







# The Philippine Journal of Orthopaedics

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eISSN 2012-3264

#### **ORIGINAL ARTICLES**

Relationship Between Surgery Timing and Hidden Blood Loss in Proximal Femoral Nailing of Pertrochanteric Fractures in Elderly Patients: A Cross-sectional Study

Four-Strand Modified Kessler versus Cruciate Technique for Repair of Zone Two Flexor Tendon Injuries: A Randomized Controlled Trial

Filipino Translation, Cross-Cultural Adaptation and Validation of the American Orthopaedic Foot and Ankle Society's (AOFAS) Ankle-Hindfoot Scale

A Comparative Study of Proximal Femoral Nail Antirotation in Peri-trochanteric Fractures in Lateral Decubitus and Supine Position

Cross-cultural Adaptation and Validation of the International Knee Documentation Committee Subjective Knee Form in Filipino

Characteristics, Treatment Patterns, and Clinical Outcomes of Patients Diagnosed with Fungating Soft Tissue Sarcomas

#### CASE REPORTS / CASE SERIES

Large Exostosis of the Distal Radius in a Pediatric Patient Causing a Dysplastic Ulna and Distal Radioulnar Joint Disruption: A Single-stage Management Approach

Spinal Arthrodesis with Acute Reduction in a High-grade Isthmic Type IIC Spondylolisthesis

Management of Long, Comminuted Pediatric Subtrochanteric Fractures Using PHILOS (Proximal Humeral Internal Locked System): A Case Series

Surgical Treatment and Short-term Outcome for a Refractory Case of Medial Clavicle Osteitis Condensans: A Case Report

A Case Series on Arthroscopic Valvulectomy of Symptomatic Popliteal Cysts in Elderly Patients Using a Modified Gillquist Maneuver

#### SYSTEMATIC REVIEW

Clinical and Radiographic Predictors of Deterioration in Mild Cervical Spondylotic Myelopathy: A Systematic Review



#### ABOUT THE LOGO: THE TREE OF ANDRY

Nicholas Andry coined the French term "orthopédie" which is derived from the Greek words "orthos" (correct or straight) and "paidion" (child). As implied in its etymology, "orthopédie" was first practiced treating childhood spinal and bone deformities.

The main elements of the logo are the tree of Andry; the Philippine Journal of Orthopaedics wordmark; and the fountain pen. The fountain pen, in replacement of the stake, represents how research has been the backbone of orthopaedic learning and practice.











The Philippine Journal of Orthopaedics, the official journal of the Philippine Orthopaedic Association, Inc. is an open-access, English language, web-based, medical science journal published by the Association. The Journal is guided by the International Committee of Medical Journal Editors (ICMJE) "Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals."

The **Philippine Journal of Orthopaedics** shall advance the art and science of orthopaedics in the country by publishing high quality original clinical investigations, epidemiological studies, case reports, review articles, evaluations of diagnostic and surgical techniques, and the latest updates on management guidelines. The journal's target audience are local and international practitioners, clinicians, and other scientists, researchers. It shall accept manuscript submissions from consultants, fellows, residents, and other allied medical professions and specialties, not only from the Philippines but also from Asia and the rest of the world as long as these are within scope and relevant to the practice. Non-members of the Association may submit scientific manuscripts to the journal.











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The Philippine Orthopaedic Association proudly presents the May 2024 issue of the Philippine Journal of Orthopaedics. We aim to come up with two publications a year thanks to the tireless effort of our editor-in-chief Dr. Tammy Dela Rosa, FPOA, the editors, the peer-reviewers, the secretariat, and the contributors.

In this fast-changing world, we are inundated with information that makes it difficult to separate hype from science. Publishing well-thought-out and peer-reviewed articles makes for a richer environment for knowledge. In this edition, we have original articles about a preferred technique for tendon repair and issues with proximal femoral nailing. We also have five case reports that show the uniqueness of our practice in the Philippines and a systematic review of cervical spine myelopathy. We hope to continue to deliver more quality papers in the future.

We acknowledge the continued support of our sponsor, Uratex Philippines, as well as the men and women of the POA. It is ultimately hoped that the scientific articles will help us, orthopaedic surgeons, improve our understanding of the complexities of musculoskeletal conditions and the way we take care of our patients.

Please enjoy this current PJO edition and continue supporting this worthwhile endeavor.

Justinian Aquilino IV Cyril LI. Pimentel, MD, FPOA 2024 Vice President, Philippine Orthopaedic Association



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#### You Give Labs a Bad Name



As an avid reader of the New York Times online edition, I browse news and feature articles multiple times a day, frequently getting to my daily Wordle, Connections, and, of course, the Times Crossword with giddy anticipation. But through the years of getting more than world events from this paper, I have read, with increasing dismay, several articles involving the falsification of data in articles published in prestigious journals. A couple of deep dives reveal other articles showcasing manipulated data, flipped pictures of purported results, and reused images in the name of publications and grants. This reality seemed so far removed from my daily life. That is until a year or so ago when, out of a mixture of ignorance and hubris, I accepted the role of editor-in-chief of this, our official journal.

Since then, whenever I see a table of figures, pictures of surgeries, or whatnot, I recall how I flipped a picture myself in desperation to meet the deadline for a photo project on the stages of mitosis and meiosis. I got away with a passing grade; but Dostoevsky earned his place in literature, not just because of his storytelling, but also because of the truths he set out to tell, this time about crime and seemingly getting away with it.

In orthopedics, retractions have become more common, probably reflecting higher vigilance and a greater number of publications. The reasons, however, should send a chill down our collective spines. The two most common reasons were fraudulent data and plagiarism – two examples of inexcusable academic misconduct.<sup>3</sup>

Research, and indeed all of science, is built on a level of trust. When information is presented as science or a result of scientific research, there is an implicit and absolute understanding that to the best of the authors'/presenters' knowledge, everything is backed up by rigor and integrity. It should be reasonable to assume that the data that someone publishes today is the highest brick of "truth" in a pyramid of "truths" and that every brick from the base up to the pinnacle is solid and unimpeachable. This may seem like an impossible task, but hey, that's science.

I bring these up today to highlight that we, the editorial board of the PJO, strictly believe in the importance of integrity. Indeed, authors and investigators bear most of the burden for these instances of misconduct. However, the board needs to keep its side of the bargain by striving to keep our journal at par with international standards; by strict peer review, vigilance when dealing with data, and using anti-plagiarism software. The integrity of our journal is a promise we make to our readers and a vow we intend to keep.

# Tammy L. Dela Rosa, MD, MMedSc Editor-in-Chief

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eISSN 2012-3264 (Online) https://doi.org/10.69472/poai.2024.01









### Relationship Between Surgery Timing and Hidden Blood Loss in Proximal Femoral Nailing of Pertrochanteric Fractures in Elderly Patients: A Cross-sectional Study\*

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#### **ABSTRACT**

**Objectives.** Pertrochanteric fractures, commonly incurred by the elderly from low-energy falls, require surgical management. Visible blood loss in the perioperative period is inconsistent with the big drop in hemoglobin seen postoperatively. This discrepancy is attributed to the hidden blood loss (HBL), which must be anticipated in anemia management. This study aimed to determine if the amount of HBL in elderly patients with pertrochanteric femur fractures treated with short proximal femoral anti-rotation nailing (PFNa) is affected by the delay in time to surgery.

Methodology. This is a detailed cross-sectional study from a single institution. Two hundred and ten patients admitted and operated on from January 2017 to December 2019 at an orthopedic specialty hospital were included in the study. Patient's age, sex, AO classification, weight, height, operative time, and hematocrit levels on admission, within 7 days before surgery and immediately postoperatively; visual blood loss and blood transfused were reviewed and retrieved from medical records. Cases were grouped into early (<30 days) or late (≥30 days) surgery groups based on the time from injury to surgery. Total blood loss and hidden blood loss were computed based on the data.

**Results.** There was no significant difference in the demographic and clinical characteristics of patients in both groups. Mean HBL was 113.65 mL ( $\pm$ 99.25 mL) in the early surgery group and 95.32 mL ( $\pm$ 111.79 mL) in the late surgery group. Mean HBLs were 31.47% and 27.88% of the total blood loss computed for the early and late surgery groups, respectively. Using an independent t-test, we noted no significant difference in the HBLs between the groups (p = 0.22).

**Conclusion.** Delay in treatment of pertrochanteric fractures fixed with short proximal femoral anti-rotation nailing did not significantly affect the amount of hidden blood loss. However, the computed hidden losses, which make up a large percentage of the total blood loss, should be considered in the postoperative management of anemia.

**Keywords.** blood loss, surgical, operative time, treatment delay, proximal femur fractures, fracture fixation, intramedullary, cross-sectional study

#### INTRODUCTION

Pertrochanteric femur fractures are a common injury in the elderly population. Due to a globally aging population, hip fractures are estimated to number around 2.56 million by the year 2050.¹ Innovations in technology accompany this upsurge in cases. Most fractures are treated using either a proximal femoral nail anti-rotation (PFNa) or a dynamic hip screw (DHS). Although there is still no gold standard, fixation with a PFNa has grown in favor among surgeons because it allows the patient to ambulate and bear weight on the affected extremity early without fearing implant-related complications.<sup>2-4</sup>

Perioperative blood loss comprises the volume lost during surgery plus the amount collected in post-surgical drains.

eISSN 2012-3264 (Online)
Printed in the Philippines.
Copyright© 2024 by Dollete et al.
Received: October 15, 2023.
Accepted: November 7, 2023.
Published Online: March 14, 2024.
https://doi.org/10.69472/poai.2024.02

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<sup>\*</sup>This study was presented at the 31st POA Midyear Conference Research Contest Podium Presentation Category on April 19-21, 2023 at the SMX Convention Center, Bacolod City.

However, this amount does not correlate with the postoperative drop in hemoglobin.<sup>5</sup> Sehat et al., – in an experiment comparing visible blood loss and total blood loss in arthroplasty cases – proposed that the difference is accounted for by hidden blood loss (HBL).<sup>6</sup> Other sources of loss include those from the initial trauma, intraoperative extravasation into the soft tissues, or other blood loss incurred during hospitalization. In recent years, there has been growing interest in defining HBL, especially in the aged population. If left untreated, HBL may aggravate pre-op comorbidities and lead to anemia.<sup>5</sup> Most studies have focused on quantifying HBL<sup>7</sup> or determining its associated risk factors,<sup>8</sup> but to date none have studied time from trauma to surgery as a variable.

A systematic review and meta-analysis by Klestil et al., 9 noted that hip fractures treated within 48 hours had a lower risk for mortality and perioperative complications. This is, however, not observed in our local setting. Delays to surgery are common due to socio-economic factors and scant resources (including blood products). Whatever the reason for the delay, we still push for surgery to improve hip function and reduce complications when compared to nonoperative treatment. Computing for the HBL may help in estimating the amount of blood products needed to manage postoperative anemia. This study aims to determine if HBL in elderly patients with pertrochanteric fractures treated by PFN is affected by delay in time to surgery.

#### **METHODOLOGY**

This was a detailed cross-sectional study. Exemption from ethics review was granted by the institution's ERB before starting data gathering (POCERB 2020-01-002). The data analyzed was limited to non-identifiable figures, waiving the need for informed consent. The STROBE cross-sectional reporting guidelines<sup>12</sup> were used as a checklist in making this study.

A review of the surgical census and medical records was done to collect data about patients with pertrochanteric fractures treated by closed reduction and short proximal femoral antirotation nailing (PFNa). Patients aged 60 years old and above who were seen at the ER, admitted, and operated on from January 2017 to December 2019 were included. Excluded from the study were patients with 1) fractures of other extremities surgically treated during the same operative time, 2) pathologic fractures, 3) patients with hematologic or GI comorbidities, and 4) patients also taking blood thinners or were given Tranexamic acid.

The sample size was calculated based on the estimation of population proportion for pertrochanteric femur fractures. Assuming that the incidence of hip fractures in the elderly population is 18%,<sup>13</sup> and using a level of significance equal to 5%, the sample size required was 196 patients.

The following data were obtained from the medical and radiographic records of patients: age, sex, height, weight, AO

classification, time from initial trauma to surgery, total operative time, intraoperative visible blood loss from the operative record, and complete blood counts done on admission, within 7 days before surgery and up to 72 hours post-surgery. Patients were classified into either early (time from injury to surgery <30 days) or late ( $\ge$ 30 days) surgery group.

The patient's total blood volume (PBV) was computed using the formula as reported by Nadler et al., <sup>14</sup> as follows:

*PBV, in liters* =  $K1 \times h^3 + K2 \times w + K3$  where h = height in meters, and w = weight in kg, Men: K1 = 0.3669, K2 = 0.03219, K3 = 0.6041, Women: K1 = 0.3561, K2 = 0.03308, K3 = 0.1833

After computing the total blood volume, we computed the estimated blood loss, and the peri-operative hidden blood loss, using the formula by Gross, <sup>15</sup> as follows:

$$Estimated blood loss (mL) = PBV \times [(Hct_{adm} - Hct_{pod1}) / Hct_{av}] \times 1000$$

*Total perioperative blood loss* (mL) = estimated blood loss + blood transfused

 $\label{eq:Perioperative HBL (mL) = 0} Perioperative blood loss - intraoperative visible blood loss$ 

Where Hct<sub>adm</sub> was the hematocrit count done during admission, Hct<sub>pod1</sub> was the hematocrit count on the first postoperative day, and Hct<sub>ave</sub> was the average of all hematocrit counts done from admission to discharge.

The data was encoded in Microsoft Excel and analysis was done using SPSS 20 software. Data was described as mean  $\pm$  standard deviation. Comparison of the demographic and clinical characteristics was performed using the independent sample t-test. A confidence interval of 95% was used to determine statistical significance.

#### **RESULTS**

A total of 210 patients were included in the study. One hundred and thirty-five patients were categorized in the early surgery group, while 75 patients were categorized in the late surgery group.

The mean age for the early surgery group was 77 years old and 76 years old for the late surgery group (Table 1). Most patients were women. Most of the patients were classified under AO 31-A1.2 (2-part pertrochanteric fracture). The operative duration averaged 73.17 minutes in the early surgery group and 72.64 minutes in the late surgery group. The mean visual blood loss for the early surgery and late surgery groups were 247.37 mL and 245.33 mL, respectively. The averages of hematocrit levels taken on admission, pre-operatively within 7 days, and immediately postoperatively are listed in the table.

There were no significant differences between the two groups in terms of age, sex, AO classification, operative duration, HCT levels, intraoperative visual blood loss, postoperative blood transfused, and total blood loss.

Our results showed that there was no significant difference noted between the two groups in terms of peri-op HBL (p = 0.22) (Table 2). The mean peri-operative HBL of patients with delayed time to surgery was 95.32 ml compared to 113.65 ml of patients with early time to surgery.

#### **DISCUSSION**

Increased perioperative blood loss may lead to many complications and a poor prognosis, especially in the elderly population. Therefore, it is important to identify the causes of perioperative blood loss. Hidden blood loss accounts for a high percentage of the total perioperative blood loss in patients with pertrochanteric fractures.<sup>5</sup> If HBL is disregarded, it may lead to anemia or low blood volume. Patient-blood management programs resulted in positive outcomes for patients under orthogeriatric care. It showed a reduction in complication and mortality rates, as well as improvement in functional recovery in the transfused patient group. 16 Foss 17 also reported that uncorrected anemia is associated with decreased mobility and an increased 30-day mortality and length of hospital stay in surgically treated hip fracture patients. Higher hemoglobin levels post-op were also shown to be related to a shorter hospital stay and lower odds of readmission.<sup>18</sup> This fueled the increased interest in quantifying hidden blood loss and studying its risk factors.

Hidden blood loss was computed to be nearly 75% of total blood loss according to Yu et al.,<sup>19</sup> or 86.8% to 89.4% according to Li et al.,<sup>20</sup> using the same formula. In this paper, we computed the mean HBL of 210 patients to be 107.10 mL comprising 30.27% of the mean total blood loss. While published literature reports figures twice as large as ours, there is no denying that HBL makes up a sizeable amount of total blood loss. One of the limitations of this study is the inconsistency in measuring visible blood loss, given that it is a retrospective study. The estimation of visible blood loss may vary from person to person, thus one of the recommendations for a future prospective paper is the creation of a protocol for standardized measurement.

Most available literature on hidden blood loss in surgically treated pertrochanteric fractures excluded patients whose surgeries are delayed. Our "early surgery" setting happens at an average of 18 days after the injury, which is a stark contrast to the immediate surgery done at 1-3 days post-injury in published literature. This delay is brought about by many factors, including patient socio-economic factors and limited hospital resources.

Our results cannot therefore be compared side-by-side to available studies. Yu et al.  $^{16}$  noted an HBL of 272.2 mL ( $\pm$  7.6 mL) in their subset of patients who underwent fixation using a PFNa. A similar study by Yang et al.,  $^{21}$  computed HBL of 787.7 mL ( $\pm$  250.9 mL) in the group fixed with a PFNa. Tian et al.,  $^{11}$  computed HBL to be around 326 to 596 mL, depending on the need for postoperative blood transfusion. Our computed HBLs were 113.65 mL ( $\pm$  99.25 mL) for the early surgery and 95.32 mL ( $\pm$  111.79 mL) for the late surgery groups, with no significant difference between the two groups.

Table 1. Demographic and clinical characteristics of patients by time from injury to surgery (n=210)

	Early Group (n=135)	Delayed Group (n=75)	Р
Age in years (mean, range)	77.00 ± 8.82 (60 to 96)	76.29 ± 7.71 (60 to 94)	0.56 (NS) †
Sex			
Male	23 (17.0%)	11 (14.7%)	0.66 (NS) ‡
Female	112 (83.0%)	64 (85.3%)	
AO Classification			
AO 31-A1.2	70 (51.9%)	37 (49.3%)	
AO 31-A1.3	27 (20.0%)	21 (28.0%)	0.38 (NS) ‡
AO 31-A2.2	26 (19.3%)	9 (12.0%)	
AO 31-A2.3	12 (8.9%)	8 (10.7%)	
Operative duration in mins (mean, range)	73.17 ± 27.84 (25 to 148)	72.64 ± 27.88 (26 to 140)	0.89 (NS) †
Hct (admission) (mean, range)	0.35 ± 0.05 (0.23 to 0.46)	0.33 ± 0.05 (0.20 to 0.46)	0.07 (NS) †
Hct (pre-operatively) (mean, range)	0.35 ± 0.04 (0.28 to 0.54)	0.35 ± 0.03 (0.28 to 0.46)	0.80 (NS) †
Hct (immediate post-op) (mean, range)	0.32 ± 0.04 (0.18 to 0.42)	0.32 ± 0.03 (0.22 to 0.42)	0.60 (NS) †
Visual blood loss (mL), mean	247.37 ± 100.38	245.33 ± 110.03	0.89 (NS) †
Blood transfused (mL), mean	328.36 ± 143.85	327.46 ± 169.62	0.96 (NS) †
Total blood loss (mL), mean	360.85 ± 150.08	341.18 ± 173.88	0.39 (NS) †

 $<sup>^*</sup>p$ >0.05 - Not Significant; p≤0.05 - Significant

Table 2. Comparison of the peri-operative HBL between the two groups

	Early Group (n=135)	Delayed Group (n=75)	p*
Periop HBL (ml). Mean ± SD	113.65 ± 99.25 (31.48%)	95.32 ± 111.79 (27.88%)	0.22 (NS)†

p>0.05 – Not Significant; p≤0.05 – Significant

<sup>†</sup>T-test; ‡Chi-square test

In theory, the absence of fracture hematoma, resolution of third-spacing post-injury, and optimized medical status may account for the lower computed HBL. However, we do not have supporting literature for this, owing to the lack of studies on delayed treatment.

This study has its limitations, such as the study design, the lack of an electronic medical database, and the limited resource setting of the institution. The authors suggest pursuing a prospective study to control confounding factors, such as measurement of visible blood loss, and uniform date of blood extraction for CBC results up to the third day postoperatively. Future studies may reveal a relationship between time to surgery and the amount of blood loss.

#### CONCLUSION

In the setting of delayed treatment of elderly patients with pertrochanteric femur fractures fixed with short proximal femoral antirotation nailing, the amount of hidden blood loss is not significantly affected by the timing of surgery. However, this hidden blood loss still makes up almost a third of the total blood loss and should be anticipated in the management of postoperative anemia.

#### STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

#### **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

#### **FUNDING SOURCE**

None.

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#### **ORIGINAL ARTICLE**









# Four-Strand Modified Kessler versus Cruciate Technique for Repair of Zone Two Flexor Tendon Injuries: A Randomized Controlled Trial

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#### **ABSTRACT**

**Background.** This is the first double-blinded randomized study in which the four-strand modified Kessler and the four-strand cruciate technique were compared in a series of zone II flexor tendon injuries in patients of working age (19–60 years old).

**Objective.** To compare the functional outcomes of zone II flexor tendon repair with the four-core modified Kessler versus the four-core cruciate technique done at our institution's Hand Clinic.

**Methodology.** This double-blinded randomized controlled trial was conducted from September 2022 to August 2023. The sample population consisted of eight fingers of eight patients who sustained traumatic zone II flexor tendon lacerations. Statistical analysis was made between the functional outcomes using the Strickland formula at the fourth, sixth, and eighth weeks, and the FIL-DASH (Disabilities of the Arm, Shoulder, and Hand - Filipino translation) score at the third, and sixth months postoperatively.

**Result.** At weeks four, six, and eight post-operatively, Strickland scores in the four-strand cruciate group were significantly higher than those in the four-strand modified Kessler group (p < 0.02, p < 0.03 and p < 0.02). FIL-DASH scores at three and six months did not differ significantly between the groups.

**Conclusion.** The four-core cruciate technique resulted in significantly better short-term functional outcomes than the four-core modified Kessler technique. More studies are needed to improve on these findings.

**Keywords.** Modified-Kessler, cruciate, functional outcome, FIL-DASH, double-blinded randomized controlled trial, flexor tendon injury

#### **INTRODUCTION**

Traumatic flexor tendon injuries are common causes of emergency room visits and require surgical treatment to restore function and prevent long-term disability. <sup>1-3</sup> These injuries are often caused by lacerations from sharp objects, crush injuries, and sports-related trauma. <sup>4</sup> Surgical repair of flexor tendons is challenging due to the need for precise fusion of transected ends, postoperative mobilization to prevent adhesions, and the risk of re-rupture. <sup>5-7</sup> Early post-operative mobilization reduces the risk of contracture, expedites the healing process, and enables patients to regain full mobility with enhanced grip strength, enabling prompt return to work. <sup>8,9</sup>

A widely recognized technique for tendon repair is the fourstrand modified Kessler stitch, which was derived from the twostrand Kessler suture. This method is initiated in the middle of the cut tendon end and a modified Kessler core is inserted that will pass through only one lateral half of the tendon; thus, apposing only the lateral half of the tendon. The same suture of the modified Kessler core is continuously inserted into the elSSN 2012-3264 (Online) Printed in the Philippines. Copyright© 2024 by Bonifacio et al. Received: April 9, 2024. Accepted: May 13, 2024. Published Online: May 20, 2024. https://doi.org/10.69472/poai.2024.13

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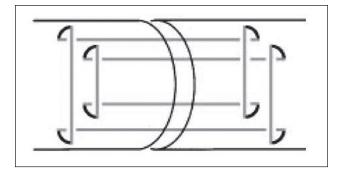


Figure 1. Four-strand modified Kessler tendon repair.

unopposed half of the cut tendon ends, which completes four strands.10 The final phase is done through a double knot to connect the gap between the tendons (Figure 1). Meanwhile, the four-strand cruciate technique is done by making a 2 mm slit on the side of the tendon and 1 cm from the tendon edge.<sup>11</sup> The needle is inserted through the slit passing through the severed tendon edge and longitudinally passing out of the tendon edge. This is followed by the passing of the needle into the corresponding severed tendon edge and going longitudinally out of the side of the tendon. The suture is further reintroduced a few mm distal to the exit point without locking and directed crosswise to move out in the middle of the tendon laceration site. The suture is reinserted into the opposite tendon segment in a crossing fashion and exits on the opposite tendon 1 cm from the laceration site. This is followed by the reinsertion of the suture without locking passing longitudinally across the laceration site. Finally, this is passed back moving through the middle of the laceration site to move out next to the free tendon edge (Figure 2).<sup>11</sup>

The choice of repair method depends on factors such as the timing of the repair, the extent of the injury, and the surgeon's preference. Primary repair is preferred within three weeks of injury, while secondary repair or tendon grafting may be necessary for older injuries. <sup>12</sup> Despite advancements in surgical techniques, there is no consensus on the best approach, and surgeons often rely on their experience and the specifics of each case. <sup>13-15</sup> The goal of repair is to minimize gaps, maintain tendon vascularity, secure suture knots, and provide adequate strength for healing. <sup>14,15</sup> Overall, surgical repair of flexor tendon injuries requires a tailored approach based on the individual patient's needs and the specifics of the injury.

In a recent study on adult sheep tendons, a six-strand (three figure-of-8 sutures) cruciate repair, a 10-strand (six figure-of-8 and cruciate) repair using a combined technique (four figure-of-8 and 10-strand sutures) were compared. Biomechanically, the combined repair was the most robust in terms of both gap and failure (single cyclic tensile). <sup>16</sup>

This paper aimed to compare the functional outcomes of patients with zone II flexor injuries who underwent surgery in a tertiary government hospital using the two suturing techniques, namely the four-strand modified Kessler and cruciate techniques.

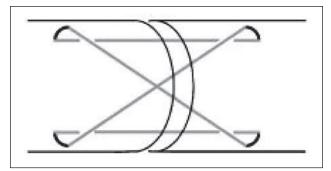


Figure 2. Four-strand cruciate technique tendon repair.

#### **METHODOLOGY**

#### Study design

A double-blind randomized controlled trial was conducted. Each eligible patient was assigned a patient reference number by the primary investigator. A patient reference number assigned as "odd" served as the control group (Arm A: Four-strand core modified Kessler Technique) while patient reference numbers that were "even" served as the experimental group (Arm B: Four-strand cruciate Technique).

In the postoperative evaluation of patients using Strickland's evaluation system<sup>17</sup> and Disabilities of the Arm, Shoulder, and Hand - Filipino translation (FIL-DASH),<sup>18</sup> both the patient and the team of evaluators (one orthopaedic resident who did not perform the procedure and one hand specialist) were blinded to the repair technique.

#### Participant selection

#### Inclusion Criteria

- patients aged 19 to 60 years
- isolated clean laceration on any finger or thumb of either hand (dominant or non-dominant) with a zone II flexor tendon injury of both flexor digitorum superficialis and flexor digitorum profundus tendons needing repair
- no comorbidities

#### **Exclusion Criteria**

- grossly contaminated or infected wounds
- multiple injuries other than flexor tendon injury
- crushing injury of flexor tendon
- intraoperative identification of non-zone II flexor tendon injury
- comorbidities

#### Sample size

Sample size could not be computed a priori due to lack of the required inputs from previous studies for sample size calculation.

The actual sample size achieved was four patients per arm. This comprised the total number of patients enrolled in the study. There was a zero dropout rate.

The final sample size of four patients per arm achieved statistically significant results at a 5% level of significance for all time points (weeks four, six, and eight). Moreover, statistical power achieved by the said sample size was 73, 78, and 91%, for outcome measurement at weeks four, six, and eight.

#### **Data collection**

The assigned procedure was explained and informed consent was taken by the principal investigator for each eligible patient. Participants were randomly assigned to the two arms based on an odd-even scheme of the patient reference number. Arm A underwent four-strand modified Kessler repair and Arm B underwent four-strand cruciate repair. One surgeon (the primary investigator) performed both procedures. Non-absorbable, braided, sterile (Ethibond 5-0) sutures were used for both techniques. Each method was followed by repair of epitenon with continuous running sutures using a non-absorbable nylon suture (Ethilon 4-0). Postoperatively, rehabilitation was done following the Belfast technique, which includes a dorsal blocking splint for six weeks with early active and passive mobilization facilitated by a single JBLMGH rehab therapist. All patients were evaluated postoperatively at four weeks, six weeks, and eight weeks for assessment of total active motion. A comparison was made between the two methods using the Strickland evaluation methodology as outlined in the following equation.

Strickland =  $\{[(active flexion PIP + DIP) - (extension deficit PIP + DIP)]/175°\} \times 100\%$ 

The Strickland scores are classified as follows; Excellent: 85% to 100%, Good: 70% to 84%, Fair: 50% to 69%, Poor: <50%.

The functional outcomes were evaluated by a single team of evaluators (one orthopaedic resident who did not perform the procedure and one hand specialist). Range of motion and tendon excursion were measured using a standard finger goniometer and data was measured using the Strickland evaluation system.

Furthermore, functional outcome was also evaluated in the third and sixth months using FIL-DASH scoring. The main part of the FIL-DASH is a 30-item disability/symptom scale concerning the patient's health status during the preceding months. The items ask about the degree of difficulty in performing different physical activities because of the arm, shoulder, or hand problem (21 items), the severity of each of the symptoms of pain, activity-related pain, tingling, weakness, and stiffness (five items), as well as the problem's impact on social activities, work, sleep, and self-image (four items). Each item has five response options. The scores for all items are then used to calculate a scale score ranging from 0 (no disability) to 100 (most severe disability). The score for the disability/symptom scale is called the DASH score. In this study, the Filipino version (FIL-DASH) was used.

#### **Data analysis**

The baseline characteristics were summarized using frequency distributions. The baseline comparability of groups was determined through Fisher's exact tests. An independent t-test was used to compare the outcomes in the two groups. A  $p \le 0.05$  was taken as statistically significant.

DASH scores were used to evaluate patients at the third and sixth months postoperatively. At least 27 of the 30 items of the questionnaire were answered. The computation and transformation of scores were based on the standard scoring guide as shown below. A higher score indicates greater disability. The standardized response mean was calculated as the mean change scores divided by the standard deviation of the change scores.

DASH disability/symptom score =  $\{[(sum of n responses)]/n - 1\} \times 25,$ 

where n is equal to the number of completed responses.

#### **Ethical considerations**

The study was conducted in compliance with the ethical principles outlined in the Declaration of Helsinki and the National Ethical Guidelines for Health and Health-Related Research of 2017. Prior to study initiation, there was a review and approval of the study protocol and informed consent and subsequent amendments by the Jose B. Lingad Memorial General Hospital Research Ethics Committee (JBLMGH REC), a Level 2 Philippine Health Research Ethics Board (PHREB) – accredited research ethics committee.

There was no direct benefit for the subjects joining this study. However, the results of the study may have indirect benefits. This study will be published and may serve as references for future studies on this topic.

Before a subject's participation, the investigator obtained written informed consent after explaining the aims, methods, anticipated benefits, and potential risks of the study. The informed consent was signed and personally dated by the subject and the person who conducted the informed consent discussion. One copy of the signed informed consent was given to the subject. Furthermore, participants could withdraw anytime from the study. Disclosure of potential conflicts of interest was discussed regularly.

The investigator preserved the confidentiality of all subjects taking part in the study. The investigator ensured that the subject's anonymity was maintained. The risk to the subject's privacy was minimal and no sensitive information was obtained. All data were encoded using a password-protected Excel spreadsheet. A code number was assigned for each patient and recorded in a separate password-protected spreadsheet. Only the primary investigator had access to this

file. After encoding, all data collection forms were kept in a secured cabinet. The researchers intended to adhere fully to the provisions of the Data Privacy Act of 2012.

#### **RESULTS**

A total of eight patients (four patients per arm) participated in this study. The two groups were found to be comparable at baseline (i.e. no significant differences were noted in the two groups in terms of age, gender, handedness and affected digit; p > 0.05) (Table 1).

At four weeks post-operatively, two patients in the four-strand modified Kessler Technique group had good functional outcomes based on Strickland's Evaluation System and the other two patients had fair functional outcomes. On the other hand, one patient in the four-strand cruciate Technique had excellent functional outcomes, two patients had good functional outcomes and one patient had fair functional outcomes. There were significant differences in the Strickland scores between the groups favoring the cruciate technique at weeks four, six, and eight (p <0.05) (Table 2).

In terms of FIL-DASH, the cruciate group had lower mean scores (indicating less disability) at months 3 and 6 but the differences were not statistically significant (p > 0.05).

Table 1. Baseline characteristics of the patients

		Four-strand modified Kessler technique n = 4	Four- strand cruciate technique n = 4	₽ <sup>F</sup>
Age	19–30 years old 31–45 years old	2 2	2 2	1.00
Gender	Male Female	3 1	2 2	1.00
Handedness	Dominant Non-dominant	3 1	3 1	1.00
Affected Digit	Index Finger Middle Finger Ring Finger Small Finger	1 2 1 0	0 1 2 1	1.00

<sup>\*</sup>F = Fisher's exact test. 2-tailed

**Table 2.** Comparison of functional outcomes of patients who underwent four-strand modified Kessler technique and four-strand cruciate technique

Functional	Four-strand modified	Four-strand	₽°
Outcome based	Kessler technique	cruciate technique	
on Strickland	n = 4 (%)	n = 4 (%)	
Scores	Mean ± SD	Mean ± SD	
Week 4	62.00 ± 9.90	77.25 ± 6.65	0.02*
Week 6	71.25 ± 11.06	87.25 ± 3.95	0.03*
Week 8	81.00 ± 8.68	95.50 ± 1.29	0.02*
Functional	Four-strand modified	Four-strand	₽ <sup>b</sup>
Outcome based	Kessler technique	cruciate technique	
on FIL-DASH	(Mean ± SD)	(Mean ± SD)	
Month 3	5.63 ± 2.20	4.60 ± 1.75	0.25
Month 6	2.48 ± 1.65	1.25 ± 1.44	0.15

<sup>\*</sup>Significant difference at  $\alpha = 0.05$ 

#### **DISCUSSION**

Regaining full function of the finger after a flexor tendon laceration is one of the most difficult tasks in the field of hand surgery. Improvements in technique and post-operative care have achieved reliable flexor tendon repairs, optimizing digital motion and functional outcomes. The surgical repair of zone two flexor tendon injuries has been the subject of considerable discussion. However, adhesion formation, suture rupture, and fixation of sutures on pulley edges remain potential consequences of inadequate repair.<sup>6</sup>

Theoretically, increasing the number of sutures that cross the repair site can reduce the likelihood of rupture. Core sutures with more strands crossing the repair site have a higher degree of tensile strength compared to those with a similar design but fewer strands.<sup>3</sup>

The advantages of multi-strand suture techniques demonstrated in vitro do not necessarily translate into improved outcomes in vivo. Numerous studies have indicated that multi-strand techniques may possess a higher degree of in vitro gliding resistance. In a 2001 study conducted by Zhao et al., gliding resistance and adhesion formation were compared between two-strand modified Kessler and a four-strand Becker repair in a dog model. The two techniques were selected due to their relatively low gliding resistances and their respective postoperative mobility protocols. The study found that the gliding resistance of the Kessler group was significantly less than that of the Becker group, with the gliding resistance being significantly lower at three and six weeks post-operatively. Therefore, it was suggested that gliding resistances may be more significant than the strength of the suture, provided a post-operative low-force gliding protocol is employed.

Our study was designed to compare the functional outcome of zone II tendon repair with four-strand modified Kessler as compared to the four-strand cruciate technique. Many studies have investigated this in animal and in vitro models, but this is the first double-blinded randomized study in which the four-strand modified Kessler and four-strand cruciate technique have been compared in a series of zone II flexor tendon injuries in patients of working age (19–60 years old).

A similar study conducted with a prospective case-control design by Dawood found better functional results in four-strand cruciate repair especially in zone II, with excellent results in 33.3%, good in 50%, and fair in 16.6% of cases, as compared to modified Kessler repair with no excellent results, 33.3% good, 50% fair and 16.6% poor results. Navali et al., found that four-strand suture repair prevented tendon ruptures and achieved excellent and good functional results in 90% of cases.

In the present study, no ruptures were encountered during early postoperative rehabilitation. The four-strand cruciate technique yielded significantly better Strickland scores at weeks four, six, and eight (Table 2). A single knot is required for

a = one-tailed independent t-test assuming unequal variance

b = one-tailed independent t-test assuming equal variance

this repair, which has been shown to have several advantages. While each additional knot has been demonstrated to reduce the strength of the suture material, a single knot results in a reduced tendon manipulation during the repair favoring significant difference during evaluation. The suture repair limbs that run along the contours of the tendons promote anatomical alignment and proper placement. If the suture can glide at the non-locking corners, this method balances the stress across all four strands of the repair. The other four-strand repair methods tested were dual-strand, which unevenly distributes stress across the multiple sutures, resulting in a final tensile strength that is only as high as the initial failure point of the suture.<sup>11</sup>

Thanks to the simplicity of design, ease of execution, and superior mechanical properties of the four-strand cruciate technique, it is a good choice for flexor tendon repair.

#### **CONCLUSION**

The four-strand cruciate technique provides better functional outcomes than the four-strand modified Kessler technique. Using the right surgical technique helps patients recover faster, return to work earlier, and provide for their families. Further studies on more patients are needed to add to the strength of these findings.

#### **ACKNOWLEDGMENTS**

This study was made successful through the aid of significant persons who guided and helped the Researchers throughout the completion of the study. Their utmost gratitude is given to the following individuals who helped broaden their perspective as they went through the study:

Their respective Department and Chairman, Dr. Ivan John P. Concepcion and faculty for sharing their knowledge and expertise and for supervision.

Their JBLMGH-PETRO-OBRAS professors and their statistician, Ms. Madonna Valenzuela who, through their guidance and encouragement helped them a lot with ideas and writing skills.

Their patients who eagerly participated and committed to this research.

#### STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

#### **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

#### **FUNDING SOURCE**

None.

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# Filipino Translation, Cross-Cultural Adaptation and Validation of the American Orthopaedic Foot and Ankle Society's (AOFAS) Ankle-Hindfoot Scale

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#### **ABSTRACT**

**Background.** Injuries about the foot and ankle account for a significant number of injuries that may lead to substantial functional impairment and disability. A standardized method of outcome assessment is necessary to evaluate and monitor patients' progress. One commonly used evaluation system is the American Orthopaedic Foot and Ankle Society (AOFAS) Ankle and hindfoot scale. The purpose of this study was to translate the AOFAS Ankle-Hindfoot scale into Filipino and establish its cultural adaptation and validity.

**Methodology.**: This was a single-center cross-sectional study that included patients with ankle and hindfoot conditions who took part in the evaluation of the proposed Filipino translation of the Ankle-Hindfoot Scale, following the guidelines set by the AOFAS. Construct validity and test-retest reliability were analyzed using intra-class correlation coefficients; internal consistency was analyzed with Cronbach's alpha. A Rasch principal components analysis was also used to test for reliability and construct validity.

**Results.** Cronbach's alpha was measured at 0.98 in terms of function, while the item separation index showed good internal consistency of the construct. Rasch analysis confirmed that the construct is a multidimensional metric. Intra-class correlation using Pearson's coefficient was significant at 0.8779 (p <0.05), showing good test-retest reliability, while convergent validity confirmed a strong positive correlation between the overall AOFAS score and the health perception domain of the Medical Outcomes Study Questionnaire Short Form-36 (SF-36 v2) (Tagalog) (Pearson = 0.647).

**Conclusions.** The Filipino translation of the AOFAS Ankle-Hindfoot Scale was successfully translated and culturally adapted to Filipino patients, with good internal consistency. We have likewise demonstrated a good correlation against a general health outcome measure (SF-36 v2) in terms of change in health perception and role limitation (physical).

Keywords. AOFAS, ankle and hindfoot, validation, cultural adaptation, Filipino

#### **INTRODUCTION**

Injuries about the foot and ankle account for a significant number of injuries that may lead to functional impairment and disability. Foot and ankle pain represents a substantial community burden among middle-to-older age individuals, being present in approximately one out of five individuals. Among the younger population, particularly athletes, 25% of reported injuries over a 6.5-year period occur around the foot and ankle. Treatment is better evaluated using standardized outcome measures, as evidenced in scientific literature, which allows the health care professional to quantify disability and monitor the progress of treatment in a patient, and/or for public researchers to evaluate the outcome of different health services.

eISSN 2012-3264 (Online)
Printed in the Philippines.
Copyright© 2024 by Robles and Gamboa.
Received: September 16, 2023.
Accepted: February 14, 2024.
Published Online: March 15, 2024.
https://doi.org/10.69472/poai.2024.03

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The American Orthopaedic Foot and Ankle Society (AOFAS) survey was developed in 1994 by Kitaoka's group for clinicians to assess outcome measures for patients with foot and ankle dysfunctions,2 and at present is one of the most widely used outcome measures by clinicians. Originally in the English language, it explores both subjective and objective experiences of pain, alignment, and function, intended to be completed collaboratively by both the clinician and the patient. The AOFAS has different versions assigned to the four different regions of the foot and ankle, namely: the ankle-hindfoot, midfoot, metatarsophalangeal (MTP)-interphalangeal (IP) for the hallux, and MTP-IP separately for the lesser toes. This evaluation tool is subdivided into three categories: pain, functional aspects, and alignment. The total score consists of 100 points, with a higher score indicating a more favorable clinical status.2 Since its inception, this assessment tool has been one of the most widely used by orthopedists due to its ease of application and its replicability. There have since emerged several versions of this scoring system in other languages: Persian, Turkish, Italian, Dutch, German, Arabic, and Portuguese, with some studies limiting their translation to the Ankle-Hindfoot score.3 The need to translate and culturally adapt this questionnaire emerged from the need to maintain the content validity of the instrument. As outlined by Guillemin et al., cross-cultural adaptation is required in the setting when the source questionnaire will be used in another country with another language, wherein there exists a culture change that may influence the participant's response. Crosscultural adaptation therefore ensures that the psychometric properties of the construct are retained to reach semantic, experiential, and conceptual equivalence.4 Currently, there has been no version of the AOFAS survey in the Filipino language for any of the versions. The high rate of injuries and pathologies centering on the ankle and hindfoot in daily activities, sports, and the like, led us to a particular interest in developing a Filipino translation of the Ankle-Hindfoot scale.

This study was developed to establish the validity, cultural adaptability, and internal consistency of a Filipino translation of the AOFAS Ankle-Hindfoot scale.

#### **METHODOLOGY**

#### **Translation process**

Translation of the AOFAS questionnaire was done following the guidelines set by the American Association of Orthopedic Surgeons. Permission to translate the AOFAS Ankle-Hindfoot scale into Filipino was sought and subsequently granted by the research division of the American Orthopaedic Foot and Ankle Society Research Committee after correspondence.

Forward translation (English to Filipino) was done by two native Filipino speakers who are fluent in the English language. The first translator was knowledgeable on the AOFAS (AOFAS T1) while the second translator had no prior experience to produce a version (AOFAS T2) that could be generally understood by the general population. Upon

consultation with linguist experts from the Sentro of Wikang Filipino, both translations were compared, discussed, and then synthesized to resolve any discrepancy and labeled as AOFAS T1-2. It was agreed early on to retain the English terminologies in the anatomical and joint movements portion of the scale.

Backward translation (Filipino to English) of the questionnaire to the original language was then performed by another set of people to check the validity of the translated version and compare it to the original AOFAS version. Two back translations were done by two bilingual persons with English as their mother tongue and likewise had no medical background and were blinded to the purposes of the study.

An expert committee comprising the investigators, a language professional, and the forward and backward translation team consolidated all versions and components of the questionnaire to develop a pre-final version in terms of semantic, idiomatic, experiential, and conceptual equivalence. The anatomical and joint movement portion of the questionnaire (AOFAS physician part) was not translated into Filipino terms as there were no equivalent translations, and doing so may have caused an error in its application. No pre-testing was done.

#### Study design

This was a cross-sectional study that included patients with established diagnoses of ankle and hindfoot conditions, which were confirmed via imaging and clinical examination. A list of patients was selected from the Institute of Orthopedics and Sports Medicine patient census (for both private and social service cases) and screened for eligibility. The study covered patients who had a diagnosis of a single ankle and hindfoot pathology from Jan 1, 2018, to Dec 31, 2019.

Inclusion criteria for the study were: (1) 18 years of age and above with a good comprehension of the Filipino language, (2) patients with an existing single ankle and hindfoot pathology as confirmed by an orthopedist who could answer the administered AOFAS Ankle and Hindfoot scale questionnaire. Exclusion criteria were: (1) patients who had coexisting injuries, such as polytrauma; (2) patients with cognitive impairment; and (3) patients who were nonambulatory at the time of the survey.

#### **Data collection and outcomes**

The list of patients was retrieved from the existing census of the institution, with priority for the social service patients who fit the eligibility criteria, followed by the list of private patients. Data collected included patient demographics, diagnosis, operative details if any, and additional contact details from the electronic hospital records. Patients who were administered the questionnaires were contacted via a telephone call and video consult, after giving verbal and signed consent (sent electronically by the principal investigator). The first 30 patients who responded were included in the study. The same subset of patients was also made to answer the Tagalog

version of the Medical Outcomes Study Questionnaire Short Form-36 (SF-36) v2, the score of which was used to check for construct validity.

Intra-interviewer reproducibility was checked by contacting the same patients within 7–14 days to obtain a new assessment using the Filipino AOFAS Ankle-Hindfoot scale.

The internal consistency of the questionnaire was assessed using Cronbach's alpha using a 95% confidence interval. A good value of the alpha was set at 0.7 to 0.95.5 A Rasch Principal Components Analysis (item reliability index, person separation, and reliability index) was also employed to further check the reliability of the instrument.6

Test-retest reliability and intra-interviewer reproducibility were determined by obtaining the intra-class correlation

**Table 1.** Sociodemographic and clinical characteristics of patients included in validation and translation process of AOFAS scale for ankle and hindfoot

	Median (Range); Frequency (%)
Age (years)	47.5 (21–75)
Sex Male Female	11 (36.6%) 19 (63.33%)
Educational attainment College graduate High school graduate	14 (46.67%) 16 (53.33%)

Table 2. Distribution of diagnoses

Diagnosis	Frequency (%)
Achilles tear	1 (3.33)
Achilles tendinitis	5 (16.67)
Post-traumatic arthritis	1 (3.33)
Malunion, bimalleolar ankle	2 (6.67)
Bimalleolar fracture	2 (6.67)
ATFL sprain	4 (13.3)
Lateral malleolus fracture	6 (20)
Plantar fasciitis	8 (26.67)
Trimalleolar fracture	1 (3.33)
Grand Total	30 (100)

**Table 3.** Distribution of scores based on AOFAS Ankle-Hindfoot Scale

Statistics	Mean	Median	SD	Min	Max
Pain	32.333	30	5.040	20	40
Function					
Restraints in activities	8.300	7	1.512	7	10
Maximum walking distance	4.133	4	0.973	2	5
Walking surface	4.400	5	0.932	3	5
Gait abnormality	7.733	8	1.015	4	8
Sagittal mobility	7.467	8	2.030	0	8
Hindfoot mobility	5.600	6	1.522	0	6
Stability	8.000	8	0	8	8
Alignment	10.000	10	0	10	10
Overall	87.967	89	7.499	67	100

(ICC) and Pearson's correlation coefficient, with the level of significance set at 5%. Koo and Li outlined the interpretation of the ICC with a coefficient of <0.50 as poor, 0.50–0.75 as moderate, 0.75–0.90 as good, and above 0.90 as excellent.<sup>7</sup>

To establish construct validity, we correlated the results against a widely accepted and commonly used general health outcome tool, in the form of a validated SF-36 questionnaire, for which a Filipino version is also available.<sup>8</sup>

#### Statistical analysis

WINSTEPS® Rasch Analysis and Rasch Measurement software and R statistical software were used for data analysis for the previously mentioned outcome measures. The Rasch analysis is a statistical model-based latent trait psychometric technique that allows examination of the fit between the rating scale data and the prescriptive Rasch statistical model.

Descriptive statistics were used to summarize the clinical and demographic data of the study participants. Frequency was utilized for nominal variables, while the median was employed for data with a range, and the mean, along with its standard deviation and coefficient of variation, was utilized for quantitative variables. The level of significance was set at a p < 0.05.

#### **RESULTS**

We analyzed a total of 30 patients who had an existing diagnosis of ankle and/or hindfoot conditions. The mean age was 45 years old with a range of 21 to 75 years old, with a higher percentage of female participants (Table 1). Fifty-three percent of the patients had attended high school while the rest held a college degree. The diagnosis of plantar fasciitis comprised the greatest number of respondents, followed by patients who sustained isolated lateral malleolus fractures (Table 2). The mean overall score for the AOFAS Ankle-Hindfoot scale was 87.96 (range: 67-100), with an SD of 7.499 (Table 3) while the summary of the corresponding SF-36 v2 subscale scores are likewise seen in Table 4.

Table 4. Distribution of scores based on SF-36 v2 (Tagalog)

Statistics	Mean	Median	SD	Min	Max
Physical function	78.833	80	7.953	55	90
Social function	73.200	78	23.116	11	100
Mental health	86.267	88	12.236	36	100
Pain	78.667	78	14.587	33	100
Change in health	64.167	62.5	18.198	25	100
Role limitation – physical	76.667	75	28.567	0	100
Role limitation – mental	88.933	100	26.705	0	100
Energy/vitality	77.667	80	9.803	55	100
Health perception	79.700	82.5	15.733	35	100

Table 5. Reliability using Cronbach's alpha of AOFAS Ankle-Hindfoot Scale

Item	Size	Sign	Correlation	Item-test correlation	Item-rest covariance	Cronbach's alpha
Pain	30	+	0.6043	0.319	2.1714	0.6691
Restraints in activities	30	+	0.3324	0.2385	2.8868	0.6667
Maximum walking distance	30	+	0.3928	0.3358	2.9090	0.6647
Walking surface	30	+	0.7271	0.6957	2.6942	0.6424
Gait abnormality	30	+	0.6331	0.5899	2.7270	0.6464
Sagittal mobility	30	+	0.6331	0.5409	2.4153	0.6211
Hindfoot mobility	30	+	0.6331	0.5661	2.5589	0.6321
Overall	30	+	1	1	0.4588	0.3997
Test Scale					2.3527	0.6693

**Table 6.** Reliability using Rasch Model for the AOFAS Ankle-Hindfoot Scale

Dimension	Cronbach's alpha		Persons	Item
All	0.6693	Separation	1.46	7.10
		Reliability	0.68	0.98

The questionnaires were administered via videoconference. Each item of the questionnaire was read and explained to the patient and their corresponding response was recorded. Due to the nature of the format, it took more time to gather the responses. The clinician-reported items were also challenging to document through videoconference.

# Internal consistency, reliability, and construct validity

Internal consistency for the nine-item scale was computed to have a Cronbach alpha of 0.6693 (acceptable is at least 0.7) (Table 5). Rasch component analysis was made to also check for internal consistency with a computed item separation

index of 0.98 (Table 6). A separate analysis was also done to check for reliability based on the dimension of Function (Table 7) alone and was found to have a good Cronbach's alpha value of 0.814. Rasch analysis was used to further analyze the overall low Cronbach alpha. Taking all questionnaire items together, the index of raw variance was found to be 81.9% but with noted unexplained variance above the ideal value of 2 (Eigenvalue: 4.62) (Table 8). Point Measure Correlation indices were all positive which indicates that all questionnaire items pointed towards the same construct (Figure 1).

Test and retest reliability were determined to be at 0.8779 (95% CI: 0.7596 - 0.94, p < 0.001) for both ICC and Pearson's correlation coefficient (Table 9).

Construct validity measured by obtaining Pearson's correlation coefficient a showed a strong positive correlation between the overall AOFAS score and the health perception domain of the SF-36 v2 (Tagalog) (Pearson = 0.647, p = 0.0001, 95% CI: 0.3661-0.8138), a moderate positive correlation between AOFAS scale and the domains of social function (Pearson

Table 7. Reliability using Cronbach's alpha for the Function dimension of the AOFAS Ankle-Hindfoot Scale

Item	Size	Sign	Correlation	Item-test correlation	Item-test covariance	Cronbach's alpha
Restraints in activities	30	+	0.5335	0.3157	0.9646	0.8439
Maximum walking distance	30	+	0.4282	0.2822	1.0646	0.834
Walking surface	30	+	0.6501	0.5456	0.9432	0.7976
Gait abnormality	30	+	0.9126	0.8762	0.7674	0.7411
Sagittal mobility	30	+	0.9126	0.8146	0.5209	0.7289
Hindfoot mobility	30	+	0.9126	0.8496	0.6184	0.7151
Test Scale	0.8132	0.814				

ENTRY	TOTAL	TOTAL		MODEL  IN	IFIT   OUT	TFIT	PTMEAS	UR-AL EXACT	MATCH	- 1
NUMBER	SCORE	COUNT	MEASURE	S.E. MNSQ	ZSTD MNSQ	ZSTD	CORR.	EXP.   OBS%	EXP%	Item
					+	+	<u></u>	+	+-	
13	240	30	-2.15	.33  .65	97 .25	-2.88	.00	.60  93.3	72.2	I0013
3	124	30	2.48	.17 1.07	.32 1.55	1.68	.48	.72 53.3	40.2	10003
1	234	30	-1.54	.30 2.46	2.67 2.84	3.11	.53	.66  .0	74.1	10001
5	132	30	2.25	.17  .69	-1.15  .86	41	.71	.73  70.0	39.8	10005
7	232	30	-1.37	.29  .49	-1.43  .22	-2.68	.87	.67  93.3	73.3	10007
9	224	30	80	.24 3.17	4.36 1.85	2.01	.87	.67 73.3	60.3	10009
11	168	30	1.14	.17 .46	-2.56 .35	-3.29	.87	.68 73.3	40.6	I0011
İ				+	+	+		+	+-	·
MEAN	193.4	30.0	.00	.24 1.28	.2 1.13	4		65.2	57.2	į
P.SD	47.0	.0	1.77	.06 1.00	2.3 .92	2.5		29.6	15.3	ĺ

Figure 1. Point measure correlation index.

Table 8. Standardized Residual Variance in Eigenvalue Units

Dimension	Eigenvalue	Observed	
All			
Raw variance explained by measures	31.7657	81.9%	
Unexplained variance in 1st contrast	4.62	14.6%	

= 0.4975, p = 0.0052), energy/vitality (Pearson = 0.4891 p = 0.0061), and role limitation (physical) (Pearson = 0.4188, p = 0.0213) (Table 10).

#### **DISCUSSION**

The original AOFAS questionnaire is in the English language, and while our familiarization with English allows us to understand and converse in it, using the original AOFAS questionnaire in our country can introduce cultural differences that may impede accurate interpretation or measurement. As such, these instances require cultural adaptation and translation to maintain the equivalence and validity of the instrument.<sup>4</sup>

This study was able to create a Filipino translation of the AOFAS Ankle-Hindfoot scale that shows internal consistency in terms of a single dimension (function) (Cronbach alpha 0.814) but not for the other dimensions, or the tool as a whole. The Rasch model analysis for internal consistency consisted of the following measures: person reliability index, an item reliability index, an item separation index, and a person separation index.<sup>7</sup> The person reliability and item reliability values range from 0 to 1 and can be interpreted much like Cronbach's alpha. Taking these into account, our construct showed strong item reliability with a value of 0.98, indicating good internal consistency of the variables measured.

Separation index values range from 0 to infinity, with higher values indicating better separation. The item separation

index reveals how well a sample of people can separate items. Analysis of the construct demonstrates a good item separation of 7.10. According to Linacre in 2012, an item separation index score of 3 or more is desirable. Pearson reliability index reveals how well a set of items separates persons measured, with a value of 1.5 as acceptable, 2 as good, and 3 as excellent. The low person indices (separation: 1.46, reliability 0.68) in this analysis may suggest that the instrument may not have sufficient sensitivity to consistently differentiate between respondents (high and low performers) or that more welltargeted items may be needed in the instrument. 10 Rasch analysis9 also looked at the dimensionality of the construct. Entries from the AOFAS Ankle-Hindfoot scale were entered in the WINSTEPS software to measure raw variance. The index of raw variance was greater than the standard of 40% (Observed 81.9%) with an unexplained variance. Despite this, the positive point measure correlation index in its subanalysis supports the finding that the items point toward the same construct, making the questionnaire sound.

The statistically significant values for both Pearson's and ICC for test-retest reliability suggest that the assessment questionnaires are reproducible over time in the same patient. This was consistent with the other translations of the AOFAS Ankle-Hindfoot scale.<sup>3,11</sup> Repeat examination was done within at least one week, not exceeding fourteen days. Strong correlations and reproducibility of the values indicate excellent consistency among the questions, suggesting they are easily understood by respondents.

Convergent validity of the AOFAS Ankle-Hindfoot Scale was done by comparing the scores with a validated SF-36 v2 (Tagalog)<sup>8</sup> using Pearson's correlation coefficient. The SF-36 is a widely accepted and generic outcome instrument to assess a patient's health status, including musculoskeletal conditions.<sup>12</sup> Good construct validity was demonstrated for Persian and Italian translations of the AOFAS when

Table 9. Intra-class correlation and Pearson's correlation of AOFAS overall score

AOFAS	Coefficient	Test stat	Р	Lower limit	Upper limit
Pearson's	0.8779	9.7008	<0.001*	0.7571	0.9407
ICC	0.8779	15.3781	<0.001*	0.7596	0.94

<sup>\*</sup>Significant at 5%

Table 10. Pearson's correlation of AOFAS overall score and SF-36 v2

SF-36		AOFAS							
5r-30	Pearson	Test Stat	р	Lower limit	Upper limit				
Physical function	0.3173	1.7706	0.0875	-0.0485	0.6081				
Social function	0.4975	3.0348	0.0052*	0.1672	0.7274				
Mental health	0.2023	1.0929	0.2837	-0.1704	0.5243				
Pain	0.2212	1.2001	0.2402	-0.1511	0.5385				
Change in health	-0.1859	-1.0013	0.3253	-0.5119	0.1869				
Role limitation – physical	0.4188	2.4401	0.0213*	0.0689	0.6769				
Role limitation – mental	0.3139	1.7493	0.0912	-0.0523	0.6057				
Energy/vitality	0.4891	2.967	0.0061*	0.1564	0.7221				
Health perception	0.6417	4.4275	1.00E-04*	0.3661	0.8138				

<sup>\*</sup>Significant at 5%

correlated with SF-36 v2, while other translations made use of other functional outcome forms. We have made use of a corresponding language version of the SF-36, which has 8 subscales. The Filipino version of the AOFAS Ankle-Hindfoot scale has the strongest positive correlation with the health perception domain of SF-36 v2 (Pearson = 0.647, p = 0.0001, 95% CI: 0.3661–0.8138) and moderate positive correlation with the domains of social function, energy/vitality and role limitation (physical). Although vitality/energy and social function showed statistically significant values, there were no corresponding questions of these items seen in the AOFAS ankle-hindfoot assessment scale.

At the height of the pandemic when this study was performed, the authors shifted to videoconferencing to administer the questionnaires. This proved advantageous since it was convenient for both the patient and the interviewer. However, it took a longer time to complete the questionnaire, as the interviewer would read the questions and the itemized responses one by one to the patient. Answering the anatomic items also proved to be a challenge as the interviewer would need to demonstrate the examination and wait for a return demonstration by the patient.

#### **CONCLUSION**

The translation of the AOFAS Ankle-Hindfoot scale into Filipino along with its cultural adaptation, internal consistency, and convergent validity suggests that this clinical assessment tool may be used for Filipino patients with foot and ankle pathologies.

This study is however limited in terms of not being able to do an inter-interviewer analysis as well as testing for responsiveness of the tool, which needs more time to measure (i.e., one-month interval). The manner of the examination (videoconference) may likewise influence the response rate of patients, as we have observed a longer time it took to examine in this setting.

#### STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

#### **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

#### **FUNDING SOURCE**

None.

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#### **APPENDICES**

<b>Appendix A.</b> AOFAS Anl	kle and Hindf	oot Scale (Fi	lipino)
------------------------------	---------------	---------------	---------

Pan	galaı	n ng Pasyente:		
Cas	e#n	g Pasyente:		
		<u> </u>		
I.		nanakit (40 puntos) Wala [+40] Bahagya, paminsan-minsan [+30] Katamtaman, araw-araw [+20] Matindi, halos laging nananakit [0]		gittal Motion (flexion plus extension)  Normal o bahagyang restriksiyon (30° o higit pa) [+8]  Katamtamang restriksiyon (15°–29°) [+4]  Malaking restriksiyon (mababa sa 15°) [0]
II.	Mg	kayahan (50 puntos)  a limitasyon sa gawain, kinakailangang suporta  Walang mga limitasyon, walang suporta [+10]  Walang mga limitasyon sa mga pang-araw-araw na gawain, may mga limitasyon sa mga gawaing panlibangan, walang suporta [+7]  Limitadong gawaing pang-araw-araw at panlibangan, tungkod [+4]  Malaking limitasyon sa mga gawaing pang-araw-araw at panlibangan, walker, saklay, wheelchair, brace [0]  akamalayong distansiya sa paglalakad, mga kanto Higit sa anim [=5]	plu	galaw ng likurang bahagi ng paa o hindfoot (inversion is eversion)  Normal o bahagyang restriksiyon (75%–100% na normal) [+6]  Katamtamang restriksiyon (25%–74% na normal) [+3]  Hindi maitatanggi ang restriksiyon (mababa sa 25% ng normal) [0]  tatagan ng sakong-hindfoot (anteroposterior, varusgus)  Matatag [+8]  Tiyak na hindi matatag [0]
		Apat – anim [+4] Isa – tatlo [+2] Wala pang isa [0]  ng nilalakaran Walang hirap sa kahit anong uri ng nilalakaran [+5] May kaunting hirap sa mga hindi patag na daraanan, baitang, dalisdis, hagdan [+3] Matinding hirap sa mga hindi patag na daraanan, baitang, dalisdis, hagdan [0]	□ □ Kal	Normal, plantigrade foot, maganda ang alignment ng ankle-hindfoot [+6]  Karaniwan, plantigrade foot, may kaunting malalignment ng ankle-hindfoot; walang sintomas [+3]  Hindi maayos, nonplantigrade foot, malalang malalignment, mga sintomas [0]
	Ab	normalidad sa porma paglalakad Wala, kaunti [+8] Halata [+4] Hindi maitatanggi [0]		Puntos para sa pananakit + Puntos para sa kakayahan + Puntos para sa alignment = Kabuoang Marka/100 puntos

Appendix B. AOFAS Ankle and Hindfoot Scale (English)

Patient Name:			
Patient MRN:	_		
Date:	_		
I. Pain (40 points)		Sagittal motion (flexion plus extension)	
None	+40	Normal or mild restriction (30° or	
Mild, occasional	+30	more)	+8
Moderate, daily	+20	Moderate restriction (15° - 29°)	+4
Severe, almost always present	+0	Severe restriction (less than 15°)	+0
II. Function (50 points)		Hindfoot motion (inversion plus eversion)	
Activity limitations, support requirements		Normal or mild restriction (75% -	
No limitations, no support	+10	100% normal)	+6
No limitation of daily activities,		Moderate restriction (25% - 74%	+3
limitations of recreational activities,	+7	normal)	+5
no support		☐ Marked restriction (less than 25% of	+0
Limited daily and recreational	+4	normal)	+0
activities, cane			
Severe limitation of daily and		Ankle-hindfoot stability (anteroposterior,	
recreational activities, walker,	+0	varus-valgus)	
crutches, wheelchair, brace		Stable	+8
Maximum walking distance, blocks		Definitely unstable	+0
Greater than six	+5	W Al' (40 ' )	
Four-six	+4	III. Alignment (10 points)	
One-three	+2	Good, plantigrade foot, ankle-hindfoot	+10
Less than one	+0	well aligned  Fair, plantigrade foot, some degree of	
	. 0	ankle-hindfoot malalignment	+5
Walking surfaces		observed, no symptoms	13
No difficulty on any surface	+5	Poor, nonplantigrade foot, severe	
Some difficulty on uneven terrain,		malalignment, symptoms	+0
stairs, inclines, ladders	+3		7.
Severe difficulty on uneven terrain,	. 0	IV. Total Score (100 points):	
stairs, inclines, ladders	+0	Pain Points +	
		Function Points +	
Gait abnormality		Alignment Points =	
None, slight	+8		
Obvious	+4		
Marked	+0	Total Points/100 points	

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# A Comparative Study of Proximal Femoral Nail Antirotation in Peri-trochanteric Fractures in Lateral Decubitus and Supine Position\*

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#### **ABSTRACT**

**Objectives.** One of the most widely used cephalomedullary devices for unstable peri-trochanteric fractures is the Proximal Femoral Nail Antirotation (PFNA). Adequate reduction and fixation are crucial to achieving ideal function and fracture union. Many factors can contribute to the ease of reduction; one of these is the positioning technique. Numerous reports have addressed the advantages and disadvantages of the positioning techniques for antegrade femoral nailing. This study aimed to compare the quality of fixation, adequacy of reduction, bony union, and functional outcomes in PFNAs done in the lateral decubitus and supine position.

**Methodology.** This study was a retrospective cohort study conducted at the Philippine Orthopedic Center. Adult male and female (21 to 65 y/o) patients who underwent open reduction with a PFNA either in the supine or lateral position were included in this study. Thirty-nine (39) patients were identified, where six were lost to follow-up, and four did not have retrievable postoperative radiographs. A total of 29 patients were included. The data were analyzed using the T-test in two population means and Fisher's Exact Test.

**Results**. At a 95% level of confidence, the study showed no significant differences in the distribution of Tip-Apex Distance (TAD), adequacy of reduction, and bony union at six months post-operatively between the lateral and supine position. In contrast, there was a significant difference in the distribution of the Cleveland index score. Regarding the Harris Hip Score (HHS), there was also a significant difference in the average score between the two groups, but all had an excellent functional outcome.

**Conclusion.** The preferred surgical position for performing an open reduction and fixation with a PFNA remains controversial. This study showed that the surgical position did not affect the TAD, adequacy of the reduction, and bony union. All patients from both groups had excellent functional outcomes at six months post-operatively, but the lateral position group had superior Cleveland index scores. Surgery with the PFNA in the lateral decubitus position can be performed in small rural hospitals that lack a fracture table. With proper surgical technique, this may be safe, executable, and may benefit more patients with peritrochanteric fractures.

Keywords. peri-trochanteric fractures, subtrochanteric fractures, cephalomedullary nail, PFNA, hip surgery

#### **INTRODUCTION**

Peri-trochanteric fractures are fractures occurring between the extracapsular part of the neck to a point 5 cm distal to the lesser trochanter. They include intertrochanteric and subtrochanteric fractures and make up about 50% of hip fractures. Subtrochanteric fractures pose a challenge to surgeons because of their anatomical peculiarity. This is an area of great stress concentration and is subjected to several deforming forces due to its muscular insertions. Complex fractures with medial support failure more often lead to fixation failure and reoperation. 1.2

In addition to the obstacles faced in obtaining an anatomic reduction, the surgeon must maintain the reduction throughout the healing process using an appropriate fixation device.<sup>3</sup> Given the shorter lever arm and load-sharing characteristics of IM nails, they are the most commonly used

eISSN 2012-3264 (Online)
Printed in the Philippines.
Copyright© 2024 by Condor and How.
Received: October 3, 2023.
Accepted: March 3, 2024.
Published Online: March 15, 2024.
https://doi.org/10.69472/poai.2024.04

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\*This study earned 3<sup>rd</sup> Place at the POC 25<sup>th</sup> Residents Research Forum on February 18, 2022. It was a presented at a podium during the Philippine Orthopaedic Association Annual Convention on June 16, 2022 and as an e-poster at the 5<sup>th</sup> AO Trauma AP Scientific Conference on May 28, 2022. devices in unstable fractures. One type of cephalomedullary device is the Proximal Femoral Nail Antirotation (PFNA). The Proximal Femoral Nail is a promising minimally invasive implant, with better biomechanical stability, minimum soft tissue dissection, minimal blood loss, minimal infection, and wound complications. 4,5 This surgical treatment aims to provide stable fixation that allows an early range of motion. However, proper placement of the blade is essential to avoid the risk of screw or blade cut-out. The tip of the blade should be 10 mm from the joint line in the anteroposterior and lateral projections. This corresponds to a tip-apex distance (TAD) of 20 mm. A study by Nikoloski et al., suggested that the optimal TAD for a PFN is between 20 to 30 mm. Another way of assessing the quality of implant placement is the Cleveland index. This assesses the position of the compression screw of a PFN and the helical blade of a PFNA. A center-center or center-inferior placement of the compression screw or helical blade is considered optimal.6

The Harris Hip Score (HHS) was developed to assess functional outcomes following hip surgery and is intended to evaluate various hip disabilities and treatment methods in an adult population. The domains covered are pain, function, absence of deformity, and range of motion. The pain domain measures pain severity and its effect on activities and the need for pain medication. The function domain consists of daily activities (stair use, using public transportation, sitting, and managing shoes and socks) and gait (limp, support needed, and walking distance). Deformity considers hip flexion, adduction, internal rotation, and extremity length discrepancy. Range of motion measures hip flexion, abduction, external and internal rotation, and adduction.

It is also essential to assess the bony union objectively. The University of Toronto and McMaster University teams developed the radiographic union score for hip (RUSH) which increases agreement among surgeons and radiologists in assessing fracture repair. The RUSH is a standardized radiographic assessment of hip fracture union based on callus bridging and the appearance of the fracture line. It may provide prognostic information that could predict healing outcomes in patients with hip fractures.<sup>7</sup>

Adequate reduction and fixation are crucial to achieving ideal function and radiographic healing post-operatively. Many factors can contribute to the ease of reduction; one of these is the positioning technique. Numerous reports have addressed the advantages and disadvantages of the positioning techniques for antegrade femoral nailing. The supine position on a fracture table offers sustained longitudinal traction without the need for an assistant, and circumferential access to the injured extremity. However, it is difficult to establish the correct starting point and to accommodate obese patients and patients with multiple injuries. Complications include pudendal nerve neuropraxia, erectile dysfunction, perineal sloughing due to continuous traction, pressure necrosis, and compartment syndrome of the opposite leg. A study by Ganjale et al., stated that it was difficult to reduce comminuted

subtrochanteric fractures (where different fragments and segments are being pulled by strong muscles around the hip) in the supine position. This caused prolonged operative time, higher risks of conversion to open technique, more bleeding, higher chances of infection, and longer anesthesia time.<sup>10</sup>

In contrast, in the lateral position, the muscles around the hip are relaxed, and the distal limb is free for easier mobilization. Reduction and fixation of proximal femoral fractures in the lateral position with fluoroscopy in rural hospitals that lack a fracture table may be executable and probably safe. <sup>10</sup> Aside from this, the lateral position allows improved access to both the piriformis fossa and the trochanteric entry points in obese patients and allows conversion to an open approach. Also, there was no significant difference in the functional outcome and out-of-bed activity time in the lateral position compared to the supine position. <sup>8</sup>

The Proximal Femoral Nail Antirotation (PFNA) is widely used in our setting. To our knowledge, there are no local studies published about peri-trochanteric fractures treated with PFNA in the lateral decubitus position, and the choice of position is solely surgeon-based. There have been international studies concerning the complications brought about by the supine position. The lateral decubitus position does not require a fracture table, (so it can be used in primary hospitals), avoids additional set-up time, and is easier to convert to open reduction.

#### **OBJECTIVES**

The objectives of this study were to compare the quality of fixation (tip-apex distance and Cleveland index), adequacy of reduction, bony union, and functional outcomes in PFNAs done in the lateral decubitus position and supine position. We also aimed to describe the incidence of complications and difficulties (malreduction, non-union, screw malrotation, difficulty in establishing an entry point, pudendal nerve neuropraxia, perineal sloughing, and compartment syndrome of the contralateral leg, helical blade cut-out) encountered.

#### **METHODOLOGY**

This study was a retrospective cohort conducted at the Philippine Orthopedic Center (POC). Convenience sampling through records review of the patient census was done to acquire a sample of patients with peri-trochanteric fractures (from trauma and adult orthopedic services at the Philippine Orthopedic Center) who underwent open reduction and internal fixation with PFNA between January 01, 2018 and December 31, 2020. The participants were nonrandomly assigned to two groups – lateral or supine position – as the attending physician saw fit. A total sample size of at least 18 patients (n = 17.48), 9 patients for each treatment group, was calculated based on a 5% level of significance (95% level of confidence) and a 6.4% coefficient of variation, with a margin of error of at most 0.11 Specifically, the sample size, n, was computed using the following formula:

$$n = [(Z_{\alpha/2} CV)/\epsilon]^2$$

where  $Z_{\alpha/2}$  is the tabular value at alpha level of significance; CV is the coefficient of variation (usually <0.10);  $\epsilon$  is the margin of error.

After IRB approval, we included patients based on the following criteria: 1) adults (21 to 65 years old), 2) patients who underwent open reduction PFNA either in the supine or lateral position, and 3) patients with unstable type intertrochanteric fractures with or without subtrochanteric extension and subtrochanteric fractures (AO/AOTA 31A2, 31A3, 32A1, 32A2, 32A3 and 32B2). Patients with significant co-morbid conditions (American Society of Anesthesiologists Physical Status classification III–V), open peri-trochanteric fractures, polytrauma patients, and patients with pathologic fractures were excluded from this study. Thirty-nine (39) patients were included in the study, where six were lost to follow-up, and four did not have any retrievable postoperative radiographs on the picture archiving and communication system (PACS). A total of 29 patients were included.

Demographic characteristics were taken, such as age, gender, height, weight, BMI, comorbidities, the timing of surgery, injured side (laterality), intraoperative blood loss, and intraoperative time.

#### Surgical technique

Patient position and reduction of the fracture:

#### Supine positioning

The patient was positioned supine on a fracture table. The ipsilateral arm was elevated in a body strap or taped to the trunk while the uninjured leg was secured on a leg holder. The torso was pushed 10° to 15° to the contralateral side to ensure that the ipsilateral hip was in an adducted position

(Figure 1). After positioning, the surgical site was prepared aseptically, and sterile drapes were then applied. The fracture was exposed, reduced, and fixed using a direct lateral approach.

#### Lateral positioning

The patient was positioned in a lateral position on a radiolucent top operating table, with the fractured limb on top and freely movable at the hip. Trunk-supporting bolsters were placed anteriorly and posteriorly and were well secured with body straps to stabilize the patient in a lateral position, (Figure 2A). The C-arm was placed contralateral to the surgeon (Figure 2B). To avoid any bony overlap on the lateral view, the contralateral hip was maximally flexed with the knee in 90 degrees of flexion. After positioning, the surgical site was prepared aseptically, and sterile drapes were then applied. The fracture was exposed (Figure 3), reduced (Figure 4), and fixed using a direct lateral approach.

#### Insertion of the nail

The C-arm was used to guide nail insertion (Figure 5). In the AP view, the nail insertion point was on the tip or slightly lateral to the tip of the greater trochanter. The guidewire was inserted laterally at an angle of 6° to the shaft. In the lateral view, the guidewire was placed in the center of the medullary canal to a depth of about 15 cm.

A cannulated drill bit was used over the guidewire to open the entry point; reaming was done manually through the protection sleeve. The nail was then inserted manually. The guidewire for the helical blade was then inserted superomedially, using the C-arm for positioning. The final position of the guidewire was at the inferior part of the femoral neck. In lateral view, the wire was positioned in the center of the femoral neck. The correct screw length was indicated on the measuring device and calculated to end approximately 5 mm before the tip of the guidewire. A hole was drilled, and the femoral neck helical blade was inserted (Figure 6). Distal





**Figure 1.** Supine position. **(A)** Patient placed on a fracture table with both leg secured with contralateral hip flexed and abducted. **(B)** Placement of the perineal post.





Figure 2. Lateral positioning with placement of a bolster to secure the body (A); contralateral hip maximally flexed (under the green plastic drape) and positioning of the C-arm (B).



**Figure 3.** Direct lateral approach preoperative surgical markings.

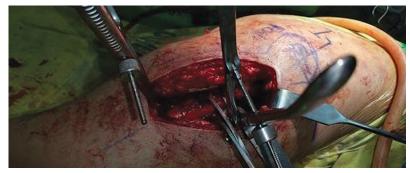


Figure 4. Direct reduction of the fracture using bone clamps.

locking screws were then placed. After proper placement of the nail, copious washing and hemostasis were done. Surgical wounds were primarily closed, and a dressing was applied.

Outcome measures (TAD, Cleveland index, adequacy of reduction, bone union, and functional outcome at six months post-operatively) were recorded and compared.

#### Quality of fixation

The quality of fixation was assessed using the tip-apex distance for PFN described by Nikoloski et al., and the Cleveland index by Cleveland et al. They suggest that a tip-apex distance for helical blade-based proximal femoral nails should be 20 to 30 mm. A TAD of less than or equal to 20 results in a possible axial cut-out (medial migration), and a TAD of more than or equal to 30 mm results in a cephalad cut-out. The Cleveland index was used to assess the position of the compression screw in PFN and the helical blade in PFNA. A center-center or center-inferior placement of the compression screw or helical blade was considered optimal.<sup>12</sup>

#### Adequacy of reduction

Adequate reductions were defined as displacements less than 5 mm and angulations deviating less than 10 degrees from the normal neck-shaft angle. <sup>13</sup>

**Table 1.** Demographic and clinical characteristics of patients by treatment group

Characteristics	Lateral decubitus p	osition	Supine posit	ion
Characteristics	Mean ± SD / n	%	Mean ± SD / n	%
Sex, n (%)				
Male	9	69.2	14	87
Female	4	30.8	2	12
Age, Mean ± SD	50.77 ± 13		38.69 ± 11	
Height, Mean ± SD	160.08 ± 8		167.06 ± 10	
Weight, Mean ± SD	64.00 ± 10		61.94 ± 10	
BMI, Mean ± SD	24.77 ± 2		22.25 ± 2	
Comorbidities, n (%)				
Normal health	9	69.2	9	5
Mild systemic disease	3	23.1	5	3
Diabetes	1	7.7	1	6.3
Missing	0	0	1	6.3
Intraoperative time, Mean ± SD	158.62 ± 51		171.6 ± 60	
Intraoperative blood loss, Mean ± SD	808.46 ± 410		785.6 ± 742	

**Table 2.** Average scores of quality of reduction and functional outcome

	Lateral Mean ± SD	Supine Mean ± SD
Tip-apex distance (mm)	21.69 ± 3.5	22.25 ± 7.5
Harris hip score (points)	92.24 ± 5.2	94.70 ± 3.4

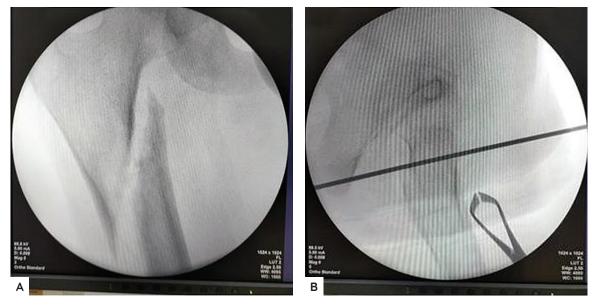


Figure 5. Intraoperative radiographs (Lateral position), AP view (A) and lateral view (B).

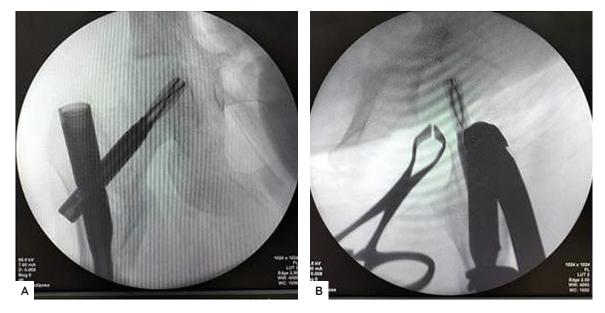


Figure 6. Final placement of the helical blade on AP (A) and lateral view (B) (lateral group).

Table 3. Distribution of categorical outcomes

Catagorian autoomos	Lateral ded	cubitus position	Supine	position
Categorical outcomes	Count	Percentage (%)	Count	Percentage (%)
TAD				
Acceptable	9	69.2	7	43.8
Not acceptable	4	30.8	9	56.3
Cleveland Index				
Optimal	13	100.0	9	56.3
Sub-optimal	0	0.0	7	43.8
Adequacy of reduction				
Acceptable	12	92.3	11	68.8
Not Acceptable	1	7.7	5	31.3
Bone union (RUSH score)				
Complete bony union	10	76.9	15	93.8
Inadequate bony union	3	23.1	1	6.3

Table 4. Result of T-test and Fisher's exact test

Outcomes	Test	Р	Interpretation
Tip- apex distance	Fisher's exact test	0.2642	Not significant
Cleveland index	Fisher's exact test	0.0084	Significant
Harris hip score	T-test	0.0001	Significant
Adequacy of reduction	Fisher's exact test	0.1834	Not significant
Bone union (Rush score)	Fisher's exact test	0.2994	Not significant

#### Bony union at six months post-op

The RUSH score assesses four component scores of cortical bridging, cortical disappearance, trabecular consolidation, and trabecular disappearance. The four cortices (anterior, posterior, medial, lateral) were each given a score from 1 to 3 for cortical bridging, and a score from 1 to 3 for cortical fracture disappearance. Trabecular consolidation and trabecular disappearance were each given a score of 1 to 3. The maximum score is 30 (perfect healing) and the minimum score is 10 (no signs of healing).<sup>7</sup>

#### Functional outcome six months post-op

Functional outcome was assessed using the Harris Hip score at six months post-op. A score of <70 indicates a poor outcome, 70–79 fair, 80–89 good, and 90–100 excellent.

Complications encountered during the follow-up period such as infection, non-union, implant failure, and pudendal nerve injury were documented.

#### Statistical analysis

Descriptive statistics such as counts, percentages, means, and standard deviations were used to describe patients' demographic and clinical characteristics in each treatment group. After the data for each population were gathered, the data were analyzed using the T-test procedure in two population means and Fisher's exact test assuming that the data gathered follows a normal distribution.

#### **RESULTS**

Most patients in both groups were male (70% for the lateral group, and 88% for the supine group). On average, patients

in the supine group were younger (mean, 39 years old vs 51 years old), slightly taller, weighed slightly less, and had slightly lower BMIs. Few patients had mild systemic disease. The mean intraoperative time was 158.62 minutes for the lateral group, while it was 171.63 minutes for the supine group. Intraoperative blood loss was higher for the lateral group, albeit with high variability. One patient in the supine group experienced pudendal nerve palsy postoperatively that completely resolved after three months.

The results showed no significant differences in the average TAD scores for the supine group (21.69 mm) compared to the lateral group (22.25 mm) (Table 2). However, a higher percentage of patients in the lateral group had an acceptable TAD (69.2% vs 43.8%, p=0.2642), Cleveland index (100% vs 56.3%, p=0.0084), and adequacy of reduction (92.3% vs 68.8%) compared to the supine group (Table 3). The supine group had higher RUSH scores (93.8% vs 76.9%, p=0.2994) and Harris Hip scores (p=0.0001).

#### **DISCUSSION**

Davis et al. in 1969 used the lateral position to facilitate reduction and exposure for the first time in intertrochanteric and subtrochanteric fractures of the femur. Ozkan et al. used this position for proximal femoral nailing in 2010, and Connelly et al., for complex proximal femur locked plating in 2012. In our study, the lateral position group had shorter intraoperative times. This is consistent with other studies that reported difficulties in reducing the fragments in supine on a fracture table. The difficulties were attributed to the pull of strong muscles around the hip, specifically in subtrochanteric fractures. In the lateral position, all the muscles around the hip are relaxed, and the affected limb is freely draped and

movable. Muscular forces around the hip in the sagittal plane are more effectively neutralized in the lateral position, whereas the coronal plane forces are easily neutralized with a firm pillow between the legs. <sup>13</sup> Although placing the patient on a fracture table gives the advantage of sustained traction with less manpower, the lateral position offers the advantage of moving the distal segment to align with the proximal freely with gentle longitudinal traction. In contrast to other studies, our lateral position group had more intraoperative blood loss. This may be due to the difficult reduction in chronic fractures, requiring a longer incision for better exposure.

This study had eight overweight patients managed in the lateral position and two in the supine position. A higher average BMI was reported in the lateral group, as this position may have been chosen by the surgeon to facilitate nail insertion. In the supine position, it is difficult to insert a nail and jig assembly for obese patients or patients with a sagging abdomen.

There were no significant differences in the TAD, adequacy of reduction, and six-month bony union. In this study, no incident of cut-out was recorded, even in patients with unacceptable TAD. We noted significant differences in the Cleveland index and Harris Hip Scores. All patients in the lateral position group had a center-center or center-inferior helical blade placement. Despite the significant difference in terms of the HHS, the average score of each group was >90, which was excellent. There was also one reported case of pudendal nerve palsy in the supine position group due to a prolonged intraoperative time due to the perineal pressure on the fracture table.

#### **CONCLUSION**

The preferred surgical position for open reduction PFNA remains controversial. Regardless of the position, the main goal is still to achieve a good reduction with stable fixation and early return to pre-morbid function. This study shows that the surgical position did not affect the TAD, adequacy of the reduction, and bony union. All patients from both groups had excellent functional outcomes at six months postoperatively, but the lateral position group was superior in terms of the Cleveland index.

Performing PFNA in the lateral decubitus position can be safely and effectively done in small rural hospitals without a fracture table, potentially benefiting more patients with peritrochanteric fractures.

Limitations of this study include the lack of follow-up for most patients, lack of randomization, different senior surgeons, and a short follow-up period. We also did not consider the fracture configuration in our analysis. A randomized controlled trial

with an increased sample size is recommended to strengthen the power of this study.

#### STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

#### **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

#### **FUNDING SOURCE**

None.

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#### Cross-cultural Adaptation and Validation of the International Knee Documentation Committee Subjective Knee Form in Filipino\*

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#### **ABSTRACT**

**Objectives.** The primary objectives of this study were to perform cross-cultural adaptation and to determine the validity and reliability of the Filipino IKDC Subjective Knee Form.

**Methodology.** The IKDC subjective knee form was translated and cross-culturally adapted to Filipino. The process consisted of a forward translation, backward translation, review of versions by an expert committee, modification of the Draft Filipino version, and pre-testing of the Initial Filipino version. The clinimetric properties of the final Filipino IKDC subjective form, namely face and content validity, construct validity, internal consistency and test-retest reliability were measured from 101 respondents. Face and Content validity were explored with questionnaires, internal consistency was assessed with Cronbach Alpha, test-retest reliability was measured with interclass correlation coefficients, and construct validity was analyzed by comparing scores between respondents with injured knees with respondents with healthy knees.

**Results.** The Filipino version of the IKDC subjective form has internally good clinimetric properties. Content validity showed that all items of the Filipino Version of the IKDC subjective form were perceived to be highly relevant by experts. Face validity showed that the participants graded the questionnaire items as easy to understand and relevant to their condition. Internal consistency, test-retest reliability and were interpreted as excellent and acceptable, with an ICC value of 0.99 and Cronbach's Alpha of 0.702, respectively.

**Conclusion.** The Filipino IKDC subjective knee form has good face validity, content validity and is a reliable patient reported outcome measure for knee function.

Keywords. Filipino, IKDC, translation, validation, PROM

#### INTRODUCTION

The International Knee Documentation Committee Subjective Knee Form has been validated, developed, and documented in literature as a valid patient-reported outcome measure (PROM) to evaluate ligamentous and meniscal knee injuries. <sup>1-4</sup> It studies the domains of pain, activities of daily living, and sports and/or recreation. <sup>2</sup> In the past decade, there have been many studies validating translated IKDC subjective forms from various countries. Each of their respective authors cross-culturally adapted and performed validity and reliability testing on their respective translated IKDC subjective form. <sup>5-15</sup>

According to Beaton et al., cross-cultural adaptations and translation of established PROMs made for use in the English language should be done in certain scenarios. <sup>16</sup> Specifically, for the Philippines, due to the change in language, culture, and country of use, a translation and cross-cultural adaption is warranted for the IKDC subjective. A cross-cultural adaptation and translation ensure that the translated version of the scale is linguistically and culturally accepted; this,

eISSN 2012-3264 (Online)
Printed in the Philippines.
Copyright© 2024 by Veloso and Arandia.
Received: April 7, 2024.
Accepted: May 15, 2024.
Published Online: May 21, 2024.
https://doi.org/10.69472/poai.2024.11

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<sup>\*</sup>The study was presented at the: (1) Philippine Orthopedic Center 2023 Interdepartmental Research Paper Contest on August 11, 2023, and won first prize; (2) Philippine Orthopaedic Association Free Paper Session of the 74th POA Annual Congress on November 18, 2023; and (3) Philippine Orthopedic Center 29th Residents Research Forum on December 12, 2023, and won first prize.

however, does not correspond to the validity and reliability of the translated PROM. <sup>16-19</sup> The primary objective of this study was to perform cross-cultural adaptation and to determine the clinimetric properties of the Filipino International Knee Documentation Committee Subjective Knee Form. Face and content validity, construct validity, internal consistency, and test-retest reliability were determined.

#### **METHODOLOGY**

#### Study design

This was a translation and validation study that examined the clinimetric properties of the created Filipino Version of the IKDC subjective knee form. The study was conducted prospectively at the Philippine Orthopedic Center from January 2020 to November 2022. The American Orthopedic Society for Sports Medicine permitted usage of the IKDC without license or agreement for the purpose of research.

#### Translation and cross-cultural adaptation phase

As described in the paper of Beaton et al., five general steps were followed to create the culturally adapted Filipino Version of the IKDC subjective form.<sup>16</sup> In the forward translation phase, two bilingual Filipinos drafted separate Filipino IKDC subjective forms, which were consolidated to create the initial Filipino IKDC subjective form. A separate professor from the Department of Filipino and Philippine Literature, University of The Philippines, Diliman, then performed a back translation of the initial Filipino IKDC subjective form. The initial Filipino IKDC subjective form and the backtranslated initial Filipino IKDC subjective form were reviewed by an expert committee consisting of three consultants from the Sports Section of the Philippine Orthopedic Center, the Assistant Chairperson and a Professor from The Department of Filipino and Philippine Literature, University of The Philippines, Diliman, and a Methodologist. The expert committee reviewed the previous drafts to create the Final Draft of the Filipino IKDC subjective form.

#### Pre-testing and modification phase

Fifteen patients with ligamentous knee injuries from the Philippine Orthopedic Center and three Rehabilitation Medicine Consultants from the Philippine Orthopedic Center took part in the pre-testing of the Draft of the Filipino IKDC subjective knee form. Each of these respondents was asked to interpret each item's difficulty and comprehensibility. The data collected from the pre-testing was reviewed and consolidated by a methodologist. The expert committee reviewed this data and modified the Filipino IKDC subjective knee form before utilization in the validation phase of the study.

# Validation and reliability testing phase: patients and procedures

Twenty-four patients diagnosed with ligamentous knee injuries were recruited for face validity testing. Participants

were asked to rate items of the Filipino IKDC subjective form in terms of *difficulty to understand* and *relevance to their condition*. To tackle content validity, 10 Orthopedic surgeons from the Philippine Orthopedic Center reviewed the Filipino IKDC subjective form. Each expert rated item relevance, comprehensiveness, and comprehensibility. A 4-point Likert scale was used to rate item relevance, while comment sections were available for comprehensiveness and comprehensibility.

A total of 67 respondents participated in the construct validity and reliability testing phase of the study. Forty-nine respondents had ligamentous knee injuries diagnosed by the Sports Section of The Philippine Orthopedic Center and 18 were healthy respondents. These 49 respondents were not part of the previous phases of this study. All respondents answered the Filipino IKDC subjective knee form and then answered the same form again five to seven days later.

A minimum of 14 patients, seven with injured and seven with healthy knees, were required to explore the discriminative validity based on a level of significance of 5% and a power of 80% with an absolute effect size of 1.3.<sup>20</sup> To adequately explore test-retest reliability, a minimum of 41 patients were required with a level of significance of 5% and a power of 90% with an interclass correlation coefficient (ICC) of 0.87.<sup>5,21</sup> Lastly, a minimum of 53 patients were required to explore internal consistency based on a level of significance of 5% and a precision of 0.05 with Cronbach alpha of 0.89.<sup>7,8,22</sup>

#### Statistical analysis

Descriptive statistics were used to summarize the general and clinical characteristics of the participants. Frequency and proportion were used for categorical variables (nominal/ordinal), mean and standard deviation for normally distributed interval/ratio variables, and median and range for non-normally distributed interval/ratio variables. The ICC was used to determine the test-retest reliability of the IKDC subjective knee form. To assess the questionnaire's internal consistency, Cronbach's Alpha was utilized. Mann-Whitney U test and Chi-square test were used to determine the difference in mean, median, and frequency between injured and healthy participants, respectively. All valid data was included in the analysis. Missing variables were neither replaced nor estimated. The Null hypothesis was rejected at  $0.05\alpha$ -level of significance. R version 4.2.2 was used for data analysis.

#### **RESULTS**

# Translation and cross-cultural adaptation, pre-testing

Cultural adaptation suggestions from the expert committee were incorporated for items 1, 5, 7, and 8 of the Filipino IKDC. In the physical activities included in the second choice, Skiing and Tennis were changed to "pagbubuhat ng sako" and "pagbibisikleta" which translate to carrying a sack and riding a bicycle respectively.

During the pre-testing and initial face and content validity testing, specifically for items 2 and 3 of the IKDC, participants had a hard time quantifying their subjective knee pain on a scale of 1–10. Thus, under the Likert scale in the choice section of these items, phrases were added to aid the participant to quantify each aspect the of knee symptoms the item was asking them to grade. Although item 4 seemed straightforward in its English form, in the pretesting phase, participants had difficult time differentiating between the initial choices "Wala/Kaunti/Katamtaman/Napakatigas/Labis-labis" The choices were changed to "walang paninigas o pamamaga, may kaunting nararamdamang paninigas o pamamaga, may paninigas o pamamaga ng tuhod, sobrang maga o sobrang tigas ang tuhod, hindi maigalaw and tuhod dahil sa pamamaga o paninigas."

#### **Content validity**

Four items of the Filipino IKDC were rated highly relevant by all experts, while the remaining items were rated highly relevant to quite relevant. None of the experts had any comments on Comprehensibility. For Comprehensiveness, comments on the items were regarding translation. The experts who participated in this study phase gave suggestions to improve the comprehensibility of each item.

#### **Face validity**

For item difficulty, the majority of the items were deemed easy to understand. The majority of the items were also deemed important to the respondent's knee condition.

#### Test-retest reliability and internal consistency

The Filipino IKDC subjective knee form had excellent reliability with a computed interclass correlation coefficient (ICC) of 0.998. With a computed Cronbach's alpha of 0.7026, the IKDC Filipino subjective knee form had acceptable internal consistency.

#### Survey proper and construct validity

There were 67 respondents for the survey, 49 (73.13%) with injured knees and 18 (26.9%) with normal knees. The two groups were similar in terms of age and sex (Table 1). The IKDC subjective knee form score was able to discriminate

between the two groups, with median scores of 51.72 for the injured group and 100 for the normal group (p <0.001).

#### **DISCUSSION**

The Filipino version of the IKDC subjective form has internally good clinimetric properties. Content validity showed that all items of the Filipino Version of the IKDC subjective form were perceived to be highly relevant by experts. Face validity showed that the participants graded the questionnaire items as easy to understand and relevant to their condition. Internal consistency and test-retest reliability were interpreted as excellent and acceptable, respectively.

# Translation, cross-cultural adaptation and pre-testing

The authors of the paper worked closely with the expert committee in creating the Final Version of the IKDC Filipino Subjective Knee form. This allowed comments and suggestions from all phases of the study to be reviewed and consolidated into each subsequent version. Prior to administering the final IKDC subjective in Filipino in the reliability testing phase, items were shortened or rephrased to make each item simpler to understand.

The original phrase from the second choice of the United States, English IKDC subjective knee form for items 1,5,7 and 8 is "Strenuous activities like heavy physical work, skiing or tennis." The expert committee as well as respondents from the pre-testing phase of this study shared the sentiments that tennis and skiing are unfamiliar sports to the demographic of respondents, that the Filipino IKDC subjective knee form is supposed to cater to. Instead of substituting tennis and skiing with other sports, the created item choice "Nakakapagod na aktibidad gaya ng mabibigat na trabaho, tulad ng pagbubuhat ng sako o pagbibisikleta" which tried to focus activities deemed as heavy physical work and strenuous.

#### Face and content validity

Most participants from the face validity phase found the items comprehensible and relevant to their condition. The items rated as "difficult to understand" were the lengthier items of the IKDC which required more time to read.

 $\textbf{Table 1.} \ \textbf{Profile of respondents and hypothesis testing validity with survey proper}$ 

	Total (n=67)	Injured Knee (n=49)	Normal Knee (n=18)	
	Frequency (%); Median (IQR)			P
Age, years	30 (27-31)	29 (26-31)	31 (30-31)	0.064§
Sex Male Female	46 (68.66) 21 (31.34)	36 (73.47) 13 (26.53)	10 (55.56) 8 (44.44)	0.270†
Injured knee [n=49] Left Right	27 (55.10) 22 (44.90)	27 (55.10) 22 (44.90)	-	-
IKDC subjective knee form score [n=116]	67.816 (47.989-94.253)	51.724 (37.931-69.540)	100 (97.70-100)	<0.001⁵

Statistical tests used:  $\S$  – Mann-Whitney U test;  $^\dagger$  – Chi-square test

The orthopedic surgeons who participated in the content validation phase graded all the items of the Filipino Version of the IKDC subjective form as highly relevant for evaluating ligamentous knee injuries. The majority of the comments focused on the correct phrasing of the questions and responses. The types of comments made by the experts were grouped into two main categories: appropriate wording to describe knee symptoms and syntax. One respondent commented using "pagkalas ng tuhod" instead of "pagbigay ng tuhod." The expert committee discussed how to phrase the term "pagbigay ng tuhod" (originally "giving way in your knee" in English) and agreed that it was an appropriate translation. The other comments on the syntax were reviewed by the Assistant Chairperson of The Department of Filipino and Philippine Literature, University of The Philippines, Diliman.

The changes incorporated into the Final Filipino IKDC subjective form prior to reliability and construct validity testing are enumerated below. For item 6, the choices were changed from "oo" and "hindi" to "mayroon" and "wala". The questions for items 1, 5, and 7 were simplified to "Ano ang pinakakaya mong gawin na hindi sumasakit ang tuhod," "... nang hindi namamaga and tuhod," and "... nang hindi bumibigay ang tuhod," respectively. For question 10, the phrase "paano mo titignan" was changed to "paano mo mamarkahan."

#### Reliability and construct validity

Internal Consistency of the Filipino IKDC subjective form was acceptable with a computed Cronbach's alpha of 0.7026. The study by Crawford et al., investigating the internal consistency of the Original IKDC reports a Cronbach's alpha of 0.77.<sup>23</sup> More recent studies that performed cross-cultural adaptations and validity studies report values ranging from 0.89 to 0.97.<sup>5-15</sup> These include studies from China, the Netherlands, Brazil, Thailand, Italy, South Korea, Indonesia, Greece, and Turkey.

The test-retest reliability of the Filipino IKDC subjective knee form was found to be excellent with an ICC of 0.99. This value is comparable with the Brazilian and Indonesian IKDC versions having an ICC of 0.99, the Dutch IKDC's ICC of 0.96, and the Original IKDC's ICC of 0.95. 6.11,22

A study by Anderson et al., collected normative data for the IKDC form for around 5000 knees in the United States of America; 28% of respondents had injured knees while the remainder did not have knee problems. <sup>24</sup> This study reported that respondents with a current unilateral knee problem, current treatment, or history of knee surgery had lower scores than respondents without a history of knee problems. This proves the construct validity of the IKDC subjective knee form since it can differentiate between patients with healthy knees and patients with injured knees. <sup>24</sup>

Data from this current study is similar to the data collected by Anderson et al., in 2006.<sup>24</sup> In this current study, patients with knee injuries also had lower scores than patients with healthy knees. The Filipino IKDC subjective knee form score was able to discriminate between the two groups, with median scores of 51.72 for the injured knees group and 100 for normal knee patients (p <0.001).

#### RECOMMENDATIONS AND LIMITATIONS

Although the scores between healthy and injured knees were significantly different, it only *suggests* that the Filipino IKDC has good construct validity because there is no normative data available. According to Mokking et al., to explore the construct validity of a PROM, hypothesis testing regarding "internal relationships, relationships to scores of other instruments, or differences between relevant groups" should be carried out.<sup>25</sup>

Most IKDC translation and validation studies noted the lack of a "gold standard" questionnaire to measure knee function. They translated and validated knee PROMs in their native languages to evaluate the corresponding IKDC being validated. The Amajor limitation of this study is that the knee-related PROMs used in foreign studies do not have Filipino versions.

The Medical Outcomes Study 36-Item Short-Form Health Survey (SF-36), although not a knee-specific PROM, was also used to measure the construct validity of the IKDC subjective knee form. The Filipino Version of the SF-36, while not accessible to the author at this time, could be licensed and used in the future to further investigate the Filipino IKDC subjective knee form.

Aside from determining the clinimetric properties of the Filipino IKDC as presented above, according to Mokking et al., responsiveness should also be measured. Page 325 Responsiveness is defined as the ability to detect change over time in the construct to be measured. To measure the responsiveness of the Filipino IKDC subjective knee form, it should also be administered to patients before and after ACL reconstruction surgery.

#### CONCLUSION

The Filipino IKDC subjective knee form developed in this study has good face validity, good content validity and is a reliable PROM to evaluate knee pain and function of Filipino patients with ACL injuries. Further research in responsiveness and construct validity can be done.

#### STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

#### **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

#### DATA AVAILABILITY STATEMENT

The data supporting the findings of this study are available upon request from the corresponding author.

## **FUNDING SOURCE**

None.

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# Characteristics, Treatment Patterns, and Clinical Outcomes of Patients Diagnosed with Fungating Soft Tissue Sarcomas\*

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#### **ABSTRACT**

**Objectives.** A fungating soft tissue sarcoma of the extremities (F-ESTS) is a rare clinical presentation with limited literature. This study aims to describe the important clinical characteristics, treatment patterns, and outcomes of twenty patients diagnosed with F-ESTS and treated by a single surgeon at a sarcoma unit.

**Methodology.** We conducted a retrospective clinical study on twenty F-ESTS patients treated by a single surgeon at a sarcoma unit over 25 years (1993–2018).

**Results.** The local incidence of F-ESTS was 10.9%. The mean age of patients was 49.2 years old. The most common site of occurrence was the thigh (50%) with an average size of 11.8 cm. Most tumors were deep (65%) and high grade (85%). Liposarcomas were the most common histologic diagnosis (35%). Limb salvage was done in 60% of the patients with 50% requiring reconstructive procedures. Fifteen percent of patients developed complications, 25% had local recurrence, and 65% developed distant metastases. The mean survival for this cohort was 49.2 months. Sixty percent of patients died of disease.

**Conclusion.** The majority of F-ESTS patients were younger than 65 years old, had deep and high-grade tumors, predominantly liposarcomas, most commonly found in the thigh, and had a history of surgery or biopsy. In the last 10 years, limb salvage surgery has become the treatment of choice even for patients with fungating sarcomas. Most F-ESTS patients in our study were still able to undergo limb salvage surgery. Local recurrence was seen in five (25%) patients, while thirteen (65%) patients had distant metastases. Twelve (60%) patients had died of disease.

**Keywords.** soft tissue sarcoma, fungating; limb salvage, neoplasm recurrence, local, neoplasm metastasis, neoplasm seeding, neoplasm staging, neoplasm, residual, neoplasm invasiveness

## **INTRODUCTION**

Soft tissue sarcomas (STS) are a rare and heterogeneous group of tumors of mesenchymal origin, occurring most commonly in the extremities (ESTS) followed by the trunk. The heterogeneity of STS makes it difficult to clinically confirm the malignant potential of the mass. Locally, there is a 4.7/100,000 incidence of ESTS in adults with 3500 cases per year. Treatment modalities for ESTS include surgical resection and adjuvant radio- or chemotherapy. The prognosis of ESTS depends on several patient, tumor, and treatment variables such as age, metastasis at diagnosis, tumor size, tumor site, depth, tumor grade, and surgical margins. Recently, the presence of fungation or malignant ulceration in an ESTS (F-ESTS) is a negative predictor for survival.

Cutaneous involvement from a soft tissue sarcoma is rare and is often seen in patients with primary carcinomas like breast cancer and melanoma.<sup>4</sup> The tumor can erode through the dermis and communicate to the skin surface which is

eISSN 2012-3264 (Online) Printed in the Philippines. Copyright© 2024 by Araneta et al. Received: February 15, 2024. Accepted: May 10, 2024. Published Online: May 23, 2024. https://doi.org/10.69472/poai.2024.12

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\* Presented at 73<sup>rd</sup> Philippine Orthopaedic Association Convention Podium on June 16-18, 2022.

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called malignant ulceration or simply fungation, wherein the tumor grows like a fungus.<sup>4,5</sup> The clinical burden of F-ESTS has not been extensively discussed in the literature due to its rarity. Currently, only three studies have analyzed the impact of F-ESTS on patient outcomes; Potter et al., in the United States; Parry et al., in the United Kingdom, and Okajima et al., in Japan. However, Within Asia, despite the prevalence of more advanced disease owing to geopolitical and cultural factors, there are fewer outcome-based data as emphasized by Ngan et al., in their systematic review of Soft-tissue Sarcomas in the Asia-Pacific Region (STAR) in 2015.<sup>6</sup>

This study described the important clinical characteristics, treatment patterns, and outcomes of a local study of twenty patients diagnosed with F-ESTS and treated by a single surgeon at a sarcoma unit.

## **Significance**

The presence of fungating or ulcerating lesions in soft tissue sarcoma has been associated with a high tumor grade and may increase the possibility of contaminated surgical margins or inadequate resection. The limited literature on this subgroup of patients has described abysmal survival rates for F-ESTS patients.<sup>5</sup>

From our own experience, these patients usually have been managed poorly or have had a delayed course of consult. By observing the clinical characteristics and treatment patterns in fungating sarcomas, surgeons can find room to improve the decision-making process for these patients to maximize survivorship and limit morbidity. Physicians can benefit by applying this knowledge to the urgent and rational management of such cases.

## **OBJECTIVES**

## General objective

This study aimed to describe the clinical characteristics, treatment patterns, and outcomes of F-ESTS patients treated by the senior investigator.

## **Specific objectives**

- 1. Clinical characteristics of F-ESTS patients were described in terms of:
  - a) Age
  - b) Gender
  - c) Tumor site (Upper, lower extremity)
  - d) Tumor size
  - e) Tumor depth (subcutaneous or subfascial)
  - f) Tumor grade
  - g) Tumor histology
  - h) Metastases at presentation to sarcoma unit
  - i) History of prior biopsy and/or treatment

- 2. The treatment of F-ESTS patients was described in terms of:
  - a) Surgical treatment
    - i) Limb salvage
    - ii) Amputation
  - b) Surgical margins
    - i) Radical
    - ii) Wide
  - c) Margin status
    - i) R0
    - ii) R1
  - d) Radiotherapy and Chemotherapy (Neo- and adjuvant)
  - e) Reconstruction
- The outcomes of F-ESTS patients were described in terms of:
  - a) Survival
  - b) Local Recurrence
  - c) Metastases
  - d) Complications
    - i) Surgical Site Infection
    - ii) Wound Dehiscence
    - iii) Neurovascular complications

## Scope and limitations of the study

This study described the important clinical characteristics, treatment patterns, and outcomes of twenty patients diagnosed with F-ESTS and treated by a single surgeon at a sarcoma unit. Because of the limited number of cases presented, this study could not make statistical associations or analyze the relationships of clinical and treatment factors with outcomes. Another limitation of this study is that the cases were only from a single orthopedic tumor surgeon and other F-ESTS patients treated by other tumor surgeons at the sarcoma unit were excluded.

## **METHODOLOGY**

#### Study design

This was a retrospective clinical study on F-ESTS patients treated by the senior investigator at a sarcoma unit over 25 years (1993–2018).

## **Patient selection**

#### Inclusion criteria

- Fungating extremity soft tissue sarcoma (F-ESTS) patients treated by the senior investigator
- Histologically confirmed STS by sarcoma unit pathologist
- F-ESTS patients with and without metastasis on presentation
- F-ESTS patients who received complete treatment at the sarcoma unit
- F-ESTS patients with at least a two-year follow-up in the absence of demise

#### Exclusion criteria

- Patients who did not undergo surgical management due to other co-morbidities
- Soft tissue sarcomas of the superficial trunk, neck

#### Materials and methods

The study was conducted at the Philippine General Hospital under the Department of Orthopaedics Tumor Service utilizing the data from the senior investigator's record of previously treated F-ESTS patients. The database was handled by the senior investigator and was accessed with his consent. From this database, the clinical characteristics, treatment variables, and outcomes were identified. The clinical characteristics included age, gender, tumor size, tumor depth (subcutaneous or subfascial), tumor site, tumor grade (low/ high), tumor histology, biopsy before surgery, initial treatment done before sarcoma unit, and metastases at presentation. The treatment variables included were the type of surgery done (limb salvage/amputation), surgical margins (wide/radical), margin status (R0/R1), adjuvant therapy (radiotherapy/ chemotherapy) given, and reconstruction done. The outcomes identified were the overall survival, local recurrence, metastases, complications (SSI, wound dehiscence, neurovascular injury), and the latest status of the patients. The follow-up data for two years or until demise were included to review the outcome.

A data collection form (Microsoft Excel) contained the clinical characteristics, treatment, and outcomes. The patient's identity was not reflected in the collected data or the study. The data were not photocopied or duplicated and were stored and protected in a password-protected computer that only the researchers could access during the study. After the study, access was limited to the senior investigator. Sharing of the collected data was only allowed per the senior investigator's approval.

Data privacy for this study was maintained and no other personnel accessed the collected data. There were no reports to the Philippine General Hospital Data Privacy Officer.

#### Sample size

The sample size was not computed due to the rarity of the F-ESTS. All patients with F-ESTS treated by the senior investigator over the past 25 years were included. Of the 258 STS patients' records reviewed, this yielded 20 patients with F-ESTS whose data collected included the incidence, clinical factors, treatment, and outcome.

## Statistical analysis

Descriptive statistics were used in this study to describe and summarize the data. The means for age, tumor size, and survivability in months were computed. The frequency of gender, tumor depth, tumor site, tumor grade, tumor histology, biopsy before surgery, initial surgery, surgical treatment, surgical margins, margin status, adjuvant therapy,

reconstruction, overall survival, local recurrence, metastases, and complications were interpreted in percentage.

## **Ethical considerations**

This study protocol was reviewed and approved by the University of the Philippines Manila Research Ethics Board (UPMREB) Panel. following the Data Privacy Law of 2012, Republic Act 10173, all patient information was kept anonymous and confidential. There was no external funding for this study; the primary investigator provided funding.

A waiver of informed consent was approved by the UPMREB panel in line with the National Ethical Guidelines for Health and Health-related Research of 2017 section 11.2; the study entailed no more than minimal risk and the medical records of the patients included in the study and their anonymity were maintained.

#### **RESULTS**

We reviewed 258 patients diagnosed with soft tissue sarcoma and treated by the senior investigator from 1993 to 2018. A total of 28 patients were identified to have F-ESTS. Three patients did not meet the two-year minimum follow-up and five patients were excluded due to missing data. Hence, 20 patients with F-ESTS were included for analysis in this study. Table 1 summarizes the clinical factors, treatment variables, and the outcomes of these patients.

F-ESTS patients had a mean age of 49.2 years old ranging from 17–80 years old. Most (55%) were female. The most common site for tumor occurrence was on the lower extremity, specifically the thigh, accounting for ten (50%) of the cases. On presentation, tumor size ranged from 5.5 to 20 cm with a mean of 11.8 cm. The majority of the tumors were deep, of which 85% were high grade (11 of the 13 deep tumors). Seven patients had superficial tumors, six of which were high-grade. Histologically, the most common diagnosis was a high-grade liposarcoma in six (35%) of the patients. This was followed by malignant peripheral nerve sheath tumors (MPNST) and rhabdomyosarcomas. Our study also had cases of synovial sarcoma, extra-skeletal osteosarcoma, high-grade undifferentiated pleomorphic sarcoma, low-grade spindle cell tumors, and low-grade angiosarcomas (Table 2).

Eleven patients (55%) had an unplanned excision and only two patients underwent diagnostic biopsies before consulting at the sarcoma unit. Two patients were diagnosed with metastasis on presentation (Table 2).

Limb salvage with wide margins was the treatment of choice in twelve (60%) patients while eight (40%) patients underwent amputation. An R0 margin was achieved in sixteen (80%) patients (Table 3). Out of the four (40%) cases with an R1 margin, three followed a limb salvage procedure and one had an amputation. Five patients (25%) received radiotherapy while four (20%) received chemotherapy. Two of the five

patients who had radiotherapy (RT) received it before surgery. Neoadjuvant chemotherapy was given in only one patient for a high-grade MPNST, in continuity with the chemotherapy given after an unplanned excision with a previous surgeon. A reconstructive procedure was required in 50% of the patients (skin graft in seven patients and flap coverage in three patients) (Table 3).

Post-operatively, three (15%) patients developed complications. Two of these patients had both a surgical site infection and wound dehiscence and one had wound dehiscence alone. Five patients (25%) developed local recurrence. Thirteen patients (65%) had distant metastasis on their latest follow-up. Lymph node metastasis had occurred in three patients with the following histologies: embryonal rhabdomyosarcoma, extra-skeletal osteosarcoma, and dedifferentiated liposarcoma (Table 4).

Patients in this study had a mean survival of 49.2 months ranging from two months to 13.5 years. Twelve patients (60%) died of disease while one died of other causes. Only four (20%) patients were alive without evidence of disease on the latest follow-up (Table 4).

## **DISCUSSION**

The presence of fungating STS has anecdotally denoted a poor prognosis; a 5-year survival rate of only 20.4% is expected for those patients presenting with a fungating STS. There is still a limited understanding of this aggressive tumor presentation. Potter et al., from the University of Miami Miller School of Medicine retrospectively reviewed 170 STS patients over fifteen years with twenty-four (14.1%) cases of fungating STS. Parry et al., from the Royal Orthopaedic Hospital, UK, retrospectively reviewed 2661 STS patients over eighteen years and found eighty-six (3.2%) cases of fungating STS. Okajima et al., from the Tokyo Metropolitan Cancer and Infectious Diseases Center Komagome Hospital/University of Tokyo Hospital/Saitama Medical Center Jichi Medical University retrospectively reviewed 26 patients with "Malignant fungating wounds (MFW)" over fourteen years and found that 19 (73%) patients initially presented with F-ESTS, and 7 (27%) developed fungation during the treatment course (Table 5).7

Of the 258 STS cases from the senior author's 25-year database, only 10.9% presented with a fungating mass. Although infrequent, this crude incidence was much higher than that described by Parry et al. in their retrospective series (3.2%). This may reflect delayed health-seeking behaviors in patients and/or a treatment gap in the management of these cases.

In our study, F-ESTS occurred in a younger age group compared to that in published literature (Table 5). Like the studies of Potter and Parry et al, our F-ESTS cases occurred mostly in women. The most commonly affected area was the thigh. Like the other studies, the tumor size was approximately 10 cm or larger. The most frequently encountered histology differed across the literature; locally, high-grade liposarcomas

were most common. Fewer of our cases had metastasis on presentation. Potter et al. and Parry et al., aside from including only high-grade F-ESTS, also included patients who did not undergo surgery due to advanced disease. This could account for higher rates of metastasis on presentation in their study populations.

Before presenting to the sarcoma unit, 55% of patients had an unplanned excision of their tumor by another surgeon. The residual tumor, postoperative inflammation, and iatrogenic break in the skin most likely contributed to conditions favorable for fungation. Two patients who underwent a previous biopsy procedure were spared from an unplanned excision but still presented with a fungating lesion, most likely due to the same conditions mentioned.

All studies included M1 patients at presentation. We describe two M1 patients on presentation: one who underwent amputation and eventually died of disease and one who underwent limb salvage but was still alive with evidence of disease on a four-year follow-up.

Most patients in our study (60%) were treated with limb salvage surgery using wide margins. However, three of those cases reported R1 margins following surgery which may be due to inadequate resection or tumor contamination. The presence of a large, fungating, and sometimes leaking mass during limb-salvage surgery can present a difficult and unique challenge to the tumor surgeon when trying to avoid tumor contamination. Techniques include using iodine-impregnated adhesive drapes over gauze dressings to contain the fungated component. A review of these R1 cases shows that three patients were given post-operative RT without re-excision; only one underwent pre-operative RT. Two of these initially non-metastatic patients developed both local recurrences and distant metastasis within months of their surgery at the unit. Both patients died of disease before their one-year follow-up.

Published data of non-metastatic ESTS cases in the same sarcoma unit would show an aggregate amputation rate of 12.8%. The amputation rate of our F-ESTS study (40%) was much higher, considering the overwhelmingly high-grade nature of this group. Tumor size did not seem to be a factor in amputation since the size of F-ESTS was no different than the average size of STS presenting at the sarcoma unit (~11 cm). Although our decision to amputate followed similar protocols to published studies, the amputation rate for our study of fungating cases was almost twice that of Parry et al. (23%).

The presence of local recurrence, distant metastases, and poor survival make surgeons question if limb salvage surgery (LSS) can be pursued or if all F-ESTS cases should be amputated. The outcome of LSS in our study demonstrated three (25%) cases of local recurrence, seven (58%) cases of distant metastasis, and seven (58%) cases died of disease. The amputated group demonstrated two (25%) cases of local recurrence, six (75%) cases of distant metastasis, and five (63%) cases died of disease. The oncologic outcomes of the two surgical groups (LSS vs

**Table 1.** Clinicopathologic demographics of F-ESTS patients

Case	Age at Diagnosis (Years)	Sex	Tumor site	Tumor size	Tumor depth		Tumor histology	Biopsy prior to surgery	Surgery prior to unit	Neoadjuvant Therapy	Metastases at presentation	Surgical treatment	Surgical margins	Margin status	Adjuvant RT	Adjuvant Chemo	Recon- struction	Compli- cation	Recur- rence	Metastases	Survival (months)	Latest status
1	46	М	Knee	14	SF	high	Rhabdomyosarcoma pleomorphic	No	no surgery	none	No	limb salvage	wide	R1	Yes	No	Yes	None	Yes	Yes	8	DOD8
2	48	F	Elbow	11	SF	high	Liposarcoma myxoid	No	unplanned excision	none	No	amputation	wide	R0	No	No	No	None	No	No	150	ANED150
3	33	М	Knee	11	SF	high	Liposarcoma NOS	No	unplanned excision	none	No	amputation	radical	R0	No	No	No	SSI, Wound dehiscence	Yes	Yes	3	DOD3
4	37	М	Leg	14	SF	low	MPNST	No	unplanned excision	none	No	amputation	radical	R1	No	No	No	None	No	Yes	162	AWED162
5	73	М	Thigh	16	SF	high	Liposarcoma NOS	No	unplanned excision	RT	No	limb salvage	wide	R0	Yes	No	No	None	No	Yes	12	DOD12
6	56	F	Leg	8.2	SF	low	Angiosarcoma of soft tissue	No	unplanned excision	none	No	amputation	wide	R0	No	Yes	No	None	Yes	Yes	52	DOD52
7	17	М	Foot	20	SF	high	Rhabdomyosarcoma embryonal	No	no surgery	none	Yes	amputation	radical	R0	No	No	No	None	No	Yes	2	DOD2
8	58	F	Thigh	20	SF	high	Liposarcoma pleomorphic	No	no surgery	none	No	amputation	radical	R0	No	No	No	None	No	Yes	53	DOD53
9	64	F	Foot	6	SF	high	Synovial sarcoma NOS	No	unplanned excision	none	No	amputation	radical	R0	No	No	No	None	No	No	105	ANED105
10	33	F	Thigh	n/a	SF	high	MPNST	No	unplanned excision	Chemo	No	limb salvage	wide	R0	Yes	Yes	No	None	Yes	No	16	DOD16
11	62	F	Thigh	11.5	SC	high	Liposarcoma myxoid	Yes	no surgery	none	No	limb salvage	wide	R0	No	No	Yes	None	No	Yes	30	DOD30
12	66	F	Thigh	7.5	SC	high	MPNST	Yes	no surgery	none	No	limb salvage	wide	R0	No	No	Yes	None	No	No	65	DOC65
13	32	F	Thigh	5.5	SC	high	Rhabdomyosarcoma alveolar	No	unplanned excision	none	No	limb salvage	wide	R0	No	No	Yes	None	No	Yes	14	DOD14
14	53	М	Gluteal	n/a	SC	high	Extraskeletal osteosarcoma	No	unplanned excision	none	Yes	limb salvage	wide	R0	No	Yes	Yes	None	No	Yes	47	AWED47
15	64	F	Thigh	18	SF	high	UPS	No	unplanned excision	none	No	limb salvage	wide	R1	Yes	No	Yes	None	Yes	Yes	7	DOD7
16	60	F	Thigh	11	SC	high	Liposarcoma dedifferentiated	unknown	planned wide	none	No	limb salvage	wide	R0	No	No	Yes	Wound dehiscence	No	Yes	48	DOD48
17	37	М	Thigh	12	SC	high	High grade sarcoma	No	no surgery	none	No	limb salvage	wide	R0	No	Yes	Yes	None	No	No	108	AWED108
18	25	М	Thigh	n/a	SF	high	Synovial sarcoma NOS	No	no surgery	none	No	amputation	wide	R0	No	No	No	SSI, Wound dehiscence	No	Yes	6	DOD6
19	40	М	Pelvic	7	SC	low	low grade spindle cell tumor	No	no surgery	none	No	limb salvage	wide	R0	No	No	Yes	None	No	No	53	ANED53
20	80	F	Forearm	7.5	SF	high	MPNST	No	unplanned excision	RT	No	limb salvage	wide	R1	Yes	No	Yes	None	No	No	44	ANED44

M = male, F = female, SF = subfascial, SC = Subcutaneous, MPNST = Malignant peripheral nerve sheath tumor, UPS = Undifferentiated Pleomorphic Sarcoma, NOS = Not otherwise specified, RT = Radiotherapy, Chemo = Chemotherapy, R0 = clear margins, R1 = tumor detected microscopically, SSI = Surgical Site Infection, DOD = Dead on disease, DOC = Dead on other cause, AWED = Alive with evidence of disease, ANED = Alive with no evidence of disease

Table 2. Clinical characteristics of patients with F-ESTS

Variables	N	Frequency (%)	Mean
Age	Mean = 49.2 (Range 17-80)		
Gender			
Male Female	9 11	45 55	
Tumor site			
Elbow Forearm Pelvis Buttock Thigh Knee Leg Foot  Tumor size (cm)  05-Oct Nov-15	1 1 1 1 10 2 2 2 2 2	5 5 5 5 50 10 10 10 10	11.8
16-20 Tumor depth	4	23.5	
Subcutaneous Subfascial	7 13	35 65	
Tumor grade			
Low High	3 17	15 85	

Variables	N	Frequency (%)	Mean
Tumor histology			
Liposarcoma	7	35	
Rhabdomyosarcoma	3	15	
MPNST	4	20	
Angiosarcoma	1	5	
Synovial Sarcoma	2	10	
Extraskeletal OSA	1	5	
UPS	1	5	
LGSCT	1	5	
Biopsy prior to surgery			
Yes	2	11	
No	17	89	
Surgery prior to sarcoma ur	nit		
Planned	1	5	
Unplanned	11	55	
None	8	40	
Metastases at presentation			
Yes	2	10	
No	18	90	

Patients Diagnosed with Fungating Soft Tissue Sarcomas

MPNST = Malignant Peripheral Nerve Sheath Tumor, LGSCT = Low Grade Spindle Cell Tumor, UPS = Undifferentiated Pleomorphic Sarcoma

**Table 3.** Treatment variables for F-ESTS patients

Variables	N	%
Surgery done	•	
Limb salvage	12	60
Amputation	8	40
Surgical margin		
Wide	15	75
Radical	5	25
Margin status		
R0	16	80
R1	4	20
Radiotherapy		
Neo-adjuvant	3	15
Adjuvant	5	25
Neo or adjuvant	5	25
None	15	75
Chemotherapy		
Neo-adjuvant	1	5
Adjuvant	4	20
Neo or adjuvant	4	20
None	16	80
Reconstruction		
Yes	10	50
No	10	50

R0 = clear margins, R1 = tumor detected microscopically

**Table 4.** Outcome of treated F-ESTS patients

Variables	N	%
Complications		
Yes	3	15
SSI	2	10
Wound dehiscence	3	15
Neurovascular	0	0
No	16	80
Local recurrence		
Yes	5	25
No	15	75
Distant metastases		
Yes	13	65
No	7	35
Survival (months)	Mean = 49.2 months	
	(Range: 2 to 162 months)	
Status		
ANED	4	20
AWED	3	15
DOD	12	60
DOC	1	5

SSI = Surgical Site Infection, DOD = Dead on disease, DOC = Dead on other cause, AWED = Alive with evidence of disease, ANED = Alive with no evidence of disease

Table 5. Comparison of characteristics and outcomes of F-ESTS patients in the literature

Mean	Mean age (years)	Mean tumor size (cm)	Most common location	Most common histology	Deep tumors (%)	High grade tumors (%)	M1 on presentation (%)	Crude LR (%)	Crude DM (%)	Died of disease (%)
Our study	49.2	11.8	Thigh	High grade liposarcoma	65	85	10	25	65	60
Potter et al	64.9	9.9	Thigh/ groin	UPS / MFH	46	100	33	13	44	54
Parry et al	68.8	11.4	n/a	Angiosarcoma	48	86.8	20	20	n/a	75
Okajima et al.	73	≥10 cm	Extremity	UPS / MFH	54	100	31	11	44	61

M1 = Distant metastases, LR = Local recurrence, DM = Distant metastases

amputation) were similar in terms of local recurrence and distant metastasis. The oncologic principle of doing limb salvage surgery for STS whenever marginally possible still applies regardless of whether a fungating lesion is present. We note that most surgeries done before 2010 were amputations (78% were amputations). Treatment previously leaned more towards amputation due to fear of tumor leakage and contamination. In the last decade, there was a reversal toward limb salvage surgeries even for deep highgrade F-ESTS cases (92% were LSS). Okajima et al., similarly call for the consideration of limb-salvage surgery to improve quality of life.

Following treatment protocol, all patients with deep and highgrade tumors, for which limb salvage surgery was planned, were given RT pre- or post-operatively. The use of chemotherapy, on the other hand, was much less routine in our study. Parry et al. do not analyze the effect of chemotherapy except to mention that this was rarely given (i.e., for soft tissue Ewing's, rhabdomyosarcomas, or advanced disease). In contrast, Potter et al. mostly followed a chemotherapy treatment protocol for patients with a large, high-grade sarcoma, effectively giving 61% of their patients a neoadjuvant doxorubicinbased drug regimen. They elaborated that in patients with >90% tumor necrosis after the neoadjuvant treatment, adjuvant chemotherapy was continued post-operatively while those with <90% tumor necrosis had modified adjuvant chemotherapy or were alternatively given radiotherapy. Although their regression analysis showed no benefit of chemotherapy for disease-specific survival, Potter et al. had better survival rates and local control compared to Parry et al. despite a population with inherently poorer prognosis. Potter et al., also emphasize that an aggressive multidisciplinary approach can improve survivorship for this group of patients. This does give us pause to consider that their treatment protocols may account for the difference.

Almost half of the patients in this study underwent surgical reconstruction using skin grafting, and flap coverage. Two cases of SSI and one case of wound dehiscence were recorded. Only one patient with SSI underwent re-operation (debridement). This patient previously presented with a fungating mass that was already infected. Despite multi-modal treatment consisting of adjuvant chemo and RT, the patient eventually developed malignant degeneration of his other lesions and died of disease within two years of his STS diagnosis.

The 148 ESTS patients included in Wang et al.'s study of unplanned excisions without metastasis on presentation were fairly distributed between low- (40%) and highgrade (60%) lesions. In this study of F-ESTS patients, the majority (85%) were high-grade lesions. Comparing the outcomes of this study with Wang et al., there was a slightly higher local recurrence (25% vs 23%), almost double the rate of distant metastasis (65% vs 36%), and nearly half the survival rate (35% vs 63%). The patients in this study had half the survival rate of the aggregate STS population treated at the sarcoma unit (64%). These crude differences may be attributed not only to fungation but also to the concentration of high-grade tumors in our study and the inclusion of M1 cases on presentation.

Our study had proportionally more patients who developed local recurrence and distant metastasis but had fewer patients lost to disease compared to Parry et al., likely explained by the inclusion of several inoperable F-ESTS patients in the latter study.

## **CONCLUSION**

Common clinical characteristics in our F-ESTS study were age <65 years, deep and high-grade tumors, typically liposarcomas, a predilection for the thigh, and a previous history of surgery or biopsy. Our study involved a younger age group of F-ESTS cases, with a larger proportion of deep tumors compared to the literature.

At the end of our study, five (25%) patients developed local recurrence, 13 (65%) patients had distant metastases, and 12 (60%) died of disease with a mean time to death of 21 months from diagnosis. Those who survived had a mean follow-up time of 7.6 years (range: 44 months to 13.5 years). This poor outcome was consistent with prognostic studies in the literature. However, with multi-modal treatment at a sarcoma unit, a long disease-free survival remains entirely possible. While chemotherapy use was rare and inconsistent given a lack of published evidence, there is room to improve our local control rates with a lower tolerance for radiotherapy (i.e., giving RT for all high-grade lesions regardless of depth).

Most F-ESTS patients were still able to undergo limb salvage surgery, receiving RT if they had both deep and high-grade tumors. We found that in the last 10 years, limb salvage surgery had become the treatment of choice for the patients in this

study, without grossly compromising local control or survival outcomes. This upends the notion that fungating tumors require amputation and may allay fears of contamination from "tumor leakage."

Given the inherent limitations of our study design, more collaborative and comprehensive prospective studies can be done involving other institutions and orthopedic oncologic surgeons (i.e. multi-center research) to gather more robust data on F-ESTS. The same issue can be addressed by tapping into existing registry data from cancer societies or consortiums.

## STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

## **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

## **FUNDING SOURCE**

None.

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## Large Exostosis of the Distal Radius in a Pediatric Patient Causing a Dysplastic Ulna and Distal Radioulnar Joint Disruption: A Single-stage Management Approach

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#### **ABSTRACT**

The distal radioulnar joint (DRUJ) is an important structure that stabilizes the radius and ulna. Any incongruency may result in limited forearm rotation and a weak grip. Exostosis is a benign bony cartilage-capped outgrowth protruding from the surfaces of affected bones. These occur as solitary lesions in 1-2% of the general population. Primary resection at the base is the mainstay of treatment for solitary exostosis lesions. In rare cases, an exostosis of the distal radius can grow particularly large leading to deformities of the adjacent ulna and DRUJ. In this case report, we present a patient with a large distal radius exostosis resulting in a dysplastic ulna with DRUJ involvement. Case reports on this rare phenomenon report good results following osteotomy and gradual lengthening of the ulna with an external fixator – a multi-stage surgical approach. In this case report, treatment involved resection, pinning, bone grafting with mesh, and the use of a fascial sling taken from the radial volar fascia wrapped around the distal ulna, resulting in near full return of function for the forearm and hand – a single-stage management approach.

**Keywords.** pediatric, exostosis, distal radius, dysplastic ulna, incongruent DRUJ, surgical treatment, single stage, one stage, DRUJ reconstruction

## **INTRODUCTION**

Primary bone tumors arise from bone tissue and range in severity from benign to significantly malignant. An exostosis is a benign growth of bone that typically presents with a characteristic cartilaginous cap toward which the growth is directed. When a cartilaginous cap is present in an exostosis, they are called osteochondromas. These lesions may be solitary and spontaneous or linked to genetics with multiple affected bones. They are more commonly found in long bones such as the radius, humerus, femur, tibia, the pelvis, and shoulders. Prevalence studies show about a 1% rate of occurrence of solitary lesions in the general population.<sup>1</sup>

These can cause dysplasia of adjacent bones and lengthening of adjacent tissues. Deformities of the forearm, when present, are relatively common at a 30–60% incidence rate. These disruptions, particularly around the DRUJ may cause significant impairment in patients' hand function, particularly with forearm pronation, supination, and grip strength. The DRUJ is one of the primary structures that transmit load at the distal forearm and wrist. It acts as a pivot for radioulnar rotation<sup>2</sup> and its disruption may significantly impair function. A large exostosis of the distal radius may lead to a dysplastic ulna in the form of ulnar shortening and excessive localized bowing to accommodate the lesion.

eISSN 2012-3264 (Online)
Printed in the Philippines.
Copyright© 2024 by Quintos et al.
Received: January 7, 2024.
Accepted: February 11, 2024.
Published Online: March 15, 2024.
https://doi.org/10.69472/poai.2024.05

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**Figure 1.** Pre-operative gross pictures of lateral **(A)** PA **(B)**, with pronation **(C)** and supination **(D)**. X-rays of forearm AP **(E)**, lateral **(F)** and wrist AP **(G)** and lateral **(H)** on presentation to the clinic showing gross limitations in pronation and supination and a large exostosis at the distal radius.

Treatment options for patients who have a large exostosis with DRUJ disruption and dysplastic ulna range from simple removal of exostosis to distraction procedures with external fixation of the ulna.<sup>3</sup> This case report presents a pediatric patient who came in with a large exostosis of the right distal radius causing a dysplastic ulna with disruption of the DRUJ, treated with resection of mass, osteotomy, and pinning of the ulna with bone grafting and mesh reinforcement all in a single-stage approach.

#### **CASE**

A 9-year-old right-handed girl with no known comorbidities presented to our service for a mass on her wrist and a limited range of motion of her right forearm. The mass started small four years before consultation without any inciting event or trauma. The mass grew over the years with further limitations in pronation and supination of the right forearm. One year

prior the patient began to have difficulty lifting heavier objects with her affected hand. On consult, limited pronation (30 degrees) and supination (25 degrees) were documented. The circumference of the right distal forearm was measured at 15 cm as compared to the left at 12.5 cm. Motor strength for grip was 4/5 for the right in comparison to the left (Figure 1A).

X-rays revealed an exostosis of the distal radius abutting against the ulna resulting in a dysplastic ulna. The widened radio-ulnar interval and negative ulnar height suggested DRUJ laxity. The figures below are the pre-op x-rays taken one month before surgery (Figure 1B).

The goals of treatment were to remove the exostosis, prevent recurrence, stabilize the DRUJ, and restore the normal function of the forearm and wrist. The surgeon performed a resection of the exostosis, pining of the distal ulna, and autologous bone grafting from the radius with mesh application.



**Figure 2.** Gross photo of exostosis intraoperatively. \*Area abutting the ulna.

A dorsal approach between the 4<sup>th</sup> and 5<sup>th</sup> dorsal compartments was used to expose the exostosis and dysplastic ulna (Figure 2). The exostosis was removed using an osteotome after stripping the surrounding periosteum ensuring that the entire cartilaginous cap was resected along with the stalk. As shown in Figure 3A, after resection of the exostosis, a dysplastic ulna was noted resulting from the abnormal growth of the resected mass. Corrective osteotomy was performed to align and restore the height of the ulna. The distal fragment was fixed to the intramedullary canal of the proximal ulna using a 0.062" K-wire, leaving a gap (Figure 3B). A bone graft obtained from the periphery of the exostosis resection site was

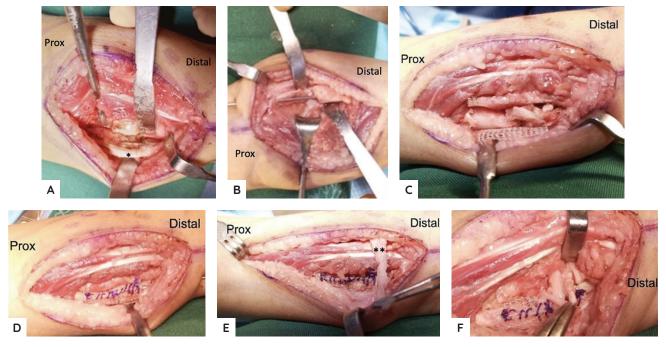
placed over the gap at the ulnar osteotomy site and secured with a resorbable mesh (Figure 3C-D). Stripped periosteum and surrounding fascia from the area of resection (Figure 3E) was then passed underneath the extensor muscles toward the ulna, looped around the ulna osteotomy site, and secured onto itself with sutures acting as a soft tissue stabilizer recreating some function of the DRUJ (Figure 3F). An ulnar gutter splint was applied post-operatively for protection.

Post-op x-rays showed complete resection of the mass with the bone graft located at the ulnar gap (Figure 4). The patient followed up at two weeks for wound inspection then at two months with repeat x-rays showing consolidation of bone graft and incorporation into the distal ulna with callus formation (Figure 5).

Histological studies revealed an osteochondroma with no malignant potential or transformation (Figure 6). The intramedullary pin was removed at 10 weeks post-operatively (Figure 7).

There was a gradual improvement in the pronation and supination of the right forearm (Figure 8, Table 1). Serial piano key tests also showed near-symmetric stability of the DRUJ throughout the range of the forearm. On serial X-rays after removal of the pin, ulnar remodeling occurred (Figure 9). The callus bridging of the gap was achieved and the patient regained 50 degrees of pronation.

At four months post-pin removal, the patient had good supination reaching up to 80 degrees and pronation of 70



**Figure 3.** Intraoperative images taken. After resection of mass noting the \*dysplastic flattened ulna **(A)**; pinning of the ulna after corrective osteotomy was performed to secure fragments **(B)**; application of bone graft and mesh done to fill the defect between proximal and ulnar fragments **(C)**; closure of mesh with absorbable sutures – securing the graft in place **(D)**; \*\* sling taken from loose periosteum and surrounding fascia from radius looped around ulna and sutured onto itself acting as a soft tissue stabilizer **(E)**; final inspection prior to closure **(F)**.



**Figure 4.** Post-op X-rays. Showing resected distal radius with an intramedullary pin of ulna. Bone graft shown at defect of the ulna in wrist AP **(A)**, lateral **(B)**, and forearm AP **(C)**, and lateral **(D)** x-ray views.



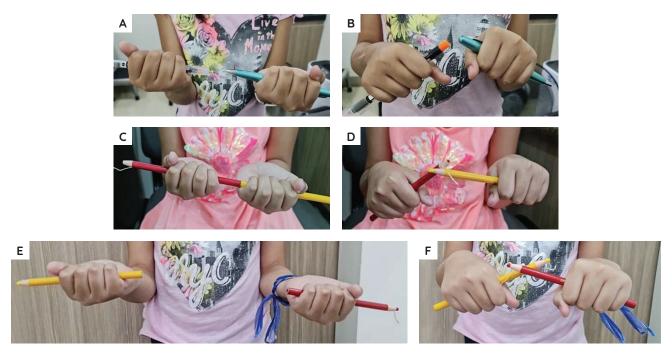
**Figure 5.** X-rays of wrist AP **(A)** and lateral **(B)** at 2 months follow-up showing intact pin and beginning consolidation of ulna with callus formation.



**Figure 6.** Histological studies revealed an osteochondroma with no malignant potential or transformation. Intramedullary pin was removed with note of some residual gapping and beginning bridging callus over area of defect (10 weeks post-op op).



**Figure 7.** Forearm AP **(B)** and lateral **(A)** X-ray views. Intramedullary pin was removed with note of some residual gapping and beginning bridging callus over area of defect (10 weeks post-op).



**Figure 8.** Gross pictures show improving pronation and supination of both forearms. Follow up pronation and supination at 2 months **(A,B)**, 4 months **(C,D)**, and at 7 months **(E,F)**.

**Table 1.** Range of motion tests after removal of pin measured in degrees from neutral position

Date	Range of Motion (deg)					
Date	Pronation	Supination				
Pre-op	0-30	0-25				
1 month	0-50	0-80				
2 months	0-50	0-80				
4 months	0-70	0-80				
7 months	0-70	0-90				



**Figure 9.** Follow-up x-rays at 1 month **(A,B)**, 2 months **(C,D)** and 4 months **(E,F)** after removal of pin showing bridging callus and progressive remodelling.

degrees. Grip strength also significantly improved as described by the patient and as clinically assessed with the finger squeeze test. A more objective tool such as a dynamometer would have been ideal to document this improvement. Follow-up X-rays showed union of the proximal and distal ends of the ulna, the graft had been incorporated into the defect. No local recurrence of exostosis was noted.

At seven months post-op, full supination was achieved at 90 degrees along with 70 degrees of pronation. X-rays showed further remodeling of the distal ulna now more vertical in orientation with less dorsal attitude.

Of significant note is the presence of some shortening of the ulna on long-term follow-up. Some consequences of this finding may be future instability and ulnar impingement syndrome. These would present with pain at the DRUJ, particularly on supination and pronation. Patients should be followed up regularly to monitor for this pathology. The patient reports being greatly thankful that the surgery was performed. She reported that she can better use her right hand for daily activities and is no longer bothered by gross deformity.

## **DISCUSSION**

The distal radioulnar joint is an important structure that stabilizes the radius and ulna close to the wrist. It is responsible for the transmission of load between the bones of the forearm and the wrist. It is a diarthrodial, synovial articulation that serves as a pivot for pronation and supination between the radius and ulna. any incongruency may result in limitations of forearm rotation and weakness in grip. The DRUJ is also synchronous with the proximal radioulnar joint and is integrated with the ulnocarpal joint. Any of these adjacent joints may be dysfunctional if the DRUJ is not intact.

The sigmoid notch of the radius contributes significantly to the stability of the DRUJ. This is the area where the radius rotates about the ulna during pronation and supination. Ulnar variance describes the relation of the height of the ulna to the articulating surface of the radius. In adults the ulnar variance averages at -0.9 mm. The tip of the ulna houses the ligamentous complex that provides stability to the ulnar side of the wrist, the triangular fibrocartilaginous complex (TFCC). Two ligaments in the TFCC provide the most stability to the DRUJ and these are the dorsal and palmar radioulnar ligaments.

Not much literature is described for reconstruction of the DRUJ in patients with large exostosis of the distal radius primarily due to the low incidence of these types of lesions with the aforementioned sequelae in the general population. Several techniques have been described to address and treat these types of deformities. Treatment options range from simple resection of mass to combined procedures such as osteotomy distraction with progressive lengthening and adjustments to address the shortening of the dysplastic ulna.

The goal of treatment is to restore the stability and strength of the DRUJ – an important structure in transmitting load between the forearm and hand and in the articulation of the radius and ulna via pronation and supination. Any disruption or incongruence can lead to limitations in the range of motion of the forearm and wrist. Further enlargement of masses at the wrist may cause weakening of grip or compression of nerves along with contractures of flexor and/or extensor tendons. <sup>2,3</sup> Our patient presented with limitations primarily in forearm supination and pronation, only achieving 30 degrees of pronation and 25 degrees of supination pre-operatively. No objective assessment for grip strength was done but the patient had a good grip and was able to hold several objects with the affected hand.

The Masada classification system was developed to communicate the location of the mass and adjacent structures. It is typically used for patients with multiple osteochondromatosis but may have applications for use in solitary lesions as in our patient. Based on the diagram, our patient may be exhibiting a type III lesion.

Multistage lengthening is described in several papers to address the ulnar dysplasia, however, for some patients, funds issues may limit their options. Aerts et. al.4 Described a similar case wherein an external fixator was fixed onto the ulna length and progressively adjusted to achieve the desired. In literature, most patients who undergo this type of distractive procedure are pediatric patients since their potential for bone healing and remodeling is greater. In these cases, patient outcomes were good, and return to function was achieved at six months. The decision to proceed with the technique utilized in this paper was to address both the exostosis and the DRUJ reconstruction with lengthening of the ulna in one sitting instead of a multistage approach. The idea was to create a gap and temporarily fix the distal ulna with a k-wire pin in distraction, filling the gap with autologous bone graft from the radius. In this way, no further implantation and no further manipulation of the ulna was needed on follow-ups. The bone graft would simply incorporate into the defect resulting in lengthening of the previously dysplastic ulna.

In terms of the DRUJ, the standard technique for stabilizing the joint is a trans-radio-ulnar pin in a position of greatest stability (more commonly neutral or supination). Other techniques have also been described such as utilization of a tendon graft sling looped around the radio-ulnar junction.<sup>5</sup> However, in cases of dysplasia of the ulna where lengthening is indicated, the option to pin through both bones distally could have aided in further stabilizing the distal fragment and maintaining the ulnar length. Intraoperatively, a fascial sling was determined sufficient to provide tension at the DRUJ. The technique described proved a viable option to achieve DRUJ stability through a single-stage approach with a dysplastic ulna. One possible complication would be the shortening of the ulna. Possible interventions to prevent this complication could be the use of a transverse pin through the distal fragment and radius to maintain ulnar length.

## **CONCLUSION**

The described single-stage procedure may be a viable treatment option for patients with a large exostosis, dysplastic ulna, and DRUJ disruption when multi-stage procedures may not be viable. It was shown that pinning in distraction, bone grafting, and the use of a fascial sling resulted in a stable DRUJ after resection of the exostosis with the return of good forearm supination and pronation.

#### RECOMMENDATIONS

This paper recommends further documentation of pre- and post-op hand functional scores such as DASH Scoring, grip strength quantification, and long-term follow-up of DRUJ function with close monitoring of potential sequelae of ulnar variance.

#### **ETHICAL CONSIDERATION**

Patient consent forms were obtained before manuscript submission.

## STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

#### **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

## **FUNDING SOURCE**

None.

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# Spinal Arthrodesis with Acute Reduction in a High-grade Isthmic Type IIC Spondylolisthesis

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#### **ABSTRACT**

The objective of this study was to present a case of Spondylolisthesis of the Isthmic Type (IIC), with Meyerding Grade III, high-grade dysplastic morphology, presenting with no neurologic deficits, which underwent spinal arthrodesis with acute reduction of L5-S1 segment. Spondylolisthesis itself is rare, presenting in 6% of the adult population with low back pain. Among these cases, 11.3% are characterized by high-grade spondylolisthesis, often accompanied by neurological deficits.

This study presents the case of a young adult woman (32 years old) with spondylolisthesis of the Isthmic Type (IIC – Acute Pars Fracture), Meyerding Grade III (slippage of >50%), presenting with no neurologic deficits. We present her outcomes after undergoing instrumented spinal arthrodesis with a reduction in terms of radiographic measurements, pain, presence of post-operative neurologic deficits, and return to work. Post-operatively, the lumbar lordosis angle improved from 84 degrees to 48 degrees. There was less pain, greater functional independence in terms of activities of daily living, and eventual return to work.

Even for high-grade spondylolisthesis, reduction of the affected level with instrumented fusion may provide excellent outcomes in terms of spinal alignment, pain, and return to work.

Keywords. pars fracture, high-grade, acute reduction, lumbar lordosis, spinal arthrodesis

## **INTRODUCTION**

Spondylolisthesis is the slippage of one vertebral segment over another in the anterior, posterior, or lateral direction.<sup>1</sup> It is a rare cause of low back pain in adults (6%), with most high-grade slips being of the isthmic type.<sup>2</sup> High-grade spondylolisthesis makes up 11.3% of adult cases with spondylolisthesis.<sup>3</sup> A literature search through PubMed reveals few reports about cases of young adults with neurologically intact high-grade isthmic spondylolisthesis (acute pars fracture) undergoing instrumented spinal arthrodesis with reduction. Most papers presented small case series of the isthmic or degenerative type, with high-grade spondylolisthesis presenting with neurologic deficits.<sup>2,4</sup> Spondylolisthesis is classified into the following types according to Wiltse (Figure 1).4 The severity of the slip is also graded according to Meyerding (Figure 2).<sup>2</sup> This paper presents a case of a young adult woman (32 years old) with spondylolisthesis of the Isthmic Type (IIC: Acute Pars Fracture),<sup>4</sup> Meyerding Grade III (slippage of >50%),<sup>2</sup> presenting with no neurologic deficits, who underwent instrumented spinal arthrodesis with reduction.

Isthmic spondylolisthesis is more common in males and is most common at the L5-S1 level. Although there is a lower incidence in females, there is a higher risk for grade

elSSN 2012-3264 (Online) Printed in the Philippines. Copyright© 2024 by Odiamar et al. Received: October 3, 2023. Accepted: January 8, 2024. Published Online: March 10, 2024. https://doi.org/10.69472/poai.2024.06

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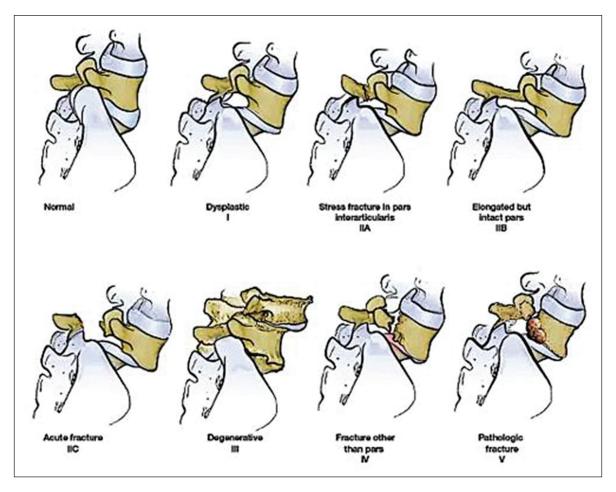


Figure 1. Meyerding classification for spondylolisthesis.

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progression. Isthmic spondylolisthesis typically presents at five to seven years old. Adult isthmic spondylolisthesis is typically asymptomatic but may present with back pain, leg pain, numbness, paresthesia, or any combination thereof. The incidence of symptoms is related to grade – grade IV presents with low back in 55–91% and radicular symptoms in 44–55% of cases. Treatment options may include pharmacologic pain control, physical rehabilitation, and bracing. Should there be a failure of conservative management, severe leg or back pain, or evidence of instability or neurologic deficit, surgical treatment is advised.<sup>5-7</sup>

Whether or not surgical treatment is recommended would also depend on the morphology of the affected segment. In the low dysplastic type, the slip involves up to 50% of the anteroposterior distance of the endplate, the endplates of L5-S1 are parallel, and there is no kyphotisation of the lumbosacral junction. In the high-grade dysplastic type, there is a slip involving greater than 50% of the anteroposterior distance of the endplate, the L5 body has a trapezoidal shape with a concave inferior endplate, and there is doming of the superior sacrum. High-grade dysplastic spondylolisthesis has a high degree of progression – and thus is an indication for surgical treatment.<sup>8</sup>

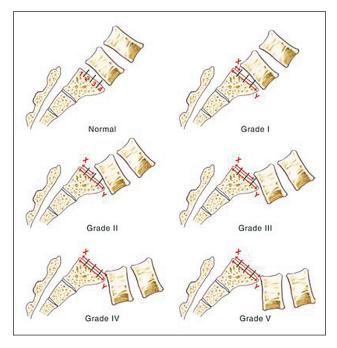


Figure 2. Wiltse classification for spondylolisthesis.

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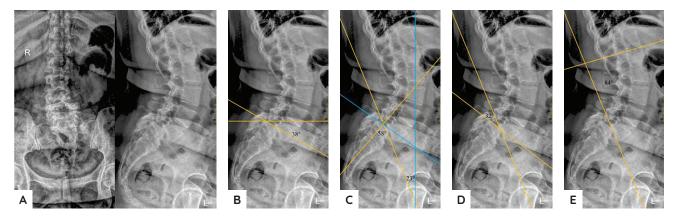


Figure 3. Showing lumbosacral AP-Lateral X-ray pre-operatively: anterior-posterior-lateral (A); sacral slope (B); pelvic incidence and pelvic tilt (C); slip angle (D); lumbar lordosis angle (E).

Controversy exists between in-situ arthrodesis, and reduction and arthrodesis. Proponents of reduction argue that with insitu arthrodesis, there is a greater risk of decompensation and pseudoarthrosis (17.8% vs. 5.5%) due to the uncorrected positive sagittal balance. In contrast, proponents of in-situ arthrodesis argue that there is a higher risk of neural complications with reduction despite the evidence still being inconclusive. <sup>1,7</sup> It is hypothesized that the dural sleeve is stretched with reduction maneuvers, putting the upper roots in tension. <sup>5</sup>

The surgical indications seen in our patient were the following: high-grade spondylolisthesis, the trapezoidal body of L5, and rounding of the superior endplate of the sacrum, as demonstrated in the pre-operative radiographs.

## **CASE**

Here, we present a case of a 32-year-old woman, a government office worker, who was injured last 2016, when she fell from standing height onto her buttocks. The radiographic examination determined that she had spondylolisthesis Grade 2. In September 2022, the patient experienced upper back pain, for which conservative management did not



Figure 4. Plain magnetic resonance imaging pre-operatively.

afford any relief. Intermittent and transient loss of muscle power and sensation over the left lower extremity was also a common occurrence. At the time of initial evaluation by an orthopedic surgeon, she did not exhibit any motor or sensory deficits, nor did she manifest with pathologic reflexes, or a positive straight leg raise test. Repeat radiographic examination revealed a Meyerding III, spondylolisthesis, L5-S1 level (Figure 3). Also seen on the radiograph were features of high-grade dysplastic spondylolisthesis, including a slip exceeding 50%, a trapezoidal-shaped L5 with a concave inferior endplate, and a doming shape of the superior endplate of S1. Additionally, magnetic resonance imaging demonstrated a spondylolisthesis of the isthmic type at the level of L5-S1, with moderate bilateral neural foraminal stenoses, worse on the left (Figure 4). Surgical management was then advised.

The operative technique proceeded as follows: the patient was placed prone on a Wilson frame with adequate padding. The hip joint was extended while the knees were flexed to avoid tension in the paralumbar muscles. An image intensifier was used to acquire anteroposterior and lateral X-ray views. From positioning alone, some reduction (one level) of the lumbosacral junction was achieved.

Dissection was carried out in the usual fashion. Intraoperatively, the surgeon noted a bilateral pars defect with a floating L5 lamina, with minimal motion at the L5-S1 segment. Pedicle screws were initially placed at L5 and S1 bilaterally. Laminectomy of L5 was performed, followed by discectomy and preparation of the disc space. The L5-S1 space was tight and could not initially accommodate the 8 mm disc shaver. A bilateral annulotomy was done to achieve release. Using a small Cobb elevator, a space was created for disc space preparation. After the disc space preparation, a bilateral facetectomy was performed. Motion was then appreciated at the L5-S1 segment. A trial of reduction was done; however, it was deemed necessary to extend the anchors superiorly to the L4 level and inferiorly to the S2 level to avoid screw pullout and to ensure a good arthrodesis. The reduction was performed using translation. The rods were placed bilaterally at S1-S2. Using reduction screws placed at L4-L5, gentle

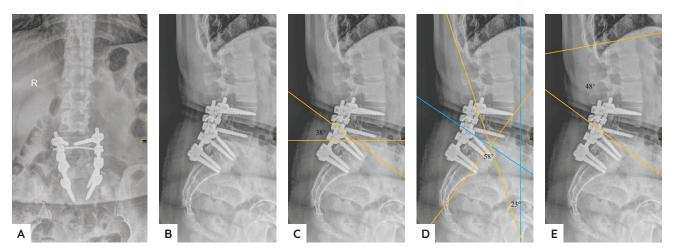


Figure 5. Showing lumbosacral AP-lateral X-ray post-operatively: AP (A); lateral (B); sacral slope (C); pelvic incidence and pelvic tilt (D); lumbar lordosis angle (E).

translation was done, watching out carefully for screw pullout. The L5 exiting roots were checked during and after the reduction. A two-level reduction was achieved. An interbody cage was not placed because it could not fit into the disc space. A local bone graft was used instead to allow interbody fusion. An onlay bone graft was also placed bilaterally (Figure 5). A satisfactory acute reduction was achieved intraoperatively – a two-grade reduction in the degree of slip, and a much-improved lordosis angle from 84 degrees to 48 degrees.

Post-operatively, the patient underwent physical rehabilitation composed of balance training, gait training, and muscle strengthening, while maintaining a chairback brace. There were no complications attributable to the index surgery. Regularly monthly radiographic examinations demonstrated the maintenance of the surgical reduction at the L5-S1 level, with no implant migration or failure (Figure 6). Although there was no loss in reduction after nine months post-surgery, the patient continued to wear a chairback brace as a precaution. The patient's compliance with rehabilitation facilitated an early return to independent function. Nine months postoperatively, the patient was already pain-free, fully independent in her activities of daily living, and had returned to work as a fully functioning member of the workforce.

## **DISCUSSION**

Reviewing the literature, our case demonstrated a two-grade improvement in the Meyerding classification as compared to Rivollier's case series, which showed a one-grade level improvement (Meyerding III).<sup>2</sup> They achieved a two-level improvement only at the Meyerding Grade IV level. No significant postoperative complications were seen.<sup>2</sup>

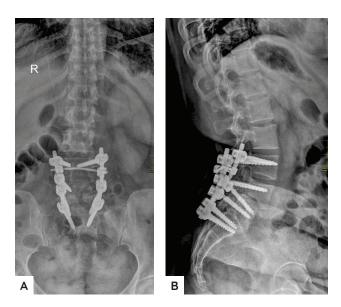
To date, regular patient re-evaluations reveal no loss of reduction and no early or late complications aligning with proponents advocating reduction before arthrodesis.<sup>1,9</sup>

In a paper by Barsotti et al., only one out of the 16 patients with high-grade spondylolisthesis who underwent an instrumented

spinal fusion with reduction had an L5 radiculopathy motor deficit at one-year follow-up, none at two-year follow-up.<sup>10</sup>

Overall, the surgery achieved what it was designated for: to lessen pain, improve functional independence, avoid neural complications, <sup>5</sup> lessen slip grade, and prevent the progression of the slip. In addition, the normal lumbar lordosis angle achieved would protect the posterior spinal ligament from excessive forces and would absorb vertically oriented loads. Excessive lumbar lordosis could cause postural and facet pain in our case due to the excessive compression forces at the apophyseal joints and anterior shear at the lumbosacral junction. <sup>11-17</sup>

Improving the spinal alignment and decreasing lumbosacral shear force would increase the chances of fusion. However, instrumented spinal arthrodesis with reduction is more technically demanding, requiring longer surgical times and increased blood loss. Our surgery lasted 9 hours and 45 minutes with an estimated blood loss of 1.2 L.<sup>10</sup>



**Figure 6.** Lumbosacral AP **(A)** and lateral X-ray **(B)** 8 months post-surgery.

Lian et al. randomized patients into reduced and in-situ groups, which showed no difference in terms of pain and functional outcomes. However, on closer scrutiny of this paper, the population consisted mostly of Grade I and II spondylolisthesis and a much smaller portion of Grade III.<sup>18</sup>

A meta-analysis by He et al. yielded 10 articles on isthmic, moderate, and serious spondylolisthesis. There was a significantly higher union rate, improvement in slippage, and shorter hospital stays in the reduction group.<sup>19</sup>

The strengths of this paper were that it presented an interesting case of a young adult with a neurologically intact high-grade spondylolisthesis who underwent acute reduction with posterior instrumented fusion and had no post-operative neurologic complications. This paper also had practical applications which could be reproducible in a similar case. The techniques employed were not novel.

The weaknesses noted in this paper are the lack of functional outcome measures done pre-operatively, immediately post-operatively, and in intervals post-operatively. We did not acquire a whole spine x-ray which would better show whole spine alignment.

## **CONCLUSION**

From the current literature, spinal arthrodesis with reduction in a high-grade spondylolisthesis is a viable option for surgical treatment. Although neurologic deficits (such as L5 radiculopathy) may occur post-operatively, the incidence is low, and the effects are transient. There is also the advantage of a higher union rate and improvement of slippage. As demonstrated in our case, there was a reduction of pain, a return to a full functional status, and no post-operative complications.

## **ETHICAL CONSIDERATION**

Patient consent form was obtained before manuscript submission.

## STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

## **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

## **FUNDING SOURCE**

None.

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## CASE REPORT / CASE SERIES









# Management of Long, Comminuted Pediatric Subtrochanteric Fractures Using PHILOS (Proximal Humeral Internal Locked System): A Case Series

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#### **ABSTRACT**

Subtrochanteric fractures in the pediatric population are rare, and there are currently no existing management guidelines. In this innately unstable fracture type, intramedullary devices preferred for adults cannot be used in children with open growth plates. A PHILOS locking plate is meant to be used in the management of proximal humerus fractures in adults, and its secondary use in children has been sparsely described in the literature.

Four pediatric patients (age range 8–14 years) with comminuted subtrochanteric femur fractures with a length averaging 7 cm (range 3.5–9.5 cm) were managed via open reduction and internal fixation using PHILOS (Proximal Humeral Internal Locked System) plates by the same surgeon and were followed up for six months, all resulting in excellent outcomes.

The PHILOS plate is a viable option in the management of subtrochanteric femur fractures in the pediatric population and long complex fracture patterns without violating the greater trochanteric physis.

Keywords. ESIN, PHILOS, pediatric subtrochanteric femur fracture

## **INTRODUCTION**

Subtrochanteric fractures in the pediatric population are rare, with an incidence of 4–10% among pediatric femoral fractures. Although there have been no guidelines in management if alignment cannot be attained by conservative means (usually with 90-90 skeletal traction), and in patients older than 10 years, surgical management is recommended using different methods such as reconstruction plates, dynamic compression plates, and adult pre-contoured locking plates. <sup>2</sup>

Gogna et al. in 2014 were the first to publish a series on the novel use of the adult proximal humerus locked plate in the management of subtrochanteric fractures in eight pediatric patients aged 10–16 years with the fracture patterns as follows: four long spiral, two short obliques, one transverse, and one comminuted.<sup>3</sup>

Chew et al. also described the use of a PHILOS plate in managing a subtrochanteric femur fracture in a 13-year-old patient with excellent results, followed over six months.<sup>4</sup>

## CASE 1

An 8-year-old boy presented with right thigh pain and deformity after being trampled on by another child during a basketball match. Radiographs show a comminuted subtrochanteric

eISSN 2012-3264 (Online)
Printed in the Philippines.
Copyright© 2024 by Morales et al.
Received: January 3, 2024.
Accepted: January 24, 2024.
Published Online: March 11, 2024.
https://doi.org/10.69472/poai.2024.08

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femur fracture with a minimally displaced proximal segment (Figure 1).

He underwent open reduction, and fixation with a 12-hole PHILOS plate under general anesthesia at 12 days postinjury. Intra-operatively, the surgeon noted that the spiral comminution was 9.5 cm long. Three Kirschner wires were initially used to maintain reduction before applying two lag screws outside the PHILOS plate. The two most proximal locking screw holes were left empty since they were directed away from the narrow femoral neck. Intra-operative blood loss was 700 cc, requiring a transfusion of one unit of packed red blood cells (PRBC). A posterior half-cylinder spica mold was placed, and the patient was allowed toe-touch



**Figure 1.** Case 1. Radiographs show a wedge spiral comminuted proximal femur, frontal **(A)** and lateral **(B)** view.

weight bearing with crutches and range of motion exercises (ROM) of the ankle.

At three months post-op, the patient could ambulate with full weight-bearing without crutches. The range of motion of the hip and knees was full and equal. On radiographs, we observed a union at the fracture site.

At six months post-op, the patient was able to ambulate with full weight-bearing without crutches, with a full range of motion (Figure 2).

## CASE 2

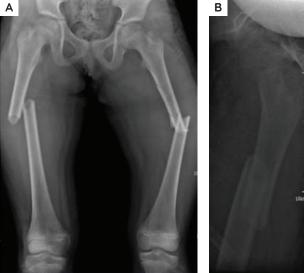
A 9-year-old girl presented with bilateral thigh deformities a few hours after being hit by a jeepney, and was subsequently diagnosed with a closed, transverse midshaft femur fracture on the right, and a closed, minimally displaced, comminuted subtrochanteric fracture on the left (Figure 3).

Initially with hemoglobin of 132 mg/dL on the day of injury, decreasing to 86 mg/dL on day two post-injury, she was hemodynamically stabilized through transfusion of two units PRBC before undergoing closed reduction and fixation using two 2.5 mm elastic stable intramedullary nailing (ESIN) rods on the right, and open reduction, fixation with a 12-hole PHILOS plate on the left under general anesthesia at seven days post-injury.

Intra-operatively, we noted that the comminution on the left subtrochanteric femur was 8 cm long. Two lag screw screws were applied within the 12-hole 26 cm PHILOS plate, which afforded compression of the fragments, converting the complex fracture into a two-part fracture. The two most proximal locking screw holes were left empty to avoid damaging the physis of the greater trochanter.



**Figure 2.** Case 1. Radiographs at six months post-op show union on both fracture sites and equal femur lengths, frontal view **(A)** and lateral view **(B)**.





**Figure 3.** Case 2. Injury radiographs show a simple midshaft femur fracture on the right and a minimally displaced, comminuted subtrochanteric fracture on the left, bilateral frontal view (A), lateral view of right (B), lateral view of left (C).

Closed ESIN fixation took 1.5 hours, while open PHILOS plating took 2.5 hours, for a total of 4 hours of surgical time. The total intra-op blood loss was 500 cc, and 1 unit of PRBC was transfused intra-op. Hemoglobin at day one post-op was 110 mg/dL. True leg lengths were equal. Post-op angulations were a one-degree varus on the right and an anatomic reduction on the left.

A posterior half-cylinder spica mold was placed on the right lower extremity, and the patient was allowed no weight bearing for a total of 12 weeks, allowing only range of motion exercises of both hips and knees starting two weeks post-op.

At three months post-op, both limbs were equal in length, and the patient was able to stand on both lower extremities with full weight bearing.

At six months post-op, the patient was able to ambulate on full weight bearing without crutches, with full range of motion.

## CASE 3

A 14-year-old male presented with left thigh pain and deformity after falling from a 10-foot-tall coconut tree. Radiographs show a comminuted, primarily reverse oblique subtrochanteric femur fracture with a pertrochanteric extension (Figure 5).

He underwent open reduction, and fixation with a 10-hole PHILOS plate under spinal anesthesia at 15 days post-injury. Intra-operatively, we noted that the fracture was 7.5 cm long. The reduction was maintained with a Lohmann, a Verbrugge, and a Kirschner wire before applying two lag screws outside the PHILOS plate. A total of four locking screws were placed proximal to and five distal to the fracture site. Intra-

operative blood loss was 800 cc, requiring a transfusion of two units of PRBC, and the surgery was done in 3.5 hours. A posterior half-cylinder spica mold was also placed, and the patient was allowed toe-touch weight bearing with crutches.

At six months post-op, the patient was able to ambulate on full weight-bearing without crutches. The range of motion of the hips and knees was full and equal.

## CASE 4

A 13-year-old male presented with right thigh pain and deformity after being hit by a van while riding a motorcycle. Radiographs show a comminuted, subtrochanteric femur fracture.

He underwent open reduction, and fixation with a 10-hole PHILOS plate under spinal-epidural anesthesia at 26 days post-injury. Intra-operatively, we noted that the fracture was 3.5 cm long with a small, comminuted fragment. The reduction was maintained with two Verbrugges. A total of five locking screws were placed proximal to and distal to the fracture site. Intra-operative blood loss was 120 cc, requiring a transfusion of two units of PRBC, and the surgery was done in 4.5 hours. A posterior half-cylinder spica mold was also placed, and the patient was allowed toe-touch weight bearing with crutches.

Radiographs at three months post-op showed callus formation but with a varus angulation of 10 degrees.

At six months post-op, the fracture was united on radiographs, and the patient was able to ambulate with ease and with full range of motion despite the initial varus angulation (Figure 4).



**Figure 4.** *Case 2.* Radiographs at six months post-op show union on both fracture sites and equal femur lengths.



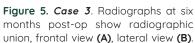
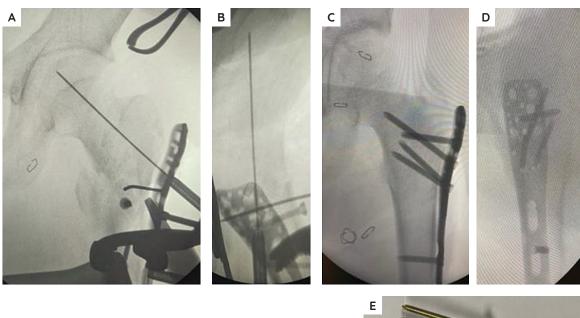
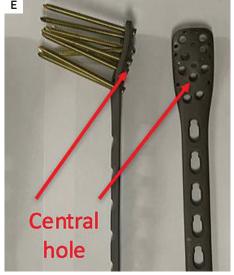




Figure 6. Case 4. Radiographs at six months post-op: fracture has united, frontal view (A), lateral view (B).





**Figure 7.** Proper placement of the central third-row proximal locking screw is most important, aimed along the center of the femoral neck on both AP and lateral views. Frontal view with central pin **(A)**, lateral view with central pin **(B)**, frontal view post-op **(C)**, lateral view post-op **(D)**, frontal and lateral view **(E)** of implant and direction of screws.

## **DISCUSSION**

Management of subtrochanteric fractures in children poses a problem due to the innate instability and increased stress at the subtrochanteric area, and the need to avoid disrupting greater trochanter physis, which contributes 30% of the femur's growth, and 15% of the overall length of the lower extremity.<sup>5</sup>

The pre-bent curve of the PHILOS plate designed for the adult proximal humerus nicely corresponds with the contour of the pediatric patients' proximal femur, which greatly assisted in providing anatomic reduction throughout the length of the entire femur. Its central locked screw angulation of 130 degrees is also placed well into the center of the femoral neck, on both AP and lateral views.

Another benefit of the PHILOS plate was its length; the longest pediatric proximal femur locked plate available in our institution fell short, compared to the 26 cm of the longest PHILOS plate. The overhang or wiper noted on the anterior-

distal femur due to the length of the plate and femoral bowing was insignificant, and there was no implant prominence appreciated clinically.

Comparing the adult proximal femur locking plate and the PHILOS plates, the width of the PHILOS plate better accommodates the width of the pediatric bone among the age range of our patients 8-14 years old. And since there are more screws with varying directions in the PHILOS plate than the adult proximal femur locking plate (9 vs 5), the PHILOS plate is more forgiving and flexible.

The most important step in these surgeries is the proper placement of the central third-row proximal locking screw, aimed along the center of the femoral neck on both AP and lateral views (Figure 7A and 7B). The first most proximal row is usually left empty because these screws angulate outward from the plate, usually beyond the width of the cortical bone (Figure 7C). The second reason why the most proximal row is left empty is that it usually coincides with the greater trochanteric physis and is avoided to prevent physeal injury.

Table 1. Demographic profile of patients and PHILOS plate used in each case

	Age (years)	Sex	Weight (kg)	Days to surgery	Fracture length (cm)	PHILOS size (hole)	Intra-op Blood Loss (cc)	Harris Hip Score 6 mos
1	8	М	38	12	9.5	12	700	91
2	9	F	32	7	8	12	500	91
3	14	М	45	15	7	10	800	88
4	13	М	48	26	3.5	10	120	91

The placement of at least four proximal locking screws is deemed sufficient for maintaining stability.

A full open incision was made in all of the cases due to the long extent of the subtrochanteric fractures averaging 7 cm (range 3.5–9.5 cm) requiring placement of lag screws (Table 1). In hindsight, a minimally invasive plate osteosynthesis (MIPO) technique could have been attempted, to decrease blood loss.

All patients were able to stand full weight bearing by three months post-op, and all fractures united as patients were able to ambulate without difficulty by six months post-op. There was no leg length discrepancy in all cases, and the range of motion for the hips and knees was full and equivalent to the contralateral limb. There were no cases of infection, no complaints of hardware prominence, nor was there a desire from the patients to have the implants removed.

At six months postoperatively, one patient still presented with a slight limp during ambulation; perhaps since this patient was non-compliant with physical therapy and gait retraining. Harris Hip scores at six months post-op were excellent with an average of 90.25 (88–91).

The results of our study are comparable with the largest case series of Gogna et al., in 2014, which studied eight children aged 10–16 years old in terms of union, absence of infection, and excellent Harris Hip scores on final follow-up. Our study differs in that two of our four cases were from a younger age group, ages 8 and 9, and that the subtrochanteric fractures in our study were all comminuted and longer, averaging 7 cm. This shows that the use of the PHILOS plate also yields favorable outcomes in younger patients and complex fracture patterns.

## CONCLUSION

We conclude that the PHILOS plate is a viable option in the management of long, comminuted subtrochanteric femur fractures in the pediatric population because it can accommodate the narrow width of the proximal femur cortical bone while providing adequate stability needed in this area of high stress. Compared to previous literature on the topic, our patients included younger patients ages 8 and 9, and longer, more complex comminuted subtrochanteric fractures up to 9.5 cm in length. No screws violated the greater trochanter physis in all our cases. The authors recommend that more cases be done to add to the growing body of knowledge regarding the matter and that an MIPO technique be attempted to minimize blood loss.

## **ETHICAL CONSIDERATION**

Patient consent forms were obtained before manuscript submission.

## STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

#### **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

## **FUNDING SOURCE**

None.

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## Surgical Treatment and Short-term Outcome for a Refractory Case of Medial Clavicle Osteitis Condensans: A Case Report

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#### **ABSTRACT**

Medial clavicle osteitis condensans presents as pain and swelling in the sternal end of the clavicle with increased bone density in the radiograph. In this case report, we present a 39-year-old woman with right medial clavicle osteitis condensans who underwent several diagnostic procedures with inconclusive findings. Despite various interventions, including IV antibiotics, steroid injections, and IV pain medications, relief was only temporary. Abnormal bone marrow signals involving the sternal end of the right clavicle on further imaging, together with non-response to treatment, prompted surgical intervention. Medial clavicle and first rib resection were performed resulting in significant improvement in pain and function on short-term follow-up.

Keywords. condensing osteitis, medial clavicle osteitis condensans, bone sclerosis

## INTRODUCTION

Condensing osteitis is a rare benign disorder found in multiple bony locations such as the clavicle and the iliac bone. It is characterized as a localized, inflammatory thickening of the affected bone. Although first reported in 1974, medial clavicle osteitis condensans remain a rare entity with few documented cases in the literature. This idiopathic disorder presents as an increase in bone density at the medial clavicle and usually affects women of childbearing age. Clinical manifestations include pain on the abduction of the arm, fullness, and sclerosis over the medial end of the clavicle with sparing of the sternoclavicular joint. The exact etiology of condensing osteitis remains elusive. Patients usually recall no traumatic episode, although possible mechanical stresses related to activity are sometimes implicated. The diagnosis can be difficult because of the non-specific symptoms and imaging that frequently resemble bone malignancies. Treatment modalities range from conservative symptomatic management to surgical excision. In this case report, we present a patient with long-standing medial clavicle osteitis condensans, highlight the clinical and radiological features, describe the treatment, and report the progress of this rare condition.

**CASE** 

This is a case of a 39-year-old woman who presented with pain and swelling localized to the right medial clavicle, with a history of shoulder pain and limitation of motion over the past 16 months. She was initially treated for rotator cuff syndrome and took pain medications which provided minimal to no relief. Subsequent ultrasound revealed

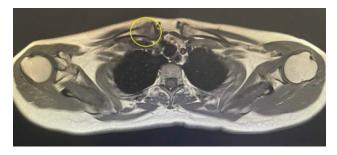
elSSN 2012-3264 (Online)
Printed in the Philippines.
Copyright© 2024 by Dolojan and Sibonga.
Received: April 8, 2024.
Accepted: May 1, 2024.
Published Online: May 5, 2024.
https://doi.org/10.69472/poai.2024.07

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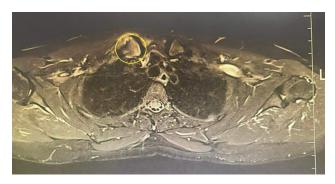
inflammatory processes at the right sternoclavicular region, while a chest CT scan with contrast showed an unremarkable bony thorax, including ribs, vertebrae, and sternum. Physical therapy also failed to alleviate symptoms. Four months before the admission, the pain persisted, accompanied by a palpable enlargement in the medial clavicle and numbness and pins and needles sensation in the right arm. A plain MRI of the right clavicle revealed no focal mass lesion or abnormal fluid collection in the area of interest. EMG-NCV results were normal with no focal entrapment neuropathy and no evidence of brachial plexopathy. Despite treatment with IV antibiotics along with steroid injections and IV pain medication, relief was only temporary. A thoracic outlet syndrome study revealed a possible symptomatic vascular thoracic outlet syndrome on the right arm. The patient underwent intraarticular acromioclavicular, glenohumeral, and sternoclavicular steroid injections which afforded no relief. Further imaging with upper thorax MRI/MRA revealed abnormal bone marrow signals involving the sternal end of the right clavicle and apposing clavicular notch exhibiting a mass effect. Figures 1, 2 and 3 show the axial and coronal cuts of the patient's MRI/MRA. Based on the MRI findings, the lesion was thought to have no overlying soft tissue mass. A core needle biopsy was initially contemplated. However, using a Jamshidi needle biopsy in the medial clavicle could place the subclavian vessels at risk. Given the two options of core needle biopsy and open excision biopsy, the patient opted for the latter, since this offers both diagnostic and therapeutic benefits. She was then admitted for surgical management.

A focused examination of the patient's right shoulder showed swelling and a 3 x 3 cm circumscribed, hard, tender, non-movable mass at the right medial clavicle (Figure 4). The range of motion of the shoulder was limited due to pain. The active and passive range of motion of shoulder abduction and forward flexion were 0–100 degrees. The patient had a 30% neurosensory deficit on the radial, median, and ulnar nerve distribution of the right upper extremity. The Roos and Adson test for thoracic outlet syndrome was positive. The patient's Fil-DASH score was 95 points indicating severe disability. The X-ray of the clavicle in AP and Serendipity view (Figure 5) revealed a cortical irregularity and bone thickening at the right medial clavicle.

The surgical plan for the patient at this point was Medial Clavicle Resection, Right with Possible Stabilization and Application of Iliac Bone graft. The patient was placed in a supine position under general anesthesia. After marking the skin, the incision was made starting from the medial one-third of the clavicle, following the surface of the clavicle medially, extending toward the midline of the sternum (Figure 6). The incision was then deepened to reveal the sternoclavicular joint, anterior surface of the sternum, and medial third of the clavicle. Exophytic changes on the inferior aspect of the medial clavicle and the first rib were removed using rongeur (Figure 7). A length of 0.9 cm of medial clavicle and 0.9 cm of first rib were resected using an oscillating saw and rongeur (Figure 8). The medial clavicle was noted to be stable after



**Figure 1.** T1 weighted axial cut of the upper thorax MRA showing the lesion



**Figure 2.** T2 weighted axial cut of the upper thorax MRA showing the lesion.



Figure 3. T2 weighted coronal cut of the upper thorax MRA.

doing a passive range of motion of the shoulder; it was decided that there was no need for stabilization using a plate. The operative site was washed out with a saline solution and then closed with the insertion of a Jackson-Pratt drain. The final procedure done was a medial clavicle resection, right; first rib resection, right, under general anesthesia.

Post-operative x-ray (Figure 8) confirms the approximately 1 cm shortening from the resection of the clavicle. Bone gram stain and culture studies revealed a light growth of Serratia marcescens. Thus, the patient was also treated with Levofloxacin 500 mg IV once a day for four weeks given on an outpatient basis, until normalization of ESR and CRP.

On the third postoperative day, there was less pain (NRS 6/10), a well-coaptated wound, no discharge, no dehiscence, and an improvement in the neurosensory deficit from 30% to 10% at the radial, median, and ulnar distribution. The JP drain was removed. There were no new subjective complaints,

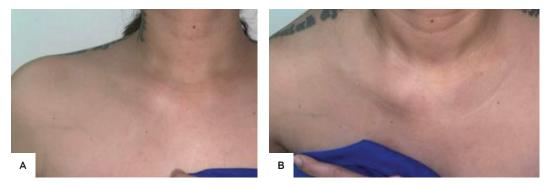


Figure 4. A gross picture of the patient's right clavicle showing frontal (A) and superior (B) view.





Figure 5. Pre-operative x-ray of the clavicle in AP (A) and serendipity (B) view.

hence the patient was discharged. Fil-DASH score at two weeks post-op was 59 points.

At two months post-op, there was a significant decrease in pain (NRS 0-1/10). The patient had a full range of motion of the shoulder (Figure 9) with no noted neurosensory deficit. She was able to do activities of daily living with minimal to no difficulty. Fil-DASH score was 17.5 points indicating significant improvement of the patient's condition. She reported satisfaction with the outcome at five months. Fil-DASH score was 15 points she was able to do activities of daily living with minimal to no difficulty.

Histopathological examination revealed mature woven bone with normocellular marrow, with islands of hyaline cartilage and some areas of fibrosis. There was no evidence of

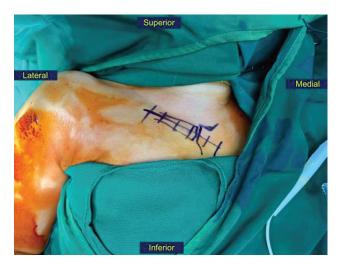


Figure 6. Skin markings over the sternoclavicular area.

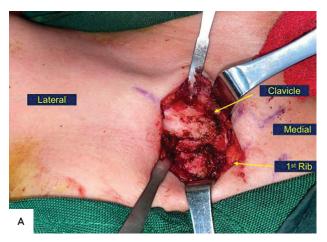
malignancy. These findings, alongside the patient's symptoms and radiographic results, are consistent with other case reports, thus favoring the diagnosis of medial clavicle osteitis condensans.

## **DISCUSSION**

Medial clavicle osteitis condensans is a rare and often overlooked condition characterized by persistent discomfort and bony irregularities at the medial end of the clavicle. It can resemble other more common conditions such as osteo-arthritis or osteomyelitis, thus knowledge about this condition is essential for appropriate diagnosis and management.<sup>1</sup>

The typical presentation of medial clavicle osteitis condensans is persistent, localized pain and tenderness at the sternoclavicular area, which worsens with movement. Patients may also experience limited range of motion and stiffness in the affected shoulder. The pain may radiate to the neck, shoulder, and arm and is frequently described as dull or aching, as experienced by our patient. Without an appropriate diagnosis and course of treatment, symptoms are often chronic and can last for years. <sup>1,2</sup>

Osteoarthritis, bone island, osteomyelitis, septic arthritis, and sternoclavicular joint dislocation are conditions similar to medial clavicle osteitis condensans. The more common conditions should first be ruled out through laboratory tests to ensure appropriate management. X-rays may show sclerotic changes and irregularities at the medial end of the clavicle, while computed tomography (CT) scans and magnetic resonance imaging (MRI) can provide more detailed information about bone morphology and soft tissue involvement.<sup>3,4</sup>



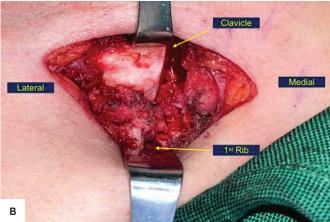


Figure 7. Intraoperative gross picture of the medial clavicle and first rib showing exophytic changes (A); post resection (B).



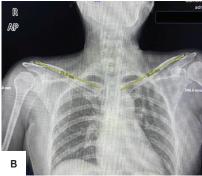




Figure 8. Post-operative x-ray of the clavicle in AP view (A); AP view with measurement confirming resection of 0.9 cm (yellow arrows) (B); serendipity view (C).

In this case, the patient also presented with shoulder pain, paresthesia, and neurosensory deficit on the right upper extremity. These symptoms could be caused by compression of the neurovascular bundle to the upper extremity resulting



Figure 9. Range of motion at 2 months postop showing full shoulder abduction (A) and full shoulder flexion (B).

in thoracic outlet syndrome or pectoralis minor syndrome. This compression could have been caused by the bony overgrowth in the costoclavicular space coming from the medial clavicle and the first rib.5

Treatment for medial clavicle osteitis condensans aims to ease discomfort and enhance functionality. Pain relief and function enhancement are typically pursued through nonsteroidal anti-inflammatory drugs (NSAIDs) and pain medications, coupled with physical therapy to improve range of motion. Occasionally, corticosteroid injections or intraarticular hyaluronic acid might offer supplementary relief. Existing literature has mostly reported improvement after conservative management.<sup>2,6,7</sup> However, there were no case reports available detailing the surgical treatment for medial clavicle osteitis condensans. Surgical measures are seldom necessary but could be considered for refractory cases, notable structural impairment, or instability.<sup>1,4</sup>

In this case report, the patient had a refractory case of medial clavicle osteitis condensans, hence surgical treatment was advised. Pre-operative planning is prepared to address instability that may occur to the remaining clavicle. According to Abbot and Lucas, a resection of the medial clavicle lateral to the costoclavicular ligament would lead to superior displacement of the medial clavicle, alongside pain

and dysfunction.<sup>8</sup> On the other hand, a study done by Bisson et al., has reported that a resection of 1.0 cm in males or 0.9 cm in females would result in minimal or no disruption of the costoclavicular ligament, thus preserving the stability of the clavicle.<sup>9</sup> Having this knowledge, the patient was advised that fixation with a Balser plate, as described by Feng et al., might be needed to restore the stability of the medial clavicle.<sup>10</sup>

Intraoperatively, there were exophytic changes on the inferior aspect of the medial clavicle extending to the first rib. Therefore, abnormal portions of both the clavicle and first rib were resected. A 0.9 cm section was resected, and upon checking the range of motion, the remaining clavicle was stable. Hence, there was no need to apply the Balser plate. Excision of the medial clavicle has provided the patient with significant improvement (even resolving the paresthesia and neurosensory deficit of the upper arm) with almost complete relief at five months post-operation.

## **CONCLUSION**

Although medial clavicle osteitis condensans is rare, it warrants consideration when evaluating sclerotic lesions near the medial end of the clavicle, especially in young and middle-aged women. Identifying this condition can prevent unnecessary tests in pursuit of a diagnosis. Understanding the clinical characteristics, diagnostic approaches, and treatment strategies is crucial for delivering the best possible care for affected individuals. In this case, surgical resection of the medial clavicle resulted in significant relief of symptoms.

## **ETHICAL CONSIDERATION**

Patient consent was obtained before submission of the manuscript.

## STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

## **AUTHOR DISCLOSURE**

The authors declared no conflict of interest.

## **FUNDING SOURCE**

None.

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## CASE REPORT / CASE SERIES









# A Case Series on Arthroscopic Valvulectomy of Symptomatic Popliteal Cysts in Elderly Patients Using a Modified Gillquist Maneuver

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#### **ABSTRACT**

The primary objective of this study was to assess the outcomes of arthroscopic management of popliteal cysts using the modified Gillquist maneuver for visualization. The original Gillquist maneuver was originally developed to gain access to the posterior knee compartment. However, there were drawbacks, including blind arthroscope insertion, which could potentially lead to unnecessary trauma to the knee and the risk of arthroscope damage. The modified Gillquist maneuver was introduced to overcome these limitations, offering the advantage of direct visualization during arthroscope insertion into the posterior compartment through either the anterolateral or anteromedial portals. This single-institution case series focused on a cohort of five patients aged over 60 diagnosed with unilateral popliteal cysts. These individuals underwent arthroscopic valvulectomy using the modified Gillquist maneuver. The results of the procedure were highly encouraging, providing symptomatic relief for all five patients. Significant improvements were observed in clinical metrics, including the Numeric Rating Scale (NRS), Rauschning and Lindgren Criteria, Lysholm Score, and Knee Range of Motion during a follow-up period of up to 24 months. These promising outcomes highlight the potential efficacy of the modified Gillquist maneuver as a viable surgical approach for managing popliteal cysts in the elderly population.

Keywords. popliteal cysts, Baker's cyst, arthroscopy, Gillquist, knee

## **INTRODUCTION**

Popliteal cysts, more commonly known as Baker's cysts, are fluid-filled masses found at the posterior aspect of the knee. They were initially described by Adams in 1840 and further studied by Baker in 1877.<sup>1,2</sup> These cysts develop due to a connection between the knee joint and a bursa located between the tendons of the gastrocnemius and semitendinosus muscle.<sup>3</sup> In approximately 50% of individuals, there is a one-way valve formed by these tendons in the knee. This valve opens when the knee is flexed and closes during knee extension. The difference in pressure within the knee joint between partial flexion and extension allows fluid to flow into both the knee joint and the bursa.4 Gastrocnemius-semimembranosus bursa enlargement due to muscle contractions and joint capsule herniation into the popliteal fossa are two other factors that contribute to cyst formation.5 The prevalence rate of popliteal cysts varies depending on the method of diagnosis. A study that used ultrasound to identify Baker's cysts in individuals with posterior knee pain found an incidence rate of 25%.6 Another study that used magnetic resonance imaging (MRI) reported incidence rates ranging from 5% to 18%.7

The clinical presentation of Baker's cyst varies. Some patients may not experience any symptoms and may only discover the cyst incidentally during a knee examination or imaging procedures like MRI. Others may exhibit symptoms such

elSSN 2012-3264 (Online) Printed in the Philippines. Copyright© 2024 by Braganza et al. Received: October 20, 2023. Accepted: December 9, 2023. Published Online: March 15, 2024. https://doi.org/10.69472/poai.2024.09

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as posterior knee pain, tightness, or discomfort. They might display Foucher's sign, where the mass is larger or firmer during knee extension and less so during knee flexion. Some patients might also show Homan's sign, which is calf pain upon dorsiflexion of the foot. If the cyst ruptures, patients may experience sharp pain, swelling, redness, and a sensation resembling fluid running down the calf.<sup>5</sup> During a physical examination, a physician may observe a prominent, fluid-filled mass at the popliteal region. This swelling might be more pronounced when the knee is extended and less so when the knee is flexed.

Various imaging methods can help diagnose a popliteal cyst. MRI remains the preferred and most accurate method for diagnosis, as it not only confirms the presence but also distinguishes it from other conditions and identifies any additional soft tissue irregularities.<sup>8</sup> On the other hand, ultrasound can serve as a more economical screening or diagnostic tool when an in-depth examination of the joint's interior is not deemed necessary.<sup>9</sup>

Treatment can be categorized into two main approaches: non-operative and operative management. Non-operative treatments include nonsteroidal anti-inflammatory drugs (NSAIDs), activity modification and rest, physical therapy, aspiration, and steroid injections. However, if the popliteal cyst persists or recurs, surgical intervention may be necessary, which can be done through either open excision or arthroscopy.<sup>4</sup>

Open excision of the popliteal cyst has been associated with a high rate of recurrence, emphasizing the need to address underlying intra-articular issues. A meta-analysis conducted by Han et al. found that arthroscopic treatment of popliteal cysts, both with and without cyst removal, yielded relatively better outcomes with low recurrence rates. Interestingly, the study also found that although arthroscopic treatment with cystectomy led to a lower recurrence rate, it was associated with a higher rate of complications compared to the group that did not have cyst removal.<sup>10</sup>

Rauschning in 1980 first described using the criteria to describe the knee symptoms of patients with popliteal cysts. <sup>11</sup> In 1982, Lysholm developed a scoring scale that monitored functional outcomes of patients who underwent a form of knee ligament surgery. <sup>12</sup> These scores have been used to evaluate outcomes in patients with symptomatic popliteal cysts who underwent arthroscopic cystectomy and valve excision. <sup>13</sup> Malinowski et al., concluded that there was an improvement in the postoperative Rauschning and Lindgren Criteria and Lysholm in patients who underwent arthroscopic cystectomy and valve excision of the symptomatic popliteal cyst.

In 1979, Gilquist et al., proposed a transpatellar tendon portal as an alternative to Johnson's posteromedial portal. This transpatellar tendon portal enables the surgeon to assess the posteromedial ligaments and eliminates the need for a secondary portal, unlike the posteromedial portal.<sup>14</sup>

The modified Gilquist maneuver involves using either the anterolateral or the anteromedial portal for arthroscope insertion. Lee et al. summarized the sequence of the modified Gilquist maneuver in six steps in 2019.<sup>15</sup>

The first step involves placing the knee in 90 degrees of flexion and positioning the arthroscope in the anteromedial portal to visualize the intercondylar notch. In the second step, the switching stick is inserted through the anterolateral portal between the lateral border of the medial femoral condyle and the medial border of the posterior cruciate ligament. It is advanced until the switching stick enters the posteromedial compartment, ensuring direct visualization to minimize the risk of iatrogenic injury. For the third step, the camera and trocar are removed from the anteromedial portal and separated. In the fourth step, the trocar sleeve is reintroduced over the switching stick in the anterolateral portal, serving as a guide to the posteromedial compartment. In the fifth step, the switching stick is withdrawn from the trocar sleeve. In the sixth step, the camera is reinserted through the trocar sleeve in the anterolateral portal to visualize the posteromedial compartment.

This modification minimizes trauma to the surrounding structures within the knee joint and provides the benefit of direct visualization of the instrument in the posteromedial compartment.<sup>15</sup>

## SURGICAL TECHNIQUE

Under regional or epidural anesthesia, the patient was positioned supine with the affected lower limb flexed at the knee to a 90-degree angle (Figure 1). The surgeon and the assistant were positioned on the side of the affected extremity. The arthroscopy towers were placed on the opposite side of the surgeon. The scrub nurse, scrub trolley, and graft preparation trolley were positioned at the end of the operating table (Figure 2). The procedure involves utilizing three portals: the anteromedial, the anterolateral, and the posteromedial.



Figure 1. Position of the patient on the operating table.

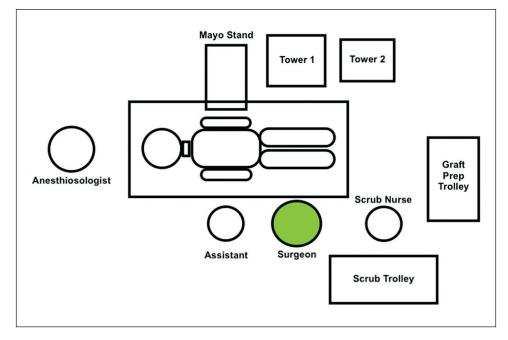


Figure 2. Operating room set-up.

To create the anterolateral portal, transverse incisions were made in the skin approximately 1 cm lateral to the lateral margin of the patellar tendon and 1 cm superior to the joint line. For the anteromedial portal, incisions were made at or 1 cm medial to the medial margin of the patellar tendon and 1 cm superior to the joint line. A 30-degree arthroscope was employed for the procedure, with a 70-degree arthroscope prepared as an alternative if necessary.

Initially, diagnostic arthroscopy was conducted through the standard anterolateral and anteromedial portals. Subsequently, the posterior compartment of the knee was visualized using the modified Gillquist maneuver.

To minimize potential harm to surrounding structures and to facilitate the precise placement of the camera for direct visualization of the posteromedial compartment, a modified Gillquist maneuver was employed. This maneuver involved flexing the knee to a 90-degree angle and introducing the arthroscope through the anterolateral portal to view the intercondylar notch.

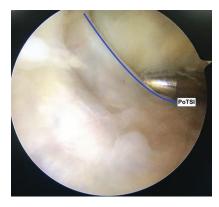
Once the region was cleared of soft tissue, a switching stick was inserted through the anteromedial portal, between the lateral edge of the medial femoral condyle and the inner edge of the posterior cruciate ligament (PCL), and advanced to the posteromedial compartment. The posteromedial compartment was identified by the posterior transverse synovial infold (PoTSI), a transverse infold overlying the medial head of the gastrocnemius (Figure 3). Subsequently, the camera and trocar were withdrawn through the anterolateral portal, and a cannula was threaded over the switching stick located at the anteromedial portal. The camera was then inserted into the anteromedial portal, enabling further examination of the posterior compartment.

The posteromedial portal was established by locating a specific soft region between the medial collateral ligament, the medial head of the gastrocnemius muscle, and the tendon of the semimembranosus defined by Lanham et al., as 2.5 cm inferior and 2.5 cm posterior to the medial femoral epicondyle, with the knee in 90 degrees of flexion.<sup>17</sup> An 18-gauge needle was carefully positioned at this identified soft spot and was visualized intraoperatively (Figure 4). A longitudinal incision was made in the direction of the previously placed cannula, and the portal was created while being directly guided by transillumination.

A blunt trocar was then introduced into the posteromedial portal. The capsular fold and the valve-like connection between the cyst and the knee joint were identified. Once the capsular fold was pinpointed, an obturator was inserted to open the valve, and valvulectomy was done with a shaver. The successful completion of the valvulectomy was confirmed by observing a release of light-yellow fluid from the cyst noted to be oily and thick, as well as the visualization of the medial head of the gastrocnemius through the anterolateral portal (Figure 5). This transformed the previously one-way valve into a two-way valve. Any intra-articular issues were subsequently managed following established protocols.

## **CASES**

Patient A was an 83-year-old woman complaining of chronic intermittent posterior knee pain at the posterior aspect of her right knee for three years with a progressively enlarging posterior knee fluctuant mass that is prominent upon knee extension. We noted a palpable posterior knee mass, with a positive Foucher's sign and a negative Homan's sign. Ultrasound revealed a popliteal cyst, and radiographs confirmed the presence of concurrent knee osteoarthritis



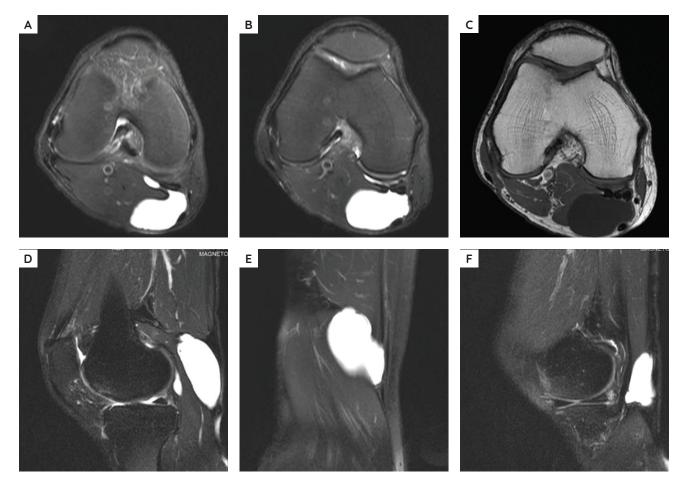
**Figure 3.** Intraoperative visualization of the placement of the trocar along the Posterior Transverse Synovial Infold (PoTSI).



**Figure 4.** Intraoperative visualization of an 18-gauge needle within the posteromedial compartment of the knee.



**Figure 5.** Intraoperative visualization of the egress of the light-yellow fluid from the cyst.



**Figure 6.** 68-year-old male presenting with posterior knee pain and popliteal mass on the left knee. T2 weighted magnetic resonance images (MRI) cross section presenting a popliteal mass at the posteromedial aspect of left knee **(A-B)**, T1 weighted MRI cross section presenting a popliteal mass at the posteromedial aspect of left knee **(C)**. T2 weighted MRI sagittal section presenting a popliteal mass **(D-F)**.

(Kellgren and Lawrence III). Initially, the patient was recommended non-surgical management, including NSAIDs and physical therapy. However, as time passed, the patient experienced worsening knee pain, leading to a gradual restriction in daily activities, particularly those involving knee movement and flexion. Eventually, the pain became so severe that the patient was unable to perform basic household

tasks for an extended period. At the initial consultation, the patient rated their pain on the Numeric Rating Scale as 7 out of 10, and their condition was classified as Grade 2 according to the Rauschning and Lindgren Criteria. Subsequently, the patient was advised to undergo arthroscopic debridement, synovectomy, and valvulectomy to address the popliteal cyst in their right knee. After the procedure, the patient was

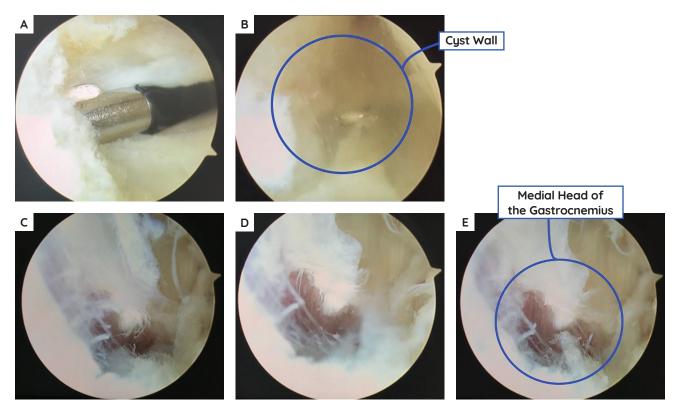


Figure 7. View from anteromedial portal. Insertion of shaver from the posteromedial portal from the posteromedial compartment towards the knee joint (A); Egress of light yellow fluid from the popliteal cyst towards the knee joint upon initiation of valvulectomy (B); Enlargement of the communication between the posteromedial compartment and knee joint (C-E); Visualization of the gastrocnemius from the anteromedial viewing portal.

instructed to start bearing weight on the knee immediately on the first day postoperatively. They were also prescribed a regimen of progressive knee range of motion exercises and exercises to activate and strengthen the quadriceps muscles and their outcomes were subsequently monitored (Table 1).

Patient B was a 63-year-old man who came in for a 4-year history of a palpable mass at the posterior aspect of the right knee accompanied by posterior knee pain upon knee flexion of more than 90 degrees. The mass was full on knee extension and soft on flexion. The patient had a positive Foucher's sign and a negative Homan's sign. An MRI revealed a popliteal cyst and chondromalacia patellae in the right knee. Initially, the patient was advised to undergo physical therapy and take NSAIDs to relieve the pain. Over time, there was a gradual increase in the size of the palpable mass, accompanied by occasional pain even when at rest. During the most painful episodes, the patient reported a pain level of 8 out of 10 on the NRS. This discomfort eventually led to the patient being unable to perform their job. Subsequently, the patient underwent arthroscopic debridement, valvulectomy of the popliteal cyst, and chondroplasty of the patella in the right knee. Starting on the first day after the operation, the patient was advised to bear weight on the knee immediately and begin a series of progressive range of motion exercises and the postoperative outcomes were monitored during the follow up period (Table 1).

Patient C was a 68-year-old man complaining of a 2-year history of posterior right knee pain and limitation of knee flexion beyond 90 degrees due to a popliteal mass accompanied by dull pain during flexion. The patient had a negative Homan's sign and a positive Foucher's sign. An MRI revealed the existence of a popliteal cyst in the right knee and chondromalacia in the patellofemoral region (Figure 6). Initially, the patient was recommended to undergo physical therapy and prescribed NSAIDs as a form of treatment. Despite diligently following the physical therapy regimen, the patient experienced a gradual increase in pain, eventually reaching a pain level of 8 out of 10 on the NRS, accompanied by limitations in their daily activities. Subsequently, the patient underwent arthroscopic debridement, chondroplasty of the patella, and valvulectomy of the popliteal cyst in the right knee (Figure 7).

Patient D was a 61-year-old man complaining of a 10-year history of posterior right knee mass with no limitation in range of motion until two months before surgery. He had a positive Foucher's sign and a negative Homan's sign. An MRI revealed a popliteal cyst measuring 7.2 x 2.4 x 4.1 cm in size. The presence of this cyst resulted in restricted knee flexion, preventing the patient from bending their knee beyond 90 degrees. Additionally, the patient experienced pain, which reached a level of 6 out of 10 on the NRS, significantly impeding their ability to walk. Consequently, the patient underwent arthroscopic valvulectomy to remove the popliteal cyst from their right knee.

Table 1. Demographics

Outcome		Pre-operative	3 months	6 months	9 months	12 months	24 months
Numeric Rating Scale (NRS)	Α	7/10	3/10	2/10	2/10	2/10 (intermittent)	2/10 (intermittent)
	В	8/10	2/10	2/10	1/10 (intermittent)	1/10 (intermittent)	1/10 (intermittent)
	С	8/10	2/10	2/10	1/10	n/a	n/a
	D	6/10	3/10	2/10	n/a	n/a	n/a
	Ε	6/10	2/10	2/10	n/a	n/a	n/a
Rauschning and	Α	Grade II	Grade 0				
Lindgren criteria	В	Grade III	Grade 0				
	С	Grade III	Grade I	Grade I	Grade 0	n/a	n/a
	D	Grade III	Grade I	Grade I	n/a	n/a	n/a
	Ε	Grade II	Grade I	Grade 0	n/a	n/a	n/a
Lysholm score	Α	35	68	74	68	82	82
	В	38	84	88	88	92	92
	С	37	82	84	82	n/a	n/a
	D	34	81	82	n/a	n/a	n/a
	Ε	38	82	82	n/a	n/a	n/a
Knee range of motion	Α	Active: 5°–100° Passive: 5°–110°	Active: 5°-110° Passive: 5°-120°	Active: 5°–110° Passive: 5°–120°	Active: 5°–115° Passive: 5°–120°	Active: 5°–115° Passive: 5°–120°	Active: 5°–115° Passive: 5°–120°
	В	Active: 5°–90° Passive: 0°–100°	Active: 5°-100° Passive: 0°-110°	Active: 5°–105° Passive: 0°–115°	Active: 5°–110° Passive: 0°–120°	Active: 5°–110° Passive: 0°–120°	Active: 5°–110° Passive: 0°–120°
	С	Active: 5°–90° Passive: 5°–100°	Active: 5°-100° Passive: 0°-110°	Active: 5°–105° Passive: 0°–110°	Active: 5°–110° Passive: 0°–115°	n/a	n/a
	D	Active: 5°–90° Passive: 0°–105°	Active: 5°-100° Passive: 0°-110°	Active: 5°–115° Passive: 0°–125°	n/a	n/a	n/a
	Ε	Active: 5°–110° Passive: 5°–115°	Active: 5°-115° Passive: 5°-120°	Active: 5°–120° Passive: 0°–130°	n/a	n/a	n/a

Patient E was a 63-year-old woman complaining of a 1-year history of a palpable posterior left knee mass with accompanying pain and limitation in knee flexion beyond 110 degrees. There was a palpable 6 x 4 cm mass within the popliteal region (Figure 8A). The patient had a positive Foucher's sign and a negative Homan's sign. An ultrasound of the posterior aspect of the left knee confirmed the presence of a popliteal cyst measuring 5.7 x 4.1 x 3.3 cm. The patient experienced discomfort, with a pain level reaching 6 out of 10 on the NRS, and the noticeable growth of the palpable popliteal mass. Consequently, the decision was made to undergo arthroscopic valvulectomy to remove the popliteal cyst from the left knee. Six months postoperatively, there was no noted recurrence of the palpable posterior left knee mass (Figure 8B) and no post-operative wound healing complications (Figure 9). The patient presented with an improving knee range of motion (Figure 10).

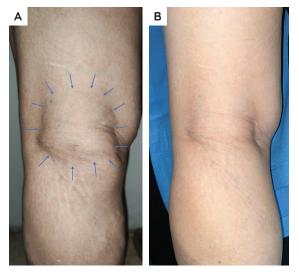
#### **DISCUSSION**

In our series, the cases involved arthroscopic procedures to enlarge the connection between the popliteal cyst and the knee joint while simultaneously addressing intra-articular issues, all without removing the cyst wall. Although open excision of the popliteal cyst is an option for surgical treatment, some studies suggest higher rates of recurrence.<sup>4,11</sup>

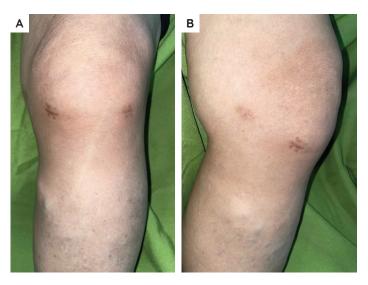
Literature is inconclusive regarding the optimal treatment for popliteal cysts due to a lack of high-quality evidence. Current surgical techniques primarily focus on enlarging the communication between the cyst and the knee joint and addressing intra-articular problems. According to the same study, this procedure has a success rate of 96.7%. The study also identified the potential need for cyst wall resection which yields a slightly higher success rate. More extended follow-up studies are required to determine the clear advantage of cyst wall resection over procedures that do not involve cyst wall removal.

Arthroscopic management is also a viable treatment option because it is minimally invasive, associated with fewer complications, and allows for early rehabilitation. This approach also enables cyst decompression while simultaneously addressing knee joint issues. The main goal of the surgery is to convert the one-way valve into a two-way valve, allowing trapped fluid within the cyst to pass into the knee joint where the fluid can be reabsorbed. Surgical failures have previously been attributed to the inability to convert the unidirectional flow of fluid between the two compartments. On the surgery is the convert the unidirectional flow of fluid between the two compartments.

The patients in our series underwent valvulectomy to enlarge the communication between the two compartments. The success of this procedure is determined by directly visualizing the medial gastrocnemius through the anteromedial portal and observing a release of light-yellow fluid from the cyst. A study by Ahn et al. found that a 5mm widening of the valve is sufficient to convert the unidirectional flow into a bidirectional flow.<sup>21</sup>



**Figure 8.** Examination of the popliteal region of Patient E. Preoperatively, presence of a 6 x 4 cm palpable mass on the popliteal region of the left knee **(A)**. Postoperatively, at 6 months follow-up, there was no recurrence of the palpable mass on the popliteal region of the left knee **(B)**.



**Figure 9.** Postoperative examination of the postoperative site of Patient E at 6 months follow up. Postoperative anteromedial and anterolateral incision sites **(A)**. Postoperative posteromedial incision site **(B)**.

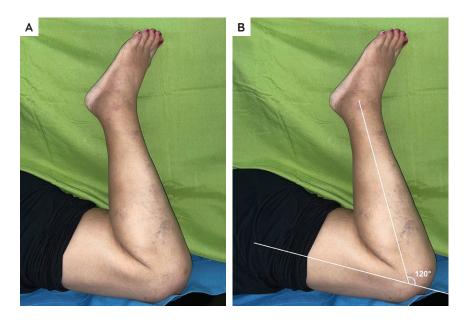


Figure 10. Postoperative examination of Patient E at 6 months follow up. Active range of motion of the left knee without the measurement (A). Improvement of the range of motion with the active knee flexion of 120° measured with a goniometer and confirmed with a digital application (B).

In our surgical approach, we incorporated a modification of the Gillquist maneuver. The original method involved creating a portal through the patellar tendon and inserting the arthroscope between the medial femoral condyle and the posterior cruciate ligament to access the posteromedial compartment. This maneuver was performed blindly and had a history of causing hardware issues, such as camera breakage, which could compromise the procedure.<sup>15</sup>

In our technique, we employ both an anterolateral and an anteromedial portal. We use a switching stick to guide the arthroscope into the posteromedial compartment, and this is done under direct visualization provided by the camera. The advantage of using a switching stick lies in the tapered design and smaller caliber compared to the trocar used for placement. This minimizes the risk of trauma as the arthroscope is

passed from the anteromedial portal to the posteromedial compartment.

Furthermore, our approach prioritizes the placement and visualization of the posteromedial compartment via the anteromedial portal. This method allows unobstructed access to the compartment and offers easier visualization using a 30-degree scope, eliminating the need to traverse any midline structures, such as the anterior cruciate ligament, as required when visualizing from the anterolateral portal.

The results from our series demonstrated favorable outcomes when evaluating patients' progress at 3, 6, 9, 12, and 24 months post-surgery (Table 2). These positive outcomes were reflected in improvements in the NRS, Lysholm score, and Rauschning and Lindgren Criteria.

Female

Patient Gender Symptomatic duration (year) Diagnostic modality Aae 83 Female Ultrasound Α В 63 Male 4 Magnetic Resonance Imaging С 68 Male 2 Magnetic Resonance Imaging D 10 61 Male Magnetic Resonance Imaging

Table 2. Outcome measures (NRS, Rauschning and Lindgren, Lysholm Score, Knee Range of Motion) of patients A to E

The success observed in our series can be attributed to the surgical focus mentioned earlier, which primarily involves enlarging the valve to convert the valve from a unidirectional to a bidirectional one. Additionally, our series also addressed any intra-articular issues present in the patients. After surgery, the rehabilitation process centered on exercises aimed at improving the range of motion and strengthening the quadriceps. Importantly, all patients showed improvement in their Numeric Rating Scale, Rauschning and Lindgren Criteria grade, Lysholm Score, and Knee Range of Motion.

63

F

Patient A had a slightly worse Lysholm score compared to patients B and C. This difference can be attributed to the concurrent presence of osteoarthritis in patient A, which likely influenced their pain levels and activity-related measurements within the outcome assessment.

This case series has certain limitations, including the small number of patients enrolled and the short follow-up period of between 9 and 24 months. This shorter follow-up period was a result of patients' decreased compliance with follow-up appointments, largely influenced by the disruptions caused by the COVID-19 pandemic.

Our case series could serve as a framework for a more extensive study. Such a study could specifically investigate the efficacy of utilizing the anteromedial portal for visualizing the posteromedial compartment and the benefits of employing a switching stick to minimize camera placement-related trauma.

#### **CONCLUSION**

A definitive standard treatment for popliteal cysts has yet to be established due to the absence of high-quality evidence demonstrating the clear advantages of one approach over another. However, the literature recognizes that arthroscopic management of popliteal cysts appears to be both minimally invasive and effective.

The primary objective of surgical management is to convert the unidirectional valve into a bidirectional one while simultaneously addressing any intra-articular issues, to reduce the likelihood of recurrence. In our case series, we used a modified Gillquist maneuver using a switching stick to minimize tissue trauma during the procedure. Additionally, we adopted the anteromedial portal as the primary viewing portal, providing straightforward access to the posteromedial compartment without the need to navigate obstructive midline

structures, which would be the case with an anterolateral viewing portal.

Ultrasound

Notably, our case series yielded positive outcomes as evidenced by improvements in the Numeric Rating Scale, Rauschning and Lindgren Criteria, Lysholm Score, and Knee Range of Motion. Although our series had a limited number of patients, this study may serve as a foundational reference point for future research endeavors focusing on this technique.

#### **ETHICAL CONSIDERATION**

Patient consent forms were obtained before manuscript submission.

#### STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

#### **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

#### **FUNDING SOURCE**

None.

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# SYSTEMATIC REVIEW









# Clinical and Radiographic Predictors of Deterioration in Mild Cervical Spondylotic Myelopathy: A Systematic Review

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#### **ABSTRACT**

**Background and Objectives.** The "best" approach for mild cervical spondylotic myelopathy (CSM) is still unclear, and the decision to perform outright surgery remains a topic of debate. Therefore, identifying clinical and imaging predictors of deterioration is crucial.

**Methodology.** This study followed the PRISMA Guidelines and reviewed published articles from 2000 to 2023 that involved adult patients with asymptomatic spondylotic cord compression and/or mild CSM who underwent conservative management. The search was conducted in MEDLINE via Pubmed, Cochrane Central Register of Controlled Trials, Herding Plus, Embase, and Google Scholar. Patient demographics, neurologic outcome, and clinical and imaging predictors were examined. We assessed study quality using the Newcastle-Ottawa Scale (NOS) for observational studies. We reported statistical data as presented and calculated RRs or ORs if not provided. Evidence quality was evaluated using the GRADE approach.

**Results.** Twelve studies were included, consisting of 1,046 patients. Cervical radiculopathy, electrophysiological abnormalities (EMG, SEP, MEP), decreased Torg ratio <0.80, cervical range of motion of >50°, and cervical instability (slippage >2 mm or segmental kyphosis) were significantly associated with myelopathy progression. MRI T2 hyperintensity of the spinal cord was associated with poor outcomes and delayed development of myelopathy. Furthermore, CSF column diameter, circumferential cord compression, cord T1 angular deformity, cross-sectional area (CSA) <70.1 mm², and cord compression ratio <0.4 were independent predictors of developing myelopathy. Progression was associated more with focal than with diffuse disc herniation.

**Conclusion.** Early recognition of clinical features and imaging predictors of deterioration may help clinicians decide when to do early surgery in patients with mild CSM. Consensus is still needed on the role of surgery in patients with mild CSM. Patients may exhibit improvement, stability, or deterioration following conservative measures.

Keywords. cervical, myelopathy, deterioration, systematic review, surgery

# **KEY POINTS**

- The natural progression of mild CSM has not been clear.
   There are no definitive recommendations regarding the need for surgery for its management.
- Identifying clinical and imaging predictors of deterioration can help clinicians anticipate symptom progression. Early recognition of these predictors can also assist clinicians in deciding on initiating early surgery, avoiding neurologic deterioration.
- Consensus is still needed on the role of surgery in patients with mild CSM. Patients may exhibit improvement, stability, or deterioration following conservative measures.

elSSN 2012-3264 (Online)
Printed in the Philippines.
Copyright© 2024 by Foscablo et al.
Received: February 27, 2027.
Accepted: April 15, 2024.
Published Online: May 3, 2024.
https://doi.org/10.69472/poai.2024.10

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#### INTRODUCTION

Cervical spondylotic myelopathy (CSM) is a condition that develops from the degenerative processes of the spine. These processes lead to facet hypertrophy, disc degeneration, vertebral subluxation, and ossification of ligamentous structures. Narrowing of the spinal canal can cause spinal cord compression, leading to vascular compromise and potentially causing irreversible damage to the involved area. Surgery is recommended for moderate to severe CSM, while its role is unclear in mild CSM. Moreover, studies showed that rigorous conservative management can be as effective as surgery in this population. 4-6

The decision to proceed with surgery remains a dilemma. Doing aggressive surgery may result in unnecessary risks and complications. Conversely, patients undergoing conservative

Table 1. Nurick arade<sup>24</sup>

Nurick Grade	Clinical Presentation			
0	Root signs or symptoms. No evidence of cord involvement			
1	Signs of cord involvement. Normal gait.			
2	Mild gait abnormality. Able to be employed.			
3	Gait abnormality prevents employment.			
4	Able to ambulate only with assistance.			
5	Chair bound or bedridden.			

management may experience neurologic deterioration.<sup>6</sup> Therefore, early detection of those at risk of progression and those who will benefit from early surgery is paramount. Likewise, it is crucial to identify the clinical and imaging predictors of deterioration.

This study was conducted to determine the clinical and imaging predictors of neurologic deterioration in patients with mild CSM. Moreover, this will also guide clinicians in identifying patients at risk of deterioration and those who might benefit from early surgery. The results should contribute to clinicians' decision-making when treating this patient population.

# **METHODOLOGY**

This systematic review used the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-analyses) Guidelines.

#### Eligibility criteria

This study reviewed published articles from 2000 to 2023 that involved adults with asymptomatic spondylotic cord compression and mild CSM (Nurick I-II, mJOA 15-17, JOA>13) (Tables 1 to 3) who underwent conservative management. The following were excluded: 1) manuscripts involving moderate to severe CSM; 2) case series/case reports; 3) conference or poster abstracts; and 4) studies published in languages other than English.

Table 2. Modified Japanese Orthopedic Association (mJOA) Score<sup>25</sup>

Category	Score	Description
Upper Extremity	0	Unable to move hands
Motor Subscore (/5)	1	Unable to eat spoon nut able to move hands
(/3)	2	Unable to button a shirt but able to eat with a spoon
	3	Able to button a shirt with great difficulty
	4	Able to button a shirt with mild difficulty OR other mild fine motor dysfunction (marked handwriting change, frequent dropping of objects, difficulty clasping jewelry, etc.)
	5	Normal hand coordination
Lower Extremity	0	Complete loss of movement and sensation
Motor Subscore (/7)	1	Complete loss of movement, some sensation present
	2	Inability to walk but some movement
	3	Able to walk on flat ground with walking aid
	4	Able to walk without walking aid, but must hold a handrail on stairs
	5	Moderate to severe walking imbalance but able to perform stairs without handrail
	6	Mild imbalance when standing OR walking
	7	Normal walking
Upper Extremity	0	Complete loss of hand sensation
Sensory Subscore (/3)	1	Severe loss of hand sensation OR pain
(, , ,	2	Mild loss of hand sensation
	3	Normal hand sensation
Urinary Function	0	Inability to urinate voluntarily (requiring catheterization)
Subscore (/3)	1	Frequent urinary incontinence (more than once per month)
	2	Urinary urgency OR occasional stress incontinence (less than once per month)
	3	Normal urinary function
Mild	15-17	
Moderate	12-14	
Severe	<14	

Table 3. Japanese Orthopaedic Association Score<sup>26</sup>

Upper Extrem	ity
0	Unable to feed oneself with any tableware including chopsticks, spoon, or fork, and/or unable to fasten buttons of any size
1	Possible to eat with spoon but not with chopsticks
2	Possible to eat with chopstick, but inadequate
3	Possible to eat with chopstick, but awkward
4	Normal
Lower extrem	ity
0	Unable to stand up and walk by any means
1	Need cane or aid on flat ground
2	Need cane or aid only on stairs
3	Possible to walk without cane or aid, but slow
4	Normal
ensory function	1
Upper extrem	ity
0	Apparent sensory loss
1	Minimal sensory loss
2	Normal
Trunk	
0	Apparent sensory loss
1	Minimal sensory loss
2	Normal
Lower extrem	ity
0	Apparent sensory loss
1	Minimal sensory loss
2	Normal
ladder function	
0	Complete uninary retention
1	Severe disturbance
2	Mild disturbance
3	Normal
Mild	>13
Moderate	9-13
Severe	<9

# Search strategy

An electronic search in MEDLINE via Pubmed, Cochrane Central Register of Controlled Trials, Herdin Plus, Embase, and Google Scholar was done, with the following search terms: (Asymptomatic spondylotic compression, mild cervical spondylotic myelopathy, degenerative cervical myelopathy, clinical predictors, imaging predictors, predictive factors of deterioration) AND (mild cervical spondylotic myelopathy management, deterioration, progression). The search used Boolean operators such as AND and OR to combine these terms effectively. Database-specific limiters for publication dates were used. A bibliography search was done within the references of included studies. Hand-searching of printed journals was not done.

#### Selection of studies

Three authors (JF, JM, IS) independently screened the titles and abstracts. Eligible full-text articles were retrieved. Full-

text copies of potentially relevant papers selected by at least one author were also reviewed. Articles that met the inclusion criteria were assessed independently, with any inconsistencies resolved through consensus.

#### Data extraction and management

The primary outcome for analysis was neurologic status on the initial and follow-up period (graded by mJOA, JOA, or Nurick grading), and clinical and imaging predictors of deterioration in mild CSM patients. Study title, author, date of publication, country where the study was conducted, study design, patient demographics, factors predicting deterioration, follow-up duration, and summary of results were extracted using a standardized extraction form. Data was independently retrieved and assessed by the three authors. Data was crosschecked, and discrepancies were resolved by a fourth author (RT).

#### Assessment of risk of bias

For the methodological quality of the individual studies, the Newcastle Ottawa Scale (NOS) for observational studies was used to score representativeness, sample size, ascertainment of exposure, and outcome. A score of 8–9 represents a low risk of bias; 6–7, a medium risk of bias; and 5 or lower, a high risk of bias. A score of at least 6 was considered acceptable. The scoring was done by three authors (JF, JM, IM). If a consensus was not reached, the fourth author (RT) settled the issue (Table 4).

#### **Data synthesis**

Studies that fulfilled the inclusion criteria were eligible for synthesis. Descriptive statistics for all studies were reported as is because there was variability in the reporting of prognostic factors and frequency of outcomes. For prognostic factors that were reported as proportions (e.g., percentage of the sample with a Pavlov ratio <0.8), we reported the calculated relative risks (RRs) or odds ratio (OR) and their 95% CIs if not provided by the authors. For continuous prognostic factors (e.g., neck range of motion [ROM]), we reported analytical statistics as detailed by the authors. We recorded comparative statistics if included in the reviewed articles. All calculations were performed using the Statistical Packages for the Social Sciences (SPSS version 29). In case of missing data or if raw data was unavailable, we tried contacting the corresponding authors through the email address listed in their article. We used narrative synthesis to summarize the results for data that could not be retrieved. Due to the heterogeneity of the included articles, meta-analysis was not conducted. Therefore, a qualitative synthesis was done (Table 5).

# Strength of evidence

We used the GRADE (Grades of Recommendation, Assessment, Development, and Evaluation) framework to assess the overall quality of evidence. Five essential factors

Table 4. Newcastle-Ottawa scale for risk-of-bias

Studies		Selection			Compo	Comparability Outcome				Score
Bednarik (2004) <sup>2</sup>	*	*	*	*	*	*	*	*	*	9
Bednarik (2008) <sup>8</sup>	*	*	*	*	*	*	*	*	*	9
Matsumoto (2000) <sup>22</sup>	*	*	*	*	*	*	*	*	-	8
Cao (2017) <sup>11</sup>	*	*	*	*	*	*	*	-	-	7
Shimomura (2007) <sup>13</sup>	*	*	*	*	*	*	*	*	*	9
Matsumoto (2001) <sup>14</sup>	*	*	*	*	*	+	*	*	*	9
Yoshimatsu (2001)⁵	*	*	*	*	*	-	*	*	*	8
Oshima (2012) <sup>6</sup>	*	*	*	*	*	*	*	*	*	9
Kong (2013) <sup>1</sup>	*	*	*	*	*	*	*	*	*	9
Sumi (2012) <sup>12</sup>	*	*	*	*	*	*	*	*	*	9
Kadanka (2017) <sup>15</sup>	*	*	*	*	*	*	*	*	*	9
Feng (2020)9	*	*	*	*	*	*	*	*	-	8

could decrease the quality of evidence: (1) Limitations in study design and/or execution; (2) inconsistency of results; (3) indirectness of evidence; (4) imprecision of results; and (5) publication bias. There were also three factors for upgrading. Three independent reviewers (JF, JM, and IS) assessed the quality of the evidence. We downgraded the quality of evidence by one level if one of the factors discussed was met.

The following grading of quality of evidence and definitions were used: (1) High quality: Further research is very unlikely to change our confidence in the estimate of effect; (2) Moderate quality: Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate; (3) Low quality: Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate; and (4) Very low quality: Any estimate of effect is very uncertain (Table 6).

# **RESULTS**

#### **Retrieval of studies**

Our literature search resulted in 563 articles. We excluded 26 duplicate studies. Another 537 studies were excluded because of irrelevant titles. We reviewed 23 full-text articles for eligibility. Out of 23 articles, 12 were included in the review based on inclusion criteria (Figure 1). Specifically, the study

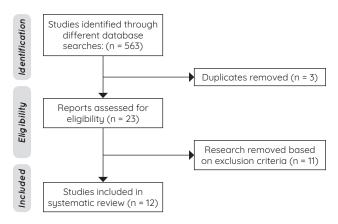


Figure 1. Flow of study selection.

of Koyanagi et al., was excluded because it involved only a surgical group of mild CMS patients.<sup>7</sup>

# Study characteristics

The included studies were published from 2000 through 2023. The major publishing countries were Japan (n=6), the Czech Republic (n=3), and China (n=3). The studies contained a total of 1,046 patients. The primary outcomes were initial and follow-up neurologic scores (mJOA, JOA, or Nurick), and clinical and imaging predictors of deterioration. The study characteristics are summarized in Table 5.

# Risk of bias assessment

Almost all studies were observational. Based on the Newcastle-Ottawa score, all were at low risk for bias. One study did not discuss the follow-up period and adequacy of follow-up (Table 4).

#### Radiculopathy, SEP, MEP, EMG and progression

In the study by Bednarik et al., involving 66 patients presenting with radiculopathy or axial pain, results from univariate logistic regression analysis indicated a significant correlation between elevated risk of myelopathy and the presence of radiculopathy (OR = 36.92, 95% CI = 4.19–325.50, p = 0.001), anterior horn cell abnormality through EMG (OR = 3.51, 95% CI = 0.89–13.87, p = 0.001), and Sensory Evoked Potential (SEP) abnormality (OR = 12.5, 95% CI = 2.97–52.41, p = 0.016). Other factors were not associated with CSM development. Male gender was noted to be significantly associated with myelopathy based on univariate analysis (OR = 4.02, 95% CI = 1.06–16.8, p = 0.038), but this was significantly associated with radiculopathy and thus was not considered as an independent factor.<sup>2</sup>

Their follow-up study (2008), involving 199 subjects presenting with radiculopathy or axial pain, showed that radiculopathy (RR = 3.68, CI = 2.03–6.69, p = 0.001), SEP (RR = 3.21, CI = 1.75–5.87, p = 0.001) and Motor Evoked Potential (MEP) abnormalities (RR = 2.91, CI = 1.60–5.29,

**Table 5.** Study characteristics and results

Author, year, type of study	Locale	Demographics	Age (years)	Follow-up	Factors predictive of neurologic deterioration	Results	Summary
Yoshimatsu, 2001 <sup>5</sup> Retrospective Observational	Japan	N = 69 Conservative = 47 Surgical = 22 Male 35 Female 34	Mean age = 67 (42-87)	Mean: 38- 39 months	Duration of symptoms     Presence of intensive conservative treatment	Multivariate analysis: improvement correlated with shorter disease duration ( $p = 0.0141$ )  Progression was associated with longer disease duration ( $p = 0.001$ )	Multivariate analysis showed significant correlations between clinical outcome, disease duration, and rigorous conservative treatment.  Conservative treatment for CSM is considered to be effective if it is performed intensively on selected patients.
Bednarik, 2004 <sup>2</sup> Prospective Observational	Czech	N = 66 Male 34 Female 32	Median age = 50 (32-75)	Minimum: 2 years	Abnormal motor evoked potential (MEP)     Abnormal EMG     Clinical signs of radiculopathy	Univariate logistic regression analysis  Radiculopathy and myelopathy (OR = 36.92, 95% CI = 4.19-325.50, $p$ = 0.001)  Anterior horn cell abnormality through EMG (OR = 3.51, 95% CI = 0.89-13.87, $p$ = 0.001)  SEP abnormality (OR = 12.5, 95% CI = 2.97-52.41, $P$ = 0.016)	Electrophysiological abnormalities together with clinical signs of cervical radiculopathy could predict clinical manifestation of preclinical spondylotic cervical cord compression.
Bednarik, 2008 <sup>a</sup> Prospective Observational	Czech	N = 199 Male 104 Female 94	Median = 51 (28-82)	Median: 44 months Minimum: 2 years	Abnormal motor evoked potential (MEP)     Abnormal EMG     MRI hyperintensity (>12 months)	Radiculopathy and Myelopathy radiculopathy (RR = 3.68, CI = 2.03-6.69, $p$ = 0.001) SEP and myelopathy (RR = 3.21, CI = 1.75-5.87, $p$ = 0.001) MEP abnormalities and myelopathy (RR=2.91, CI=1.60-5.29, $p$ = 0.001) T2 hyperintensity is associated with later (>12 months) manifestation of myelopathy ( $P$ = 0.001)	Electrophysiological abnormalities of cervical cord dysfunction together with clinical signs of cervical radiculopathy and MRI hyperintensity are useful predictors of early progression into symptomatic SCM in patients with P-SCCC.
Kadanka, 2017 <sup>15</sup> Prospective Observational	Czech	N = 112 Male 57 Female 55	Median age = 59 (40-79)	Minimum: 2 years (mean 3 years)	Radiculopathy     Electrophysiological (SEP, MEP, EMG)     Cross-sectional area     Compression ratio	Cross-sectional area (CSA) of less than or equal to $70.1 \text{ mm}^2$ (OD = $6.16$ , $95\%$ Cl = $1.61-23.72$ , $p$ = $0.008$ ) as well as compression ratio of less than or equal to $4.0$ (OD = $5.61$ , $95\%$ Cl = $1.45-21.7$ , $p$ = $0.012$ ) were independent predictors of developing myelopathy	In addition to previously described independent predictors of DCM development (radiculopathy and electrophysiological dysfunction of cervical cord), MRI parameters, namely CSA and CR, should also be considered as significant predictors for the development of DCM.
Matsumoto, 2000" Retrospective Observational	Japan	N = 52 Male: 39 Female: 13	Mean age = 55 (30-80)	Mean: 3 years	Increased Signal Intensity     Diameter of spinal canal	T2 signal hyperintensity and transverse area of the spinal cord do not correlate with poor outcome	Increased signal intensity was not related to a poor outcome of conservative treatment or severity of myelopathy in the patients with mild cervical myelopathy.
Matsumoto, 2001 <sup>14</sup> Retrospective Observational	Japan	N = 27 Male: 20 Female: 7	Mean age = 44.4 (27-69)	Mean: 3.9 years	Type of herniation Level of herniation	Although not statistically significant (p = 0.15), diffuse-type herniation spontaneously regressed more frequently than focal-type	Conservative treatment is an effective treatment option for mild cervical myelopathy caused by cervical soft disc herniation. A good outcome can be expected in patients with a median-type and/or diffuse-type herniation on magnetic resonance imaging.
Oshima. 2012 <sup>6</sup> Retrospective Observational	Japan	N = 45 Male: 27 Female: 18	Mean age = 59 (35-76)	Mean: 78 months	Cervical range of motion Segmental kyphosis Local slip	Segmental lordotic angle at the maximum segment was significantly higher in nonsurgical patients (cox HR = 4.51, CI = 1.59-12.8, $p$ = 0.001)  Local slip of >2 mm at the involved segment was also significantly higher in the surgical group (cox HR = 4.67, CI = 1.67-13.0, $p$ = 0.03)	Fifty-six percent of patients with clinically mild CSM with increased signal intensity (ISI) had not deteriorated or undergone surgery at 10 years. Large range of motion, segmental kyphosis, and instability at the narrowest canal were adverse prognostic factors.

**Table 5.** Study characteristics and results (continued)

Author, year, type of study	Locale	Demographics	Age (years)	Follow-up	Factors predictive of neurologic deterioration	Results	Summary
Cao, 2017 10 Retrospective Observational	China	N = 68 Male: 37 Female: 31	Mean age = 52.6 (38-72)	-	Torg ratio Cervical spine instability MRI T2 weighted high intensity  Terms of the content of t	myelopathy was dependently associated with cervical instability (OR 5.898, $P=0.037$ )  range of motion >50 degrees at the maximum segment involved was also noted to be significantly higher in the surgical group (cox HR: 3.25, CI = 0.42-3.17, $p=0.04$ symptomatic cases were significantly associated with T2 hyperintensity, showing a significant difference from asymptomatic cases ( $p<0.05$ )  Further analysis showed that myelopathy was associated with T2 hyperintensity (OR = 9.718, $P=0.020$ )  Torg-Pavlov Ratio was significantly associated with asymptomatic cases compared to those who present with symptoms ( $P<0.05$ )	Cervical segmental instability, a high intramedullary signal on T2-weighted MRI, and the Torg ratio had the greatest capacity to distinguish between asymptomatic and symptomatic patients with CSM with mild to moderate cervical spinal cord compression.
Shimomura, 2007 <sup>13</sup> Prospective Observational	Japan	N = 70 Male: 49 Female: 21	Mean age = 55.1 ± 11.8	Mean: 35.6 months	Circumferential spinal cord compression	associated with a decreased Torg-Pavlov ratio (OR = 0.155, <i>P</i> = 0.006)  The only noted factor associated with deterioration in mild CSM was circumferential spinal cord compression on axial MRI (OR = 26.624, CI = 1.682-421.541, <i>P</i> < 0.05)	Outcomes of mild forms of CSM during nonsurgical treatment were generally good as shown by average JOA scores. The only prognostic factor for mild forms of CSM was circumferential spinal cord compression in the maximum compression segment on axial MRI. Surgical treatment can be considered for patients with this prognostic factor.
Sumi, 2012 <sup>12</sup> Prospective Observational	Japan	N = 60 Male: 42 Female: 18	Mean age = 56.1 ± 11.8	Mean: 94.3 months	Spinal cord shape (angular)     MRI imaging findings	Ovoid deformity and T2 hyperintensity were noted to have no significant statistical relationship with myelopathy deterioration  angular deformity of the cord on T1-weighted axial scans was significantly correlated with myelopathy deterioration (OR = 8.22, P = 0.006)	The tolerance rate of mild CSM was 70% in this study, which proved that the prognosis of mild CSM without surgical treatment was relatively good. However, the tolerance rate of the cases with angular-edged deformity was 58%. Therefore, surgical treatment should be considered when mild CSM cases show angular-edged deformity on axial MR imaging, even if patients lack significant symptoms.
Kong. 2013¹ Prospective Observational	China	N = 78 Male: 45 Female: 33	Mean age = 57.8 (37-71)	Mean: 40 months	Segmental instability     Cervical spinal stenosis	segmental instability (>2 mm slippage) was significantly associated with the surgical group (P = 0.01) diameter of the CSF column was also correlated with deterioration (10.7 ± 1.8 vs. 12.1 ± 1.2 mm, P = 0.02)	Conservative treatment is effective in MCSM patients. Patients with segmental instability and cervical spinal stenosis tend to deteriorate, but conservative treatment remains the recommendation for the first action. If the myelopathy deteriorates during conservative treatment, timely surgical intervention is effective.
Feng. 2020° Prospective Observational	China	N = 200 Male: 92 Female: 108	Mean age = 55.8 ± 8.7 (28-80)	1-year follow-up	Sensory Evoke Potential (SEP) waveform	The incidence of progressive myelopathy was significantly correlated with upper SEP ( $r = 0.94$ , $\rho < 0.01$ ) and the combined SEPs ( $r = 0.95$ , $\rho < 0.01$ )	The incidence of progressive degenerative myelopathy increased with the upper and combination SEP classifications. Thus, classification of SEPs could predict the clinical decline in mJOA in CSM, reflecting the probability of worsening of myelopathy.

Table 6. GRADE certainty of evidence

Outcomes	Number of studies	Impact	Certainty of Evidence	Comments
Myelopahy progression and Radiculopathy, electrophysiologic abnormalities	3 non- randomized studies	Radiculopathy and electrophysiologic abnormalities are associated with the progression of myelopathy in two studies	⊕○○○ Very low	Purely observational studies     Two studies are similar, the latest study is the updated one
Myelopathy and segmental instability	3 non- randomized studies	Segmental instability was associated with progression of myelopathy in three studies	⊕○○○ Very low	Purely observational studies Heterogeneity exists among studies No data on follow-up in one study
Myelopathy and T2 hyperintensity	4 non- randomized studies	T2 hyperintensity was associated with myelopathy in two studies, no correlation was established in the other two studies	⊕OOO Very low	Purely observational studies     Heterogeneity exists among studies
Myelopathy and Torg-Pavlov Ratio	1 non- randomized studies	Torg ratio had the greatest capacity to distinguish between asymptomatic and symptomatic patients with CSM with mild to moderate cervical spinal cord compression	⊕OOO Very low	Purely observational studies     Lack of follow-up data
Myelopathy and T1 Cord Deformity. Circumferential Cord Compression. CSA <70 mm. Compression Ratio	4 non- randomized studies	Circumferential cord compression was associated with progression in one study Angular deformity in t1 was also associated with deterioration CSF column diameter was correlated with deterioration CSA and CR were correlated with myelopathy development	⊕○○○ Very low	Purely observational studies     Heterogeneity exists among studies
Myelopathy and Disc herniation	1 non- randomized study	Conservative treatment is an effective treatment option for mild cervical myelopathy caused by cervical soft disc herniation. A good outcome can be expected in patients with a median-type and/or diffuse-type herniation on magnetic resonance imaging	⊕○○○ Very low	Purely observational studies     The sample size is limited

**Table 7.** Sensory Evoked Potentials (SEP) waveforms by Feng et al.<sup>9</sup>

SEP waveforms					
Class I Normal amplitude and latency					
Class II with normal latency but abnormal amplitude					
Class III with normal amplitude but abnormal latency					
Class IV	Class IV abnormal latency and abnormal amplitude				
Class V	with unmeasurable or absent waveforms				

p=0.001) contributed independently to the development of myelopathy and constituted a very significant prediction model for this endpoint. Furthermore, the risk of the development of early myelopathy was seen contributed by radiculopathy (OR = 4.69, CI = 1.61–13.7, p=0.004), SEP (OR = 3.97, CI = 1.36–11.6, p=0.011) and MEP (OR = 2.94, CI = 1.04–8.75, p=0.001).<sup>8</sup>

Feng et al., conducted a study involving 200 patients, classifying 5 types of SEP waveforms (Table 7). They reported that waveforms can predict the deterioration of CSM and correlate with disease progression. According to them, the incidence of progressive myelopathy was 2.6%, 27.7%, 23.8%, 86.7%, and 100% in Class I, II, III, IV, and V of upper SEPs, respectively, and 18.8%, 39.4%, 42.3%, and 83.3% in Class I, II, III, and IV of lower SEPs, respectively. The incidence of myelopathy was 0%, 13.7%, 24.3%, 91.1%, and 100% in Class I, II, III, IV, and V respectively for the combined SEPs. The incidence of progressive myelopathy was significantly correlated with upper SEP (r = 0.94, p < 0.01) and the combined SEPs (r = 0.95, p < 0.01).

# Imaging features and progression

# Segmental instability

Oshima et al., revealed that the segmental lordotic angle at the maximum segment was significantly higher in nonsurgical patients (cox HR = 4.51, CI = 1.59–12.8, p = 0.001). Association with a local slip of more than 2 mm at the involved segment was also significantly higher in the surgical group (cox HR = 4.67, CI = 1.67–13.0, p = 0.03).<sup>6</sup> A retrospective study by Cao et al., of 68 patients found that cervical instability and a listhesis greater than 3.5 mm were noted to be associated with symptomatic cases (p < 0.05). Further analysis showed that myelopathy was dependently associated with cervical instability (OR 5.898, p = 0.037). In addition, Kong et al., noted that segmental instability (>2 mm slippage) was significantly associated with the surgical group (p = 0.01). Oshima et al., found that a range of motion >50° at the maximum segment involved was also noted to be significantly higher in the surgical group (cox HR: 3.25, CI = 0.42–3.17, p = 0.04). According to this study, segmental kyphosis, local slip, and ROM of >50° were adverse prognostic factors in mild CSM patients.<sup>6</sup>

# T2 Hyperintensity

In the study of Bednarik et al., MRI hyperintensity was associated with later (>12 months) manifestation of myelopathy (p = 0.001).<sup>8</sup> Cao et al. found that symptomatic cases were significantly associated with T2 hyperintensity, showing a significant difference from asymptomatic cases (p < 0.05). Further analysis showed that myelopathy was associated with T2 hyperintensity (OR = 9.718, p = 0.020).<sup>10</sup> On the other hand, the study of Matsumoto et al. concluded that T2 signal hyperintensity and transverse area of the spinal cord do not

correlate with poor outcomes.<sup>11</sup> Furthermore, Sumi et al. showed that T2 hyperintensity was noted to have no significant statistical relationship with myelopathy deterioration.<sup>12</sup>

# Torg-Pavlov ratio

Cao et al. studied 68 patients, finding that an increased Torg-Pavlov Ratio was significantly associated with asymptomatic cases compared to those who present with symptoms (p<0.05). However, the range of motion of the affected segment did not correlate with symptomatology. Further analysis showed that myelopathy was dependently associated with a decreased Torg-Pavlov ratio (OR = 0.155, p = 0.006). <sup>10</sup>

# T1 Cord deformity and circumferential cord compression

Shimomura et al. showed that mild CSM can be conservatively managed with good results. The only factor associated with deterioration in mild CSM was circumferential spinal cord compression on axial MRI (OR = 26.62, CI = 1.68–421.54, p<0.05). Sumi et al. investigated 55 patients with mild CSM and found that angular deformity of the cord on T1-weighted axial scans was significantly correlated with myelopathy deterioration (OR = 8.22, p = 0.006). Ovoid deformity and T2 hyperintensity were noted to have no significant statistical relationship with myelopathy deterioration. The overall tolerance rate of mild CSM is 70%, and the tolerance rate of those subgroups having angular deformity of the cord was decreased to 58%. 12

# Other findings

According to Kong et al., the diameter of the CSF column was also correlated with deterioration ( $10.7 \pm 1.8$  vs.  $12.1 \pm 1.2$  mm, p = 0.02) resulting in surgery. No significant differences were identified between the two groups in relation to age, gender, duration of disease, C2-C7 angle, MRI ISI and levels and degree of SC compression.<sup>1</sup>

Matsumoto et al. noted that diffuse-type disc herniation may be conservatively treated in patients with mild CSM. Although not statistically significant (p=0.15), diffuse-type herniation spontaneously regressed more frequently than focal type. More aggressive measures should be done in patients with a focal type of disc herniation in patients with mild CSM. <sup>14</sup>

A study by Kadanka et al. consisting of 112 subjects found that aside from radiculopathy, cross-sectional area (CSA) of less than or equal to  $70.1 \text{ mm}^2\text{ (OD} = 6.16, 95\% \text{ CI} = 1.61–23.72, <math>p = 0.008$ ) as well as compression ratio of less than or equal to 4.0 (OD = 5.61, 95% CI = 1.45–21.7, <math>p = 0.012) were independent predictors of developing myelopathy in asymptomatic patients with cervical cord compression.<sup>15</sup>

# **DISCUSSION**

The composite analysis of data derived from various studies consistently revealed that the following factors have a substantial correlation with the progression of myelopathy:

cervical radiculopathy, somatosensory evoked potentials, motor evoked potentials, EMG alterations indicating anterior horn involvement, cervical instability (slippage >2 mm or segmental kyphosis), circumferential compression of the cord, CSF column diameter, and angular deformity at T1-weighted axial scan. T2-weighted signal hyperintensity and transverse spinal cord area have been associated with poor outcomes in some studies of mild CSM patients undergoing conservative treatment. However, most studies found no correlation between MRI hyperintensity and early deterioration. Nonetheless, MRI hyperintensity was identified as a determinant of delayed development of myelopathy.

Cervical radiculopathy is a strong predictor of myelopathy development and is one of the symptomatology in patients with degenerative spine disease.<sup>8</sup> A comprehensive set of assessments, including EMG, SEP, and MEP, should be included in the initial examination for individuals with mild CSM to identify those at risk for disease progression. Electrophysiological findings reflect the long tract involved and are a strong predictor of myelopathy progression. Degenerative changes causing canal narrowing that leads to cord compression can manifest as changes in electrophysiologic reading. The study by Kanchiku et al., noted that ½ lateral cord compression was associated with long tract abnormalities.<sup>16</sup>

Segmental instability was a significant predictor of deterioration. It can lead to the progressive degeneration of the affected segments. Instability, such as vertebral slippage and segmental kyphosis, can cause an imbalance in the vector forces within each spinal unit. Abnormal soft tissue mechanics can lead to further instability and degenerative changes. The buckling of intervertebral ligaments (including the ligamentum flavum and posterior longitudinal ligament) accelerated osteophyte formation, reduction in disc space, and bulging of the disc into the spinal canal all contribute to a decrease in spinal or root canal size. <sup>17,18</sup> These events can further accelerate myelopathy development.

The Torg-Pavlov Ratio refers to the sagittal spinal canal-tovertebral body ratio, which is directly correlated with spinal canal diameter by eliminating the effect of radiographic magnification.<sup>19</sup> A smaller value indicates a smaller canal diameter and volume. In degenerative changes in the spine, small ossification of the posterior longitudinal ligament or ligamentum flavum hypertrophy can result in significant volume change, compressing the cord and further worsening myelopathy.

MRI T2 signal hyperintensity reflects parenchymal injury to the spinal cord due to compression. These signal changes can vary from reversible (edema, inflammation) to irreversible damage (gliosis, osteomalacia, necrosis) of the cord substance. According to Ramanauskas et al., there are three stages of signal hyperintensity in the cord. The early stages involve reversible edema, followed by cystic necrosis of the grey matter in the intermediate stage, and ultimately progressing into irreparable cavity and syrinx formation at later stages. The irreversible

injury occurs during the intermediate and late stages; it is challenging to distinguish between reversible and irreversible stages based solely on MRI intensity. Consequently, correlating signal hyperintensity with myelopathy progression presents difficulties.

According to Shimomura et al., the sole significant prognostic factor linked with deterioration was circumferential cord compression. The study observed that circumferential compression and severe cord distortion indicate irreversible degeneration. The CSF column diameter reflects the degree of canal stenosis since CSF displacement indicates narrowing before complete compression of the cord. Therefore, clinicians need to recognize that patients with more significant circumferential compression are at a higher risk of progression than those with only partial compression.

Angular T1 deformity correlates with adverse prognosis in patients with mild CSM. <sup>12</sup> Kemayama et al. noted that a triangular deformity was associated with more damage to the white and gray matter than an ovoid deformity; it is a critical sign of cord damage. The angular shape of the cord at the lateral recess indicates a poorer prognosis. <sup>21</sup> Another factor of interest is the CSA and Compression Ratio. These factors were useful in predicting symptomatic patients with cervical cord compression. In the study of Kadanka et al., the severity of compression on MRI could stratify patients in terms of further management. <sup>15</sup>

In patients with myelopathy secondary to disc herniation, Matsumoto et al., found that spontaneous regression was more common in patients with the diffuse type of disc herniation. In contrast, the focal type had a lesser incidence of spontaneous regression. Therefore, the latter type should be addressed aggressively.<sup>14</sup>

Most of the identified risk factors for CSM progression are poorly understood and further studies are needed to better understand and strengthen the knowledge about this condition.<sup>22</sup> Likewise, consensus is still needed on the role of surgery in patients with mild CSM. Patients may exhibit improvement, stability, or deterioration following conservative measures. Predictors can aid in identifying candidates for early surgical intervention among patients with mild CSM. Detecting such patients prone to rapid deterioration could help avert devastating neurological consequences.

#### **LIMITATIONS**

The impact of a systematic review depends on the data's quality and homogeneity. The study is limited by several factors affecting the ability to make a firm conclusion about the result. First, the study only included English articles, which may lead to an incomplete literature search, leading to publication bias. Second, the review included heterogeneous studies, and we could not conduct a meta-analysis. Thus, combining the results to make a solid basis to support the conclusion is difficult. Similar factors were discussed in different studies,

however, with conflicting results. Assessing the quality of each outcome was difficult due to inconsistencies in risk factors and comparator variables. More studies are needed to establish a strong relationship. Third, some of the studies were conducted more than ten years ago, which may affect the consistency of the results.

### **CONCLUSION**

Early recognition of clinical features such as myelopathy with radiculopathy and electrophysiologic abnormality is crucial in catching patients at risk of deterioration. Moreover, immediate identification of imaging predictors of deterioration such as segmental instability (segmental slippage > 2mm and segmental kyphosis), decreased Torg-Pavlov ratio, circumferential cord compression, CSF column diameter, and angular deformity on T1, cross-sectional area (CSA) of less than or equal to 70.1 mm<sup>2</sup> as well as compression ratio of less than or equal to 4.0 may compel us to do early surgery in these patients. The role of MRI T2 hyperintensity is still vague in predicting early deterioration, although it is associated with the development of myelopathy. Early surgery should be considered in patients with myelopathy and focal disc herniation. Diffuse-type herniation spontaneously regressed more frequently than the focal type. Early identification of these predictors can assist clinicians in determining the optimal timing for early surgical intervention.

## RECOMMENDATIONS

The findings from this study hold significant implications for the management of mild Cervical Spondylotic Myelopathy (CSM) patients. Fehling et al. outlined guidelines indicating that patients with radiculopathy and electrophysiologic abnormalities are prone to deteriorate.<sup>23</sup> By including electrophysiological tests in the initial work-up, we can identify subpopulations susceptible to early deterioration, enabling us to offer timely surgical intervention to these patients. Understanding the imaging predictors of myelopathy progression can help us anticipate patients who are prone to deteriorate and determine when early surgery should be considered. However, conflicting results exist in some predictors, and further studies are necessary. By identifying these predictors, we can significantly improve patient outcomes. In addition, this study will serve as a backbone for further studies to give us robust data on the different predictors of deterioration in mild CSM that can help in decision-making. Few data involving conservative management were collected, so it is essential to conduct further studies.

#### STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

# **AUTHORS DISCLOSURE**

The authors declared no conflict of interest.

# **FUNDING SOURCE**

None.

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