability to function in society.

Disability is the term used to describe this relationship and relates to an individual’s inability to complete a task or duty. Common themes include consideration of many factors beyond impairment, beyond loss or deficit in psychological, physiological, anatomic structure. Disability encompasses vocational, educational, psychosocial, and financial factors.

SO WHY CAN WE NOT PREDICT DISABILITY BETTER?

Is it a result of an inadequate understanding of anatomy and physiology? Probably not. Our CT scanners and MRI scanners have become increasingly better at spatial resolution. Is it a result of an inadequate understanding of function? Probably not since there are some instruments that are valid and reliable in assessing our physical capacity. Instead, it is most likely due to a failure to define disability. We can all recognize a disabled child with spina bifida or cerebral palsy. There are many disabled young children who have attended mainstream schools, received excellent education, have essentially no or few activity limitations, and participate fully in society. Why does a physically adept masonry worker who had at least the same or more opportunities in society, acquire a “disabled” label at the age of 45 simply due to back pain?

Perhaps a better model exists for evaluating structure, function, activity limitations, and participation in society. Though many factors contribute to the determination of disability, the common ones include physical, cognitive, vision, and hearing. However, mental health, culture, social institutions, and physical environments can also be variables that are necessary to control for when evaluating disability.

In our medical practices, we are very structure oriented. We rarely have difficulty identifying patients who cannot see, hear, etc . . . Physicians trained to treat patients with chronic pain are ever more adept at identifying depression and making appropriate treatment referrals.

The Social Model of Disability exists upon a spectrum that defines the degree to which an impairment is disabling in relation to individual attitudes and societal structure. The social environment may actually disable the person. It may be society’s response or lack
of response that forms the basis of social disadvantage experienced by these disabled persons. This model attempts to direct rehabilitation efforts toward society to increase access to services and to include disabled people into societal activities.

However, many of our patients complain to us that they do not have access to jobs, do not have the training to go back to work, do not have the finances to retrain or the education to make a change. These are societal factors that clearly influence disability determination. While it is not uncommon to think that all patients need to pull themselves up by the bootstraps, this is an unrealistic expectation.

More recently, the World Health Organization commissioned the International Classification of Function, Disability, and Health. This new classification system is based upon a biopsychosocial model of disease of which pain physicians are very aware. It includes the body or organ systems that are affected but also adds a dimension of functioning to incorporate all bodily functions, activities, and participation. Participation restrictions are dimensions of activities that an individual is unable to perform at a level appropriate to their capacity. Clearly, one can see that a disease process affects the individual as well as the way he interacts with his environment.

Let us explore a debilitating condition like blindness within the context of the ICF model (See Figure 2). There are numerous disease processes such as macular degeneration, retinitis pigmentosa, and optic neuritis that produce obvious structural and physiologic organ system abnormalities. The impact these diseases have upon our personal ability to ambulate, dress, and groom ourselves must be examined. Participation in some life activities, like driving, is impossible and this could be rated when their performance is at full capacity. However, without Braille books or Seeing Eye dogs within their environment allowing them to participate in societal activities, the performance of the visually impaired could be significantly less than their capacity with these adaptive aids. A patient’s motivation and social support of course are integral parts of their overall level of function.

If we next imagine how children with spina bifida can fit within this model (Figure 3), we can see that they may have impairments in lower extremity strength, mobility, bowel and bladder control and skin sensation. Children’s functional abilities can be enhanced with
the using wheelchairs and other items to enable them to perform up to their best capacity. They should have few restrictions in their ability to go to school or find a reasonable workplace. However, if a particular child with spina bifida were placed solely in a position that required him to work a physically demanding occupation, his capacity might be less than his performance, thereby making him more susceptible to disability than a similar individual working as a professional engineer.

Taking the next step, we move on to using this ICF Model as was described in Wittink et al. in the *Clinical Journal of Pain* in 2004. In this condition, adults with chronic back pain may have some limitation or impairment in pain, anxiety, range of motion, strength, endurance, cognition, attention, memory, sleep, or depression. They may also have difficulty in carrying out activities of daily living including sitting, standing, walking, or using stairs. Some may be able to sit and stand during the day, others may be able to walk only 2 blocks before they are limited by their neurogenic claudication. These activities in the blue box are easily measurable (Figure 4). However, the other boxes in the diagram including Participation, Personal Factors, and External Factors are not easily measurable and can play pivotal roles in a patient's ability to function within society. These are exactly the missing links that indicate why a Functional Capacity Evaluation is not able to provide high predictive value of disability.

**ICF MODEL FOR ADULT WITH CHRONIC BACK PAIN: TRAINING FOR THE OLYMPICS**

I will conclude by adapting Wittink’s diagram and including how these other factors are essential in the overall treatment of a patient with chronic back pain. In many senses, there are several dimensions that one needs to understand to train understand a complex problem such as chronic pain. There will obviously continue to be a need for physicians trained in the proper treatment of chronic pain. There will be a need for physical therapists who are skilled in the measurement of functional abilities, although it may not need to be with a full-scale functional capacity evaluation. The missing links include what our colleagues trained in vocational rehabilitation, social work, and psychology...
can add to improve a person’s performance up to his or her capacity. The missing ring (Figure 5) includes those personal factors that are innate and are not likely to be changed with any of our interventions. While we need to respect that all people are different in their motivation and inner workings, we must make sure that we are not a part of the problem by becoming an enabler of chronic pain, confusing patients with the use of unnecessary pain medications, or further disabling them with our medical experience. By understanding these concepts we can provide our patients with excellent care and enable them to optimize their performance and capacity in fulfilling their societal roles.

Figure 4. ICF Model from Wittink.

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THE 88-HOUR FAMILY: EFFECTS OF THE 80-HOUR WORK WEEK ON MARRIAGE AND CHILDBIRTH IN A SURGICAL RESIDENCY

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ABSTRACT
The restriction of the resident physician work week to 80 hours has had dramatic affects on resident education and lifestyle. While effects on mood, psychological distress, and burn-out have been studied, the resultant changes in tangible quality of life have received little attention. Birth rate was considered a measurable, relevant outcome. The resident marital and parental status by duty month was collected from a single orthopaedic surgical residency program for the four academic years preceding and following the implementation of the 80-hour work week. The number of births to residents during these periods were also tallied. The relative prevalence of positive marital status changed very little between residents in the two time durations from 66 to 71 percent, but parental status increased from 27 to 43 percent. The number of births per married resident duty year also increased from 0.23 pre-restrictions to 0.32 post-restrictions. While the individual decisions involved in generating these observed changes are complex and difficult to entirely decipher, it is thought that an increased perception of lifestyle within the work-hour restrictions may have prompted the dramatic changes in birth rate among resident families.

INTRODUCTION
The restriction of the resident physician work week to no more than 80 hours, mandated by the Accreditation Council of Graduate Medical Education (ACGME) beginning in July 2003, was expected to dramatically affect a number of aspects of resident education. However, some of the anticipated benefits and detriments of the policy enforcement have not been clearly demonstrable. Dramatic effects on resident physician lifestyle were also anticipated from the corollary mandate for at least 88 hours of non-hospital time. These effects have yet received minimal attention in the literature.

The impetus behind the ACGME restrictions was the desire to protect patients from the medical errors that can result from sleep-deprived physician decision-making. It remains to be proven whether or not work-hour restrictions have accomplished this noble goal of decreased fatigue-related medical error.

Some of the anticipated side-effects of work-hour restrictions related to resident quality of life. It was also conjectured that work-hours restrictions during training may change the specialty and subspecialty choices made by graduating medical students, that perhaps the brutal rigor of the training programs of certain surgical disciplines may have previously deterred some candidates who will no longer avoid these specialties now that training work-hours are controlled.

Quality of life is an extremely difficult entity to measure among residents in training. One of the major challenges is the potential confounding force of a change in the complement of residency programs. If truly different people are filling the positions, they will naturally have different behaviors and different attitudes toward what an 80-hour work week means in terms of time for family, hobbies, recreation, etc.

For this pilot study, we hypothesized that the number of births per month per married resident has increased since the implementation of work-hour restrictions. While the actual time necessary to conceive a child is minimal in comparison to an 80-hour or even 120-hour work week, the decision to do so usually follows a perception that successful family life is possible within the confines of the married couple’s current schedule constraints.

METHODS
The residency records of the Department of Orthopaedics and Rehabilitation at the University of Iowa were reviewed, along with personal memory, to identify the marital status of every resident in the first four years post-work-hours-restriction, spanning from July 2003 through June 2007, and the last four years pre-work-
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hours-restrictions, spanning from July 1999 through June 2003. Common-law and formal marriages were not distinguished. Residents who spent less than a full five years in the program were included, as all numbers ultimately included the denominator of residency duty months. First through fifth post-graduate years were included.

Children born were counted for birth occurring during the pre- and post- time periods noted above. Births were anticipated for the final 5 months of the post- time period, under the assumption that pregnancies far enough along to permit birth in the next 5 months are already publicly recognized in the small and friendly community of residents. Only children born to known resident families were included. No attempt was made to measure or estimate actual conception events as these are necessarily private. Specifically, no data regarding children conceived out of wed-lock (formal or functional) were collected. Data regarding pregnancies ending in miscarriage or abortion were also not collected. Number of children born was felt to be a maximally concrete variable to measure. Children born immediately before or after the periods in question were not counted. Children born immediately before or after a resident’s months of resident duty in the Department were also not counted.

RESULTS

Twenty-four births to resident families occurred in the four years following ACGME restriction of resident work hours (Figure 1). Fifteen births occurred in the four years preceding these work hour restrictions (Figure 1). The denominator for these births changed, however, as the number of residents per class increased in the years following work hour restriction implementation. A total of 1188 months of resident duty were served during the four years preceding work hours restriction and 1260 months following. The number of births per year of resident duty increased from 0.15 to 0.22. Otherwise stated, there were 79 months of resident duty per birth prior to work hours restrictions and 53 months of duty per birth afterward.

More residents in the four years post-restrictions were married, with 892 married resident duty months following restrictions versus 781 married resident duty months in the four years pre-restrictions. The number of births per year of resident duty while married also increased from 0.23 to 0.32. This is a 40 percent increase in birth

Figure 1. The number of births to orthopaedic surgical resident families during the four years preceding and following the implementation of the 80-hour work week.
rate among married residents following the ACGME mandate. This correlates with 52 months of married resident duty per birth pre-restrictions and 37 months post-restrictions.

Residents did not appear to have children earlier or later in residency before or after the ACGME restrictions were mandated, averaging 2.73 and 2.79 years into residency at the time of birth pre- and post-restrictions, respectively (Figure 2). However, between the increased birth rate and the increased number of post-restriction residents with children born prior to the beginning of residency, the prevalence of positive parental status underwent a 60 percent increase after work-hours restrictions were mandated (Figure 3).

No resident underwent divorce during either time period studied. Two resident weddings occurred during the four pre-restriction years and five during the post-restriction years.

**DISCUSSION**

We report a 40 percent increase in births per married resident per year and a 60 percent increase in the relative prevalence of married residents with children associated with implementation of the ACGME mandate that the resident physician work week be restricted to no more than 80 hours of in-hospital duty. It is possible that this is simply due to sampling error. This study only involves four years prior to and following work hours restrictions. It only includes one residency program. Nonetheless, as a pilot study it is at least provocative that the difference may be real. A number of factors may have contributed to such an increase. One explanation is that the 80-hour work week has changed the personal attributes of residents in the program. The central challenge to this interpretation is that most of the births in the four years following work hours restriction were to residents recruited prior to the work...
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week limitations. Further, there has been no hard data to suggest that medical students are choosing specialties and programs differently since the implementation of the limited work week, although it has been widely suspected. The only hard evidence available is that voluntary attrition due to life-style concerns in general surgery programs seems not to have decreased. In our own recruitment process, some applicants have voiced that the limited work week prompted their consideration of a career in orthopaedic surgery, but no candidates vocal in this sentiment have joined our ranks.

Another interpretation is simply that residents have more time for personal pursuits, be they hunting, culinary arts, weight-lifting, or child-rearing. The difficulty with this interpretation standing alone is that 88 hours away from the hospital each week are still largely spent in educationally-related activities or sleep, and are sparse when considered against the time requirements for successful child-rearing. The previously well-quantified perception that residency is difficult for spouses and children of residents has not changed qualitatively. Certainly, spouses of residents in this residency program still regularly discuss their challenging schedule situation when together for group activities (Figure 4).

Third, residents may have an improved sense of health and well-being with the work-hour restrictions, which may promote the decision to procreate. Studies have demonstrated improved mood and reduced psychological distress among surgical residents accompanying implementation of the 80-hour work week. One contrary study noted no change in resident burn-out, specifically, however. If such a change in mood and well-being were appreciable in our program, it might explain the large changes in birth rate. In the infertility literature, a study previously demonstrated that “cognitive weariness” associated strongly with male infertility, as did construction related careers to a lesser extent. While orthopaedics has been likened to carpentry previously, the 80-hour work week has only likely affected the weariness factor from this study. It is certainly possible that increased fertility from decreased weariness among residents is partly responsible for the increased birth rate. It is not known how many sought infertility counseling or treatment prior to or following the 80-hour restrictions mandate.

Finally, the increased rate of births may be due to an increased sense of control in resident life. Residents remain very busy with clinical and education responsibilities. Resident families continue to fend for themselves without the presence of the resident for most waking hours of the week. However, the 80-hour work week has prompted vast changes in the programs for call and clinical service coverage, most of which have dramati-

Figure 3. These pie charts demonstrate the relative prevalence (in duty months) of single residents (white wedges), married residents without children (gray wedges), and married residents with children (black wedges) both before (A) and after (B) the implementation of ACGME work-hours restrictions.

Figure 3A

Figure 3B
cally increased the residents’ sense of how predictable and manageable the schedule for the typical week can be. This sense of control, even if it is control over a still too-busy schedule, must promote the settled feelings that can engender thoughts of offspring.

While this pilot study has many limitations, it highlights the possibility that implementation of the 80-hour work has improved residents’ ability to pursue non-career related life goals. Hopefully, this has been mated to an increase in resident quality of life and overall academic and professional performance.

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