



# THE SRI LANKAN JOURNAL OF ORTHOPAEDIC SURGERY

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## President's Message SLOA 2022

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As the 14<sup>th</sup> president of Sri Lanka orthopaedic association, It's an honour and pleasure for me to send this message to SLOA Journal 2022. This year, the journal is published in parallel with the annual sessions. Even though it is the only journal published by our association, it has not reflected the full range of clinical work in Sri Lanka as the number of submissions have been low.

There are large number of clinical activities happening in many orthopaedic units in Sri Lanka. However much of the work has gone unpublished. Many cases that we see in our day-to-day practice are different with many case reports and clinical research that we see in many international publications. I feel there is some gap between the clinal activities in our community and their publications. Therefore, SLOA Journal is a good platform to fill this gap. I encourage more and more clinicians and trainees in Sri Lanka to use this platform to make their publications. Furthermore, this year SLOA Journal is being published with ISSN certification. I thank the editors for the hard work done to achieve this. move. This would enable our clinical work to be waived not only locally but internationally as well. Similarly in future international authors would find this as a valuable platform to publish their work.

I thank all the authors and the editorial board for contributing to make this journal in 2022 a success.

**Dr Dilshan Munidasa**

President,

Sri Lanka Orthopaedic Association

November 2022



## **The Journal: The past present and the future**

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Journal of the Sri Lanka orthopaedic association has come a long way since it was first published in 2006. Initially the idea that the association should have a journal enabling the trainees and surgeons to publish their work came from then president of SLOA Dr Upali Banagala who also acted as the first editor. Then the aim was to publish at least one volume per year; over the years we managed to publish 8 volumes. Initially the journal started as a private circulation journal among the membership and established a research publication identity locally

Since then, we managed to obtain an ISSN number for the journal that will help the journal to achieve national and international recognition. Another milestone was to appoint an academic editorial board that will help us to proceed smoothly. Using the journal platform, a research workshop was conducted by the Sri Lanka orthopaedic association in 2022 and will plan to conduct regular workshops every six months.

In the future journal we are planning to go online making it easy for the authors to submit articles, reviewers to review, editors to manage the contents regularly throughout the year.

Our aim is to publish a minimum of two volumes per year in the future and to achieve the valuable contributions of your research will help.

Even though We were unable to publish the journal last year due to unforeseen reasons including the pandemic this year we have a comprehensive publication including over 8 case histories and 7 research articles.

Thank you for all the contributors, authors reviewers and the editorial board.

Hoping, you enjoy reading the journal,

**Upali Banagala**

Co-Editor

**Hiran Amarasekera**

Co- Editor

SLOA Journal

November 2022



## **SUBMIT TO THE JOURNAL INSTRUCTIONS FOR AUTHORS**

*Sri Lanka Journal of Orthopaedic Surgery Established in 2008*

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Sri Lankan Journal of Orthopaedic Surgery welcomes all articles including original articles, review articles, case series, and case reports from national and international orthopaedic and research community.

All articles will be peer reviewed by an editorial board with panel of experts local and international editors and reviewers.

Articles can be directly submitted on paper format to the Sri Lanka Orthopaedic association office, Sri Lanka Orthopaedic Association, C/O The College of Surgeons of Sri Lanka, 6 Independence Avenue, Colombo 07, Sri Lanka, or email to the journalsloa@gmail.com. Once accepted all articles needed to be submitted in an electronic format.

### **Guidelines for Manuscript Submission**

Typically, original and research articles should have around 3000 to 4000 words with minimum of 10 references while much smaller articles such as case reports and case series should have between 500 to 2000 words with minimum of 3 references.

All papers should have 8 authors or less. All authors should declare an author declaration and each authors contribution towards the research should mentioned at the submission. Any additional person who is not an author but have helped in the research should be associated with the collection of data or production of the manuscript should be thanked in an acknowledgement at the end of the paper.

We encourage to submit figures and tables and any diagram that will help to understand the work and add clarity for the research. Your main document should be completely blinded and all identifying information should be on a separate title page.

We strongly recommend structuring the paper according to the standards recommended by the Bone and Joint Journal UK, (Formerly JBJS Br) and the final checklist developed by the journal similar to the check lists given by many orthopaedics journals including the BJJ.

### **Structuring the Paper**

Typically, we invite following types of articles for publication. Articles can fall in to any category below.

1. Editorial: An invited personal view on a general subject or issue.
2. Expert Opinion: This is an expert in a particular field noting and writing an opinion on a particular problem or surgical technique and sharing personal experience in their practice.

3. Instructional review article: Extensive wide-ranging contemporary updated review of an important aspect of Orthopaedic surgery.
4. Annotation: A piece concentrated on a narrow aspect of Orthopaedics.
5. Specialty update: General update of aspects concerning a subspecialty, e.g. hands, oncology, sports medicine, etc.
6. Clinical: General paper dealing with a question or hypothesis related to daily clinical practice.
7. Research: A paper primarily dealing with an area of basic science (rather than clinical practice) in a specialist research area, e.g. engineering, stem cells, pathology, genetics, biomaterials, finite element analysis, etc.
8. Case Series: Study of more than one similar cases, that are either rare or the authors would like to use as a teaching tool or share experience to deliver a novel message
9. Case history: A single case noted for rarity complexity or challenging in diagnosing or treating or an unusual complication that is worth sharing among orthopaedic community.

### **General rules for writing your paper**

All appropriate data should be presented as means with ranges and/or standard deviations (SDs). Medians should only be used when the data is skewed, accompanied by an interquartile range (IQR, presented as the upper and lower quartiles).

Present information in a consistent order throughout the text. If you are referring to Test, then Control groups, or THA patients, then TKA patients, then that order should be maintained throughout the entire text, not mixed up. Maintain this order in all tables and figures. All abbreviations should be mentioned in full at the first instance it appears on the article. E.G Total hip replacement (THR) In subsequent use only the abbreviation can be used.

If you name any specific product, then it requires the name, city and state/country of the manufacturer.

Do not describe standard procedure for common operations. Only include new procedures or adaptations to standard procedure.

### **The structure of a paper**

(Adapted Sri Lanka Journal of Surgery (Taken from the guidelines given by Bone & Joint Journal (BJJ)) Editorial board 2019 )

Papers should be divided into sections. For most papers this will be: Abstract, Introduction, Patients (or Materials) and Methods, Results and Discussion, followed by a Reference list. All data presented in the abstract must be followed-up with a relevant sentence in the paper itself. All results should be presented in the abstract.

#### **Title:**

The title of a paper should clearly define the nature of the study.



**Sub-title:**

Should only be used when qualifying information about the title is required.

**Abstract:**

The abstract should be no more than 300 words summarising the most important points in the article. It is unnecessary to include an introductory paragraph in the abstract. It should be structured to include the following headings: Aims, Patients (or Materials) and Methods, Results and Conclusion. In addition, please add one or two bullet points which sum up the clinical relevance of the paper, i. e., where it fits into the literature.

**Introduction:**

The Introduction should explain the background to the study and why the study was undertaken, explaining the problem which is to be addressed, and outlining briefly its relevance to the current literature. The last sentence should outline the research question or hypothesis. There is no need to have a separate heading named as Background.

**Patients (or Materials) and Methods:**

In the Patients and Methods section the subjects of the study and the methods and outcome measures used in the investigation must be clearly described. The selection criteria should be stated, and the number of patients included and excluded should be stated and the reasons given. Any group used as controls must be defined accurately. The Patients and Methods can be broken down into separate sections as needed: e.g. Selection criteria, Serum metal ion analysis, Histopathological examination, Study population, Comorbidity, Mortality, Outcome (e.g. what tests used and who undertook the measurements), etc.

A Statistical analysis section should be included at the end of the Patients and Methods section. This should detail which statistical tests have been used in the analysis of results, the reasons why, the statistical package used and what p-value was considered statistically significant.

**Results:**

The Results section should describe the relevant results which have been analysed and state the corresponding follow-up time, give the details of which patients made it to final follow-up and details of those that didn't and why. Sometimes follow-up information fits better in the Patients and Methods section, however, it is only needed once and should not be repeated. Tables or similar diagrams can be used but must not duplicate material already expressed, in the text. The Results can be broken down into separate sections for different analysis, e.g. Details of operation, Functional outcome scores, Radiological outcomes, etc. Any complications should be included along with the number of patients who suffered them and the relevant outcomes.

All results must be backed-up with p-values or survivorship analysis. All Kaplan-Meier data should be presented with the confidence intervals. Always present exact absolute p-values, whether significant or not, unless  $p < 0.001$ .

If needed statistical analysis can be included as a sub heading and paragraph within the results section.

### **Discussion:**

The Discussion must be succinct, pointing out the relevance of the work described in the paper and its contribution to current knowledge. The results must be interpreted clearly, and any deficiencies expressed. What can be deduced from the results and how will it affect clinical practice should be clearly stated. Discussion of pertinent references must be concise. The limitations of the study should be presented and suggest how the study could have been improved for a future study. The question or hypothesis stated at the end of the Introduction should be discussed and supported or rejected. Please do not repeat your introduction. Scope for further research in the topic and be mentioned here.

### **References:**

The references should include only those that are important and have been studied in full by the authors. They should be presented using the Vancouver system by superscript numbers in the order of their appearance. Only use references which are as up-to-date as possible, unbiased and relevant. Only use the classic, original references when needed. Otherwise try and keep references to within the last 10 years. References should only be used from published work. Personal communications are not acceptable as references. Proof of acceptance is required for references cited "in press".

The list of references at the end of the text should be formatted to the same style as these examples:

#### **Journal Reference:**

Allen GM, Wilson DJ. Ultrasound and the diagnosis of orthopaedic disorders. *Bone Joint J* 2012; 95-B:1- 50.

#### **Book Reference:**

Watson-Jones R. Fractures and joint injuries. Vol. 2. Fourth ed. Edinburgh: Churchill Livingstone, 1955:744-5.

#### **Chapter in a book:**

Winqvist RA, Frankel VH. Complications of implant use. In: Epps CH Jr, ed. Complications in orthopaedic surgery. Vol. 1. Philadelphia: JB Lippincott Company, 1978:99-129.

#### **Web Reference:**

No authors listed. International commission on radiological protection. <http://www.icrp.org> (date last accessed 20 September 2009).

#### **Abstract Reference:**

Peterson L. Osteochondritis of the knee treated with autologous chondrocyte transplantation [abstract]. ISAKOS Congress, 2001.

#### **Tables:**

Tables must not duplicate data already given in the text. They should be used to present information in a clear and concise manner. All tables should be understandable without the main text and each table should have a short, descriptive heading. They should also be created using the Table tool in Word, rather than in an un-editable format such as an imported graphic.

**Figures:**

Figures should be clear and easily understandable, with a full descriptive legend stating any areas of interest and explaining any markings, letterings or notations. For radiographs please ensure you state view used and the time point at which it was taken, as well as the demographic details of the patient if applicable. All figures should be understandable without the main text.

Please note the following points when preparing your figures:

- Please ensure that any radiographs, photographs or histology are submitted as high quality (minimum 300 dpi (pixels/inch) resolution) originals (as a tiff/JPEG). Where possible, radiographs should be supplied unmarked except where explanation is necessary, i.e. without extraneous additions such as dates.
- Check that all figure captions are present and match the figure in question – they should be provided at the end of the paper.
- Please ensure decimal points are presented as points, rather than commas.
- Graphs should be presented in an editable format (i.e. EPS, excel or power point) on a plain background, without gridlines (the background for flow charts should also be plain). Where editable versions cannot be provided, please ensure arial, or similar, font (8 point) has been used, where possible.
- Graphs with a single line should be provided in black and white, however colour can be used if this is more appropriate.
- Both x and y axes should be captioned with a description of the data presented.
- If confidence intervals have been included in the paper, please could these be added to graphs.

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In the interest of all authors, we will submit a final check list in a tick box format in a future journal. As the journal evolves, we reserve the right to change the guidelines if needed.

Hiran Amarasekera  
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November 2022



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Sri Jayewardenepura

## Article 1

# Anticoagulation in orthopaedic practice

---

### Abstract

Venous thromboembolism is a known complication following fractures and surgery. The incidence in orthopaedic surgeries is higher than in other specialities. Proper post procedural anticoagulation will help prevent most thromboembolic events. Several guidelines have been published directing the use of anticoagulation, most optimal agent to use and duration. The decision to anticoagulated must be based on a case-by-case basis considering patient factors as well. The following review article attempts to address these issues regarding anticoagulation in orthopaedic practice.

### Introduction

Deep vein thrombosis (DVT) with or without ensuing pulmonary embolism is a complication of any surgery. The risk for venous thromboembolism increases with orthopaedic surgery than with other surgical procedures with the incidence of DVT ranging up to 40% to 60% in major orthopaedic surgery. [1]

This increase can be attributed to the factors that contribute towards the pathophysiology of hypercoagulability that are seen in orthopaedic procedures. These include use of tourniquet, immobilization and bed rest leading to blood stasis, surgical manipulations of the limb that cause endothelial vascular injuries, increase in prothrombotic factors such as fibrinogen, factor V111 associated with multiple trauma, use of polymethylmethacrylate (PMMA) bone cement that increases hypercoagulability etc.

Other patient related factors that increase the risk of venous

thromboembolism (VTE) include age, obesity, varicose veins, family history of VTE, thrombophilia's, combined oral contraceptives, hormone replacement therapy, pregnancy and puerperium and underlying malignancies. These patients are at increased risk for thromboembolic events.

Randomized clinical trials have concluded that the rates of venographic documented DVT and proximal DVT, 7 to 14 days after major orthopaedic surgery in patients who did not receive any VTE prophylaxis are approximately 40% to 60% and 10% to 30%, respectively. [1]

Hypercoagulability can persist for six weeks after a hip fracture, while venous function remains significantly impaired for up to 42 days following hip fracture surgery. [2,3] VTE can occur up to three months after total knee and hip arthroplasty. [4] With routine VTE prophylaxis in orthopaedic patients symptomatic VTE within three months has been reduced to 1.3% to 10%. [4]

All this points to the very important aspect of offering postoperative thromboprophylaxis to patients with orthopaedic procedures.

### **Thromboprophylaxis**

Thromboprophylaxis can be two folds, mechanical and pharmacological.

Mechanical thromboprophylaxis includes early mobilization, graduated compression stockings (GCS), intermittent pneumatic compression device (IPCD) and venous foot pumps (VFP). Although they play a contributory role in helping to reduce thrombotic events there is no strong evidence that mechanical prophylaxis alone is adequate to prevent VTE.

Pharmacological agents used in thromboprophylaxis include Aspirin, Unfractionated Heparin,

Low Molecular Weight Heparin (LMWH) (enoxaparin, dalteparin, nadroparin, tinzaparin) Fondaparinux, Newer oral anticoagulants – (Rivaroxaban, Dabigatran, Apixaban) and Vitamin K antagonists such as Warfarin.

### **Guidelines**

Many guidelines [ACCP guideline of 2012, American Association of Orthopaedic Surgery (AAOS) guidelines (2007), SIGN guidelines (2010, updated in 2015) and NICE guidelines (2018)] have addressed thromboprophylaxis in orthopaedic surgeries and agree on most aspects of treatment options. [5,6,7,8]

#### **Aspirin**

Although Aspirin is recommended and has been used in VTE thromboprophylaxis, all guidelines suggest that in the presence of other effective agent, mostly LMW Heparins, use of Aspirin is to be discouraged.

#### **Warfarin**

Warfarin is an age-old anticoagulant which has been in use since 1954. It inhibits the gamma

carboxylation of Vitamin K dependent clotting factors 11, V11, 1X and X. Since the factors that are already formed are in the circulation of the patient and need to decay it takes about 72 hours after the initiation or change in dose of warfarin for the maximum effect to be seen. Thus a patient who is started on warfarin will have the full expected effect in 72 hours (3 days). Warfarin therapy is monitored with INR and a dose that achieves a target range of INR 2- 3 is suggested as adequate dose. The initial period up to adequate anticoagulation needs to be covered with LMWH.

#### **Unfractionated and Low Molecular Weight Heparins**

Unfractionated Heparin can be administered subcutaneously or intravenously. The subcutaneous administration requires higher doses as it is less bioavailable. UFH is recommended by the latest ACCP guidelines for VTE prophylaxis in patients undergoing THR, TKR or hip fracture surgery. [9] The therapeutic effect is monitored by APTT and the Heparin dose is titrated according to the APTT value.

Due to the necessity of monitoring by laboratory tests, both warfarin and UFH are less convenient than LMWH.

LMW Heparins are derived by enzymatic or mechanical breakdown of the UFH molecules and have the most action against anti Xa. There is no need for monitoring of treatment except in a few rare instances (renal failure, very obese, pregnancy). Monitoring is by measuring anti- Xa levels. All the guidelines and many studies have concluded that LMWH is the best drug to be used in VTE thromboprophylaxis. Renal function tests and a Full Blood Count must be done prior to commencement. Dose adjustments can be made in renal impairment while severe renal failure is a contraindication for administration. The prophylaxis dose varies for each preparation. For example a daily SC 40 mg of Enoxaparin is adequate as thromboprophylaxis.

When LMWH is used for VTE prophylaxis in patients undergoing total hip replacement, it is recommended to start either 12 hours or more



pre-operatively or 12 hours or more post-operatively. [10] Suggested duration of anticoagulant treatment by ACCP is for minimum 10 to 14 days and up to 35 days. [5]

During hospitalization, the use of dual prophylaxis with an IPCD device for at least 18 hours daily along with an antithrombotic agent is recommended. Doppler ultrasonography (DUS) screening before hospital discharge is not recommended for asymptomatic patients. [5]

NICE guidelines suggest LMWH for 10 days and then aspirin for another 28 days or LMWH for 28 days in combination with anti-embolism stockings until discharge for patients undergoing elective THR. [11] For patients undergoing elective TKR, they suggest aspirin (75 mg or 150 mg) for 14 days, or LMWH for 14 days in combination with anti-embolism stockings until discharge. [11]

### **Fondaparinux**

Fondaparinux is a synthetic pentasaccharide which also has anti -Xa activity which is higher than LMWH. It is given as a subcutaneous injection at a dose of 2.5mg daily. The latest ACCP guidelines recommend fondaparinux as a method of VTE prophylaxis for patients undergoing THR, TKR or hip fracture surgery.

### **Other oral anticoagulants**

Rivaroxaban is an orally administered direct inhibitor of activated factor X (Xa). It was found to be more effective than enoxaparin in preventing VTE after THR or TKR in phase III clinical trials. Rivaroxaban is administered in a fixed daily oral dose of 10 mg for VTE prophylaxis after elective THR and TKR. [12] The latest ACCP guidelines recommend rivaroxaban as a method of VTE prophylaxis in patients undergoing THR and TKR.

It is not recommended for hip fracture surgery as it has not been evaluated in this scenario.

Dabigatran is an orally administered antithrombin inhibitor. It is recommended for VTE prophylaxis after THR and TKR at a dose of 150 mg and 220 mg daily, starting with a half dose given soon after surgery. As with Rivaroxaban it has not been evaluated in hip fracture surgery and therefore not recommended in this event.

Apixaban is also a direct factor Xa inhibitor used in VTE prophylaxis following THR and TKR. Given at a daily dose of 2.5 mg twice daily starting 12 to 24 hours after surgery, it can be continued for 35 days for THR and 12 days for TKR. [13] Apixaban is also not recommended after hip fracture surgery.

### **Anticoagulation non THR/TKR patients**

Current ACCP guidelines suggest no VTE prophylaxis rather than prophylaxis for patients undergoing knee arthroscopy. However, in the presence of other risk factors for DVT such as a history of VTE, malignancy etc. thromboprophylaxis may be initiated.

The incidence of DVT with short leg cast immobilization is in the range of 4% to > 16%. [14,15] Therefore, current ACCP guidelines suggest no VTE prophylaxis in patients with isolated lower-leg injuries requiring leg immobilization.

For patients undergoing spine surgery who do not have additional VTE risk factors, thromboprophylaxis is not necessary. However, on a patient-to-patient basis if additional risk factors are present thromboprophylaxis with UFH or LMWH can be offered.

In spinal cord injury, if acute bleeding is suspected mechanical thromboprophylaxis is the option. With time if no bleeding or haematoma is ascertained by MRI thromboprophylaxis can be initiated.

Upper limb surgeries do not require thromboprophylaxis unless prolonged local anaesthesia has been used.

## Conclusion

The risk of bleeding is present with all anticoagulants specially when over anticoagulated. It is imperative that proper doses be administered. Monitoring must be done with laboratory tests (for UFH, Warfarin ) with proper titration of doses .

Renal function tests, liver function tests, FBC and coagulation screen must be done prior to initiation of treatment.

VTE associated with orthopaedic procedures is preventable and all measures need to be taken to prevent it in the at risk patient.

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## Article 2

# Posterolateral rotatory instability of the elbow

### Abstract

Posterolateral rotatory instability (PLRI) of the elbow is the most common chronic instability pattern identified. It is the resultant ulno-humeral instability secondary to compromised lateral ligament complex. The characteristic injury is the avulsion of the lateral ulnar collateral ligament (LUCL) from its humeral attachment. Acute PLRI can present following simple or complex elbow dislocations. Chronic PLRI mostly follows trauma but also can be the result of iatrogenic injury.

Common presentations include pain, locking, pseudo locking, loss of motion and apprehension for certain movements. Stress testing that demonstrates the posterior dislocation or subluxation of the radial head plays a vital role in the diagnosis of PLRI. Diagnosis is supported by static and dynamic imaging. Examination under anaesthesia and arthroscopy enables objective diagnosis of PLRI.

Surgical repair of PLRI is often successful following acute presentations and less symptomatic chronic presentations where there is a healthy ligament remnant is available. Many surgical reconstruction techniques for chronic PLRI have been described, with good clinical and patient reported outcomes. Most common complication following PLRI reconstruction is recurrent instability which mostly occur in revision procedures.

### Introduction

Elbow instability clinically presents as inadequate joint congruity or tendency to dislocate

on examination. Acute dislocations occur most commonly during athletic activities<sup>1</sup>. After an acute dislocation ongoing instability persists in 0-8% of the patients<sup>2,3</sup>. Chronic elbow instability, 90% of the time, is the result of non-healing soft tissue injuries or bony defects following acute trauma<sup>4</sup>. Repetitive stress, congenital or acquired deformities, collagen disease, inflammatory arthritis and iatrogenic injuries due to steroid injections or prior surgery account for the remaining 10%<sup>5</sup>.

Posterolateral rotatory instability (PLRI) is the most common form of chronic instability.

Initially described by O'Driscoll et al, PLRI refers to a syndrome of ulno-humeral instability secondary to injury to lateral collateral ligament complex (LCLC)<sup>6</sup>. The resultant posterior subluxation or dislocation of the radial head relative to the capitellum gives rise to the clinical symptoms of PLRI. While PLRI can occur following simple elbow dislocations, the most common complex dislocation pattern that results in PLRI is the terrible triad injury. In terrible triad the elbow is dislocated with coexisting

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radial head and coronoid fractures<sup>5</sup>. When the ulnar humeral joint externally rotates the lateral ligament complex fails in tension<sup>7</sup>.

Most common cause of PLRI, in systematic review of 168 cases, was trauma accounting for 87%. Two thirds of this of this were elbow dislocations<sup>8</sup>. Interestingly 7% patients had PLRI following iatrogenic causes like surgical release and repeated steroid injections for lateral epicondylitis. Injury to the lateral collateral ligament (LCL) of the elbow is identified as ubiquitous lesion of PLRI<sup>3</sup>. It is typically avulsed at its humeral origin. Untreated PLRI can lead to ongoing symptoms and finally degenerative arthritis of the elbow joint.

### **Surgical anatomy**

Elbow is stabilised by is both static (osseous and soft tissue) and dynamic mechanisms<sup>9</sup>.

Static mechanism includes the osseous congruity of the ulna-humeral joint and the medial and lateral ligament complexes. The elbow flexion angle affects the stability provided by these osseous and soft tissue structures<sup>9</sup>. Bony stability is maximum at  $<20^\circ$  and  $>120^\circ$  of flexion<sup>10</sup>. Soft tissue structures like static medial and lateral collateral ligament complexes are dynamically supported by muscular stabilizers; common flexor and extensor origins, the biceps brachii and triceps brachii. The anterior bundle of the medial collateral ligament (aMCL) is the primary stabiliser against valgus strain in at  $30^\circ$ - $110^\circ$  of flexion. Anterior bundle is mainly active during extension and early flexion whereas the posterior bundle becomes the principal stabilizer from  $60^\circ$  to full flexion<sup>9</sup>.

The lateral ligament complex resists excessive varus and external rotation forces on the elbow<sup>11</sup>. The lateral ligament complex consists of, annular ligament, lateral/ radial collateral ligament and lateral ulnar collateral ligament (LUCL). LUCL origins at the lateral humeral epicondyle, partly blends with the annular ligament and inserts at the supinator crest of the ulnar. About 30% of the population has an

accessory lateral collateral ligament extending from the annular ligament to the supinator crest of the ulna<sup>11</sup>.

### **Applied biomechanics**

The common injury pattern leading to PLRI is fall onto outstretched hand with weight of the body transmitting through an axis lateral to the elbow joint<sup>12</sup>. This creates a valgus strain on the elbow with concomitant axial load. Then a supination (internal rotation) moment is applied to the elbow, as the body rotates externally pivoting around the hand. This leads to avulsion of the LCLC off its proximal attachment at the lateral humeral epicondyle.

O'Driscoll et al demonstrated simple elbow dislocations can be produced by sequential ligament failure from lateral to medial side, a concept termed "Horii circle"<sup>13</sup>. Schreiber reviewing 62 youtube.com video footages of elbow dislocation concluded that, the elbow dislocates in a position of relative extension<sup>14</sup>. He stated that sequence of disruption occurs from medial to lateral, disrupting the anterior bundle of medial collateral ligament (aMCL) first. Evidence of MRI from further studies by Schreiber et al and Rhyou et al confirms this concept, the first study showing significant partial or complete medial ligament tears in all MRI scans following simple dislocations<sup>15,16</sup>. Therefore, it is important to identify PLRI as a stage in the spectrum of elbow instability and hence look for co-existing other instability patterns.

Classification of elbow instability remains complex. Understanding a mechanistic classification of instability is important as diagnosis of instability relies upon direct appreciation of dynamic joint incongruity on stress testing. The common mechanistic patterns of elbow instability can be rotatory (PLRI and Posteromedial rotatory instability-PMRI), Axial (Monteggia and olecranon fracture dislocations), Longitudinal (Essex-Lopresti lesion) and combined patterns. While PLRI is the most common pattern, valgus instability is the second commonest. PLRI is a consequence of trauma

in 90%, however, valgus instability results from chronic repetitive valgus strains as in pitching and overhead racket sports.

## Diagnosis

Diagnosing PLRI is often challenging, mostly in chronic presentations. A clear history, a detailed clinical examination and utilising advanced imaging such as MRI is important in making a diagnosis. Acute PLRI can present with simple or complex elbow dislocation. Chronic patients commonly present with pseudo locking, loss of motion, apprehension, or pain with certain activities. History should aim to elicit risk factors for instability such as previous trauma, surgery or corticosteroid injections most often administered for tennis elbow.

While it is important to look for signs of generalised ligamentous laxity, focussed elbow examination for PLRI is essential. Assessment of the joint alignment, the presence of previous scars is important. Stress testing or provocative manoeuvres to reproduce instability are useful.

During stress testing the direction to which the elbow has the propensity to dislocate, during physiological range of motion, is identified. The direction of instability will guide to the classification and the structures that needs to be repaired. Direction of elbow instability determines the basis of mechanistic classification.

The objective of the clinical tests for PLRI are to demonstrate ulno-humeral instability indirectly by observing the posterolateral translation of radial head, while applying axial force with valgus strain with forearm in supination, taking it through the range of motion from flexion to extension. Observing features of instability like subluxation of radial head, apprehension and a palpable clunk while recreating the forces that disturb joint congruity enable diagnosis. The stress tests include, table-top relocation test, chair push up test and press up test<sup>17,18</sup>. Subluxation, apprehension frank dislocation or pain leading to inability to fully extend is described as positive tests.

Pivot shift test as described by O Driscoll is positive when the radial head subluxates when the elbow is taken through range of motion from extension to flexion while applying an axial load, a supination torque, and a valgus strain. During 20°-40°-degree flexion arc, the supination torque reaches a maximum and the valgus strain is the only lateral strain remaining when the elbow shows maximum instability. The subluxated radial head causes a skin dimple at this point. With further flexion the elbow reduces with a clunk. Pain and apprehension are also considered a positive test. This needs clinical experience to demonstrate, while can be uncomfortable in an awake patient. Also, the sensitivity of the test is less but the specificity is good.

Posterolateral drawers test is considered a better clinical test to diagnose PLRI due to its increased sensitivity and specificity<sup>19</sup>. Antero posterior translation of the forearm on a stable humerus is done while observing for skin dimpling over the radial head as it dislocates and reduces. Pain and apprehension are also considered positive signs. Eliciting subtle forms of instability clinically can be challenging and findings can be equivocal in milder forms where ligament reconstruction is helpful. Performing these tests in an acute painful elbow is often uncomfortable and can lead to misinterpretation of findings. Examination under anaesthesia should be considered in cases where there is strong clinical and radiological suspicion.

Examinations is not complete unless both collateral ligaments are tested. Under valgus strain 15°-30° flexion is used to check the MCL. It is important understand that PLRI is one presentation of elbow instability which may well co-exist with the other forms. Examination for the integrity of the nerves, specifically the ulnar nerve is mandatory.

Standard static X ray imaging gives minimal details on instability which is a dynamic phenomenon. Standard anteroposterior and lateral X rays may reveal presence of loose bodies, arthritis, coronoid dysplasia, and malalignments occurred because of a paediatric fracture. Also following an acute dislocation, this may reveal, angulations in the radial neck, rim defects in

the radial head and impaction fractures of the capitellum. 3D CT scans enable diagnosis of osseous factors of elbow in stability like coronoid fractures, radial head fractures and associated bone loss of the capitellum as in a 'Hill-Sachs' lesion<sup>5</sup>. MRI Can be helpful in assessing the extent chondral injuries, ligament tears, chondral injuries, and joint subluxations. Dynamic imaging like ultra-sonography and fluoroscopy can demonstrate radial head subluxation or ulno-humeral widening. Ulno-humeral laxity more than 4 mm is indicative of PLRI.

However, intra operative arthroscopic diagnosis is considered the most sensitive and current gold standard with regard to pathologies in other joints like wrist<sup>20</sup>. This is due to direct appreciation of disrupted structure by the operating surgeon and objective demonstration of the instability pattern<sup>21</sup>. Also, dynamic nature of diagnostic arthroscopy allows the surgeons to assess in real time, the surgical need for repair or reconstruction, assessing the disrupted anatomy and gauging the degree of instability. Arthroscopic diagnosis and classification also help to diminish the effects of confounding factors of clinical and imaging-based diagnosis like discomfort felt by the patient and having no visual impact as in pivot shift test, where the examiner feels a clunk. Arthroscopy enables intra operative decision making and guides the treatment<sup>22, 21</sup>.

## Management

Management of lateral instability can be challenging. A management algorithm is often helpful in complex situations<sup>23</sup>.

Simple acute elbow dislocation rarely needs surgery, the two indications being, irreducible dislocations and the inability to maintain reduction. These occur less than 10% in cases of acute simple elbow dislocations. Over 90% of the simple dislocations are managed with reduction and splinting. Pronation increases stability of the elbow if lateral instability is the aetiology<sup>24</sup>. In isolated medial instability the patient should be splinted in supination<sup>25</sup>.

However, if both lateral and medial ligament complexes are compromised immobilization in the neutral position is recommended. At least for 4 weeks unprotected shoulder abduction is not recommended. This is to prevent varus strain on the healing lateral ligament complex.

Some authors recommend examination under anaesthesia for simple dislocations if the mechanism of injury is of high energy, severe swelling and bruising all around the elbow, and if the patients are reluctant for active mobilisation after 1-2 weeks of non-operative management<sup>5</sup>. This is due to the extent of soft tissue injury being more dramatic where the whole distal humerus can be stripped off the soft tissue.

Ligament repair is recommended in acute stage for elbows that are unstable  $<30^\circ$ . Repair can also be attempted for early, less symptomatic chronic PLRI if soft tissues are favourable. For open surgery patient is positioned supine with the affected arm on a hand table. Use of sterile tourniquet is helpful for adequate access during the procedure. Kocher approach is used for isolated lateral access, but posterior incision is an option in the presence of co-existing medial instability. In acute elbow dislocations it is common to find a haematoma with a torn anterior capsule and brachialis muscle. Once the hematoma is cleared laxity of the lateral ligament complex and annular ligament can be observed. It is not uncommon to find the entire lateral ligament complex avulsed from the humeral origin and flipped into the radio-capitellar joint.

Once the avulsed proximal end is freed up, the footprint on the posterior aspect of the lateral humeral epicondyle can be identified. This is seen as a bare area directly lateral and slightly inferior to the centre of the olecranon fossa. After debriding the footprint, a trans osseous repair can be done with No 1 or 2 non-absorbable braided sutures. Sutures are tied at elbow flexion of  $30^\circ$ . Suture anchor repair is another option. Cast or brace is applied for initial post operative period which can be replaced by a removal splint/ brace at 10-14 days. The elbow is splinted at rest for 4 weeks, strictly avoiding shoulder abduction.

Symptomatic chronic PLRI rarely responds to non-operative management. Therefore, non-operative management is limited to very low demand patients. Lateral ligament complex reconstruction is considered in this situation<sup>26,27</sup>. Palmaris longus, semitendinosus, allograft or synthetic graft options can be used. Open surgery is performed through the Kocher's approach. A variety of bone tunnel configurations have been described which includes direct pass-through tunnels at the humeral or ulnar ends, convergent tunnels, or half tunnels to dock the ligaments. Most commonly reported technique is the docking technique<sup>27</sup>. Trans osseous repair with graft sutured onto itself, suture anchors or interference screws can be used for fixation.

Important technical points are identifying the isometric point on the humeral epicondyle for better stability. Graft tensioning during fixation is another crucial step with the final graft tensioning performed with the elbow positioned in 30° to 40° of flexion and pronation. Capsular closure is important to retain the graft at an extra-articular location.

Rehabilitation protocol is similar to acute repair.

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## Outcome

There is no difference reported between the described techniques or choice of graft regarding surgical outcome<sup>8</sup>. Most common complication was recurrent instability and 15% patients do have recurrent instability and majority of them were following revision procedures. Over 93% of the patients would achieve normal or near normal functional range post operatively. With a mean follow up of 34 months, nearly half the patients did have ongoing pain, however 87% was satisfied following PLRI reconstruction.

## Summary

PLRI commonly follows trauma, but it can be the result of iatrogenic causes like previous lateral epicondylitis release or multiple steroid injections. Diagnosis is by a detailed history, examination using stress tests and a combination of imaging including 3D CT and MRI. It is important to look for other instability patterns as PLRI is one stage in the spectrum of elbow instability. Arthroscopy is a useful adjunct in the diagnosis of PLRI and is also effectively used for surgical repair. In chronic symptomatic PLRI surgical repair results in good clinical and patient reported outcomes.

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### Article 3

## Role of Growth Modulation in the Management of Idiopathic Scoliosis: A Narrative Review of literature

**Key words:** Idiopathic Scoliosis, Growth Modulation, Spinal Deformity, Lateral Bending

### Abstract

Scoliosis is a condition in which there is a lateral curve to the spine. The cause of scoliosis is unknown, but different aetiologies have been suggested, but mostly cause has been idiopathic in nature. Muscle spasms, cerebral palsy, Marfan syndrome, and tumours (neurofibromatosis) are some of the suggested aetiologies. Progressive adolescent idiopathic scoliosis (AIS) affects mainly girls and anterior spinal overgrowth during the second growth spurt appear to be a significant cause.<sup>1</sup> The diagnosis of scoliosis is primarily a clinical suspicion followed by an AP and lateral X ray of spine in standing position. Measurement of the Cobb angle is the standard method of assessing the curvature in a quantitative manner. It is the angle between two lines, drawn perpendicular to the upper endplate of the first vertebra involved and the lower endplate of the last vertebra involved. Cobb angles are followed for both curves for those with two curves. Risser classification grades the skeletal maturity on X rays and is considered along with the Cobb angle to making a decision towards surgery.<sup>2</sup> Indications for surgical correction of scoliosis is not clearly defined however, the treatment of scoliosis is based on the severity and progressive nature of the curve. Cobb angle above 40 degrees, rapidly progressing curve, major thoracic deformity affecting the function such as breathing and cosmetic reasons, are some of the indications for surgery. There are several modalities of surgical correction. Main method has been correction of the deformity and fixation with one or two rods and pedicle screws followed by bone grafting. Harrington rods were the primarily used system, but with time many spinal instrumentation systems and rods have been introduced to the market. <sup>3</sup> Apart from using rods some have tried anterior vertebral body stapling and anterior vertebral body tethering for correction scoliosis using the principle of growth modulation. <sup>4 5</sup>

We conducted a systematic review to answer the question, 'What is the role of growth modulation in the management of idiopathic scoliosis?' All articles / publications published from 2010 May to 2019 October were used as research data. A health, related database (PubMed) was used for the search. Search criteria revealed 99 articles initially and 37 articles after filtered based on title reading. Then 18 articles were selected for the final analysis after being filtered manually by reading summary and abstracts of each article.

**Level of Evidence:** Level II, systematic review

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## Introduction

A condition in which there is a lateral curvature of the spine is scoliosis. The cause of most cases is unknown, but it is said that genetics and environmental factors play a role in it. The treatment is based on the severity and progressive nature of scoliosis. Based on that there are several modalities of treatment which includes watching periodically, applying a brace and surgical correction. The surgical correction of scoliosis involves many treatment formats out of which growth modulation is considered one. The growth modulation in managing Adult idiopathic scoliosis (AIS) is based on Hunter-Volkman law. This law tries to explain the mechanism of scoliosis. A compressive force usually halts the growth while traction force enhances it. In the vertebral column there is anteriorly a compression and posteriorly traction force. This is a possible reason for normal AP curvatures in scoliosis and a lateral bending that is significant. However, due to rotation of the vertebra, the direction of the traction and compression forces change creating disproportionate growth of vertebrae.

Hunter Volkman law-based growth modulatory surgical techniques include use of correctional implants such as Harrington rods, anterior vertebral body stapling and vertebral body tethering.

Harrington rods are used in the correction of curvatures in scoliosis and fixation of vertebra. They can be fixed with single or double rods. It's a stainless-steel rod fitted with hooks at both ends and a ratchet and is implanted through a posterior approach. Harrington rods reduce the curvature and provide more stability to a spinal fusion. Before the Harrington rod was invented, scoliosis patients had their spines fused without any instrumentation to support it; However, disadvantages of such fusions were required many months in plaster casts, and large curvatures progressed despite fusion.

Anterior vertebral body stapling is a new non fusion technique used to treat scoliosis, especially in skeletally immature patients. This concept of stapling the growth plates in achieving curve stabilization via growth modulation is known to be an effective method. Therefore, it is used as an alternative to bracing in patients with rapid progression of the curve and for patients who might need spinal fusion in the future.

Screws placed into the vertebral body modulating the growth of concave and convex sides of the spine and curve stabilization is the technique used in anterior vertebral body tethering. It is a newer method compared to stapling and it places a compressive force over the convex side of the spine with slowing growth and permits growth of the concave side ultimately creating a straight spine.

The basic principle in spinal growth modulation involves slowing the growth on the convex side and enhancing the growth on the concave side, resulting in gradual deformity correction.<sup>5</sup> In theory advantages include early recovery ability to use minimal invasive procedures, and motion preservation. This systematic review attempts to look at the published evidence of available growth modulation techniques and instrumentations.

## Materials and methods

The objective of the study was to find out the Role of growth modulation in the management of Idiopathic Scoliosis. A detailed systematic search of the database (PubMed) was done.

### Search strategy:

The following search strategy was carried out using PubMed database. The initial PubMed search was done using the terms "Growth modulation" OR "Scoliosis" which retrieved 145114 articles. Then a filter up to 10years (2010-2019) was added to the search and it retrieved 72972 articles. When a single term "Scoliosis"

was searched the result was 10716 hits. “Growth modulation” search of the database from 2010 to 2019 gave 62355 hits. Further search continued with the terms “Growth modulation” AND “Scoliosis” with the aim of filtering the number of articles for final analysis retrieved 158 results. Further filtering the search for 10 years (2010-2019) with terms “Growth modulation” AND “Scoliosis” retrieved 99 articles.

Search: **(growth modulation) AND (scoliosis)** Filters: **from 2010 - 2019**

(((((("growth and development"[MeSH Subheading] OR ("growth"[All Fields] AND "development"[All Fields])) OR "growth and development"[All Fields]) OR "growth"[All Fields]) OR "growth"[MeSH Terms]) OR "growths"[AllFields])AND((((("modulate"[All Fields] OR "modulated"[All Fields]) OR "modulates"[All Fields]) OR "modulating"[All Fields]) OR "modulation"[All Fields]) OR "modulations"[All Fields]) OR "modulator"[All Fields]) OR "modulators"[All Fields])) AND (("scoliosis"[MeSH Terms] OR "scoliosis"[All Fields]) OR "scolioses"[All Fields])

9900:22:23#12

Search: **(growth modulation) AND (scoliosis)**

(((((("growth and development"[MeSH Subheading] OR ("growth"[All Fields] AND "development"[All Fields])) OR "growth and development"[All Fields]) OR "growth"[All Fields]) OR "growth"[MeSH Terms]) OR "growths"[AllFields])AND((((("modulate"[All Fields] OR "modulated"[All Fields]) OR "modulates"[All Fields]) OR "modulating"[All Fields]) OR "modulation"[All Fields]) OR "modulations"[All Fields]) OR "modulator"[All Fields]) OR "modulators"[All Fields])) AND (("scoliosis"[MeSH Terms] OR "scoliosis"[All Fields]) OR "scolioses"[All Fields])

15800:22:02#11

This was used as the base for our search strategy. After going through the titles of the articles We manually removed 62 articles. (24 Non-human studies, 30 Studies not related to the topic, 8 duplicate studies) .Remaining 37 articles were retrieved for abstract reading. Using our exclusion criteria, we further reduced to 18 articles.19 articles excluded after abstract reading as per exclusion criteria. (2 studies in other languages, 2 animal studies, 11 studies which were not topic related, 4 studies, including a pilot study, observational study, cohort study and a single surgeon study) (Fig 1) in Using data extraction form which included Title, type of study, research question, publisher/journal, impact factor of the journal, authors, patients, comparison and outcome we retrieved following information from all the final 18 articles. (Table 1)

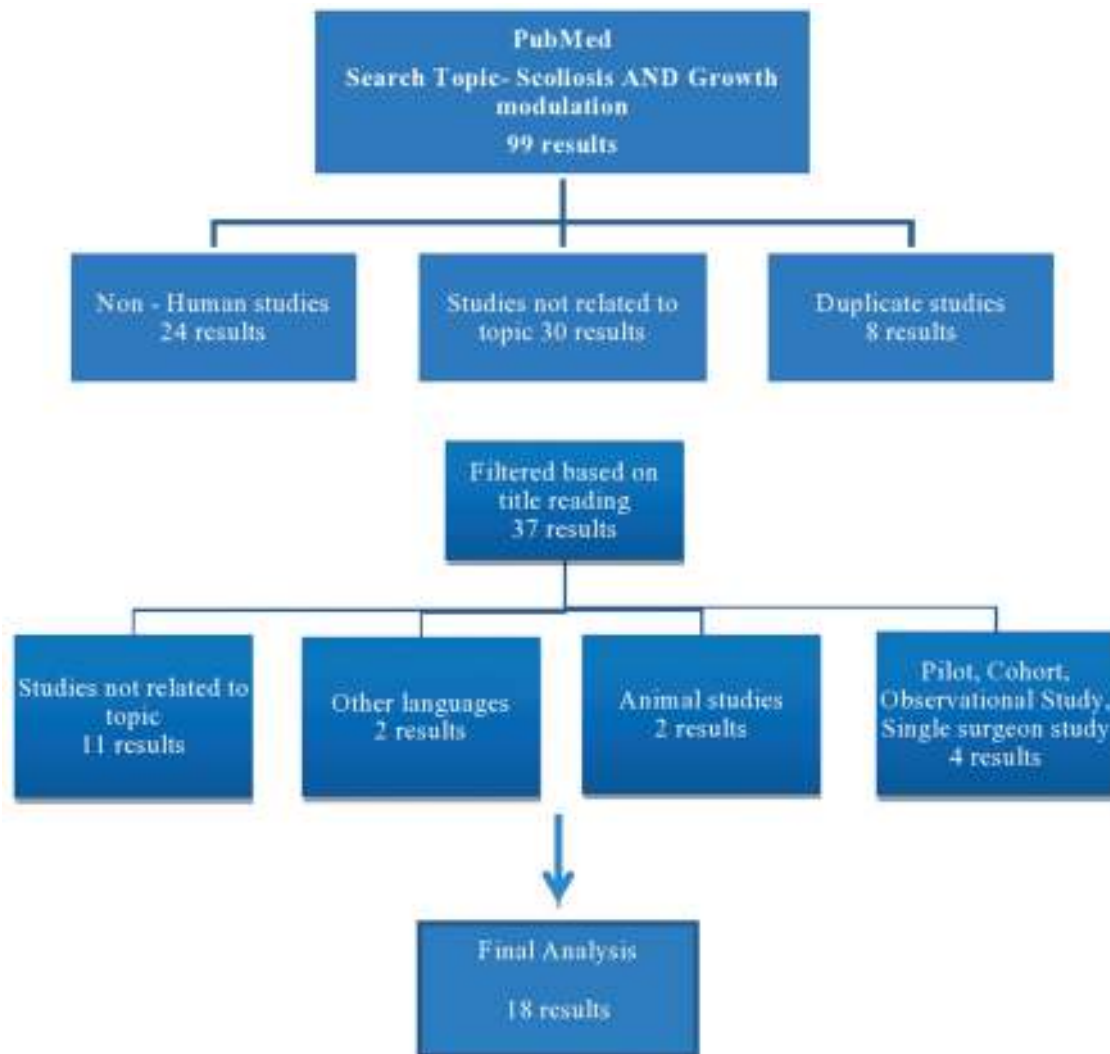
The inclusion and exclusion criteria which were used in the final selection of 18 articles are given below. (Fig 1)

#### **Inclusion criteria**

- All articles published from 2010 May to 2019 October in the search site PubMed
- Articles published in English language
- Articles that were related to the research question

#### **Exclusion criteria**

- All articles that were published in other languages
- All articles that were published before 2010 May and after October 2019
- Studies not involving humans
- Articles that were retrieved by the data base search, but were manually filtered reading the titles as they were not topic related
- Pilot studies, case series, cohort, observational studies and single surgeon studies were manually filtered
- Duplicate articles were removed manually.



**Fig 1** Flow chart of the search strategy-

## Results

Summary of final 18 articles are given below in table 1.

**Table 1** Summary of final 18 articles analysed

Title	Type of study	Publisher/Journal (Impact Factor)	Authors	Patients	Intervention	Comparison	Outcome Primary Secondary
1. Idiopathic scoliosis in children and adolescents: emerging techniques in surgical treatment <sup>7</sup>	Narrative review	World Neurosurgery (1.723)	<i>Cheung et al<sup>7</sup></i>	Children and adolescents with idiopathic scoliosis	Emerging techniques in surgical treatment	None	Robotic-assisted pedicle screw placement, vertebral body stapling, vertebral body tethering, magnetically controlled growing rods, , and sublaminar polyester bands are the emerging techniques in surgical treatment.
2. Dynamic scoliosis correction as alternative treatment for patients with adolescent idiopathic scoliosis: A non-fusion surgical technique <sup>8</sup>	Narrative review	Zeitschrift für Orthopädie Und Unfallchirurgie (0.572)	<i>Trobisch et al<sup>8</sup></i>	100 patients from different institutions	Dynamic scoliosis correction: A non-fusion surgical technique	None	Anterior dynamic scoliosis correction has promising short term results but there is a paucity of literature and optimal criteria for the best candidate has yet to be defined.

Title	Type of study	Publisher/Journal (Impact Factor)	Authors	Patients	Intervention	Comparison	Outcome Primary Secondary
3. Curve modulation and apex migration using Shilla growth guidance rods for early-onset scoliosis at 5-year follow-up <sup>9</sup>	Retrospective study	Journal of paediatric orthopaedics (2.046)	<i>Wilkinson et al</i> <sup>9</sup>	All patients with Shilla implants in place for ≥5 years yielded 21 patients	Curve modulation and apex migration using Shilla growth guidance rods	Coronal curve characteristics preoperatively, postoperatively, and at last follow-up to note changes in the apex of the primary curve.	Apex of the fused primary curve shifts in approximately 62% of patients, with nearly all of these (92%) involving a distal migration. Compensatory curves did develop after Shilla placement as well.
4. Prediction outcomes for anterior vertebral body growth modulation surgery from discriminant spatiotemporal manifolds	Narrative review	International Journal of computer assisted radiology and surgery (1.961)	<i>Mandel et al</i> <sup>10</sup>	Adolescents with idiopathic scoliosis	Anterior Vertebral Body Growth modulation	None	Achieved a higher prediction accuracy and improved the modeling of spatiotemporal morphological changes in surgical patients treated with AVBGM.
5. Anterior spinal growth tethering for skeletally immature patients with Scoliosis: A retrospective look two to four years postoperatively	Retrospective study	The journal of bone and joint surgery (4.84)	<i>Newton et al</i> <sup>11</sup>	Skeletally immature patients with thoracic scoliosis who underwent ASGT	ASGT with a minimum of 2 years of follow-up.	Skeletally immature patients with thoracic scoliosis	ASGT showed a powerful, but variable, ability to modulate spinal growth and did so with little perioperative and early postoperative risk
6. Scoliosis vertebral growth plate histomorphometry: Comparisons to controls, growth Rates, and compressive stresses	Retrospective study	Journal of Orthopaedic Research (3.14)	<i>Bylski-Austrow et al</i> <sup>12</sup>	Patients with Severe scoliosis	Hypertrophic zone heights and chondrocyte heights have been used to assess treatments that aim to modulate growth	To age matched autopsy specimens	Help assess theories of progression and potential treatments using growth modulation.
7. Thoracoscopic anterior instrumentation and fusion as a treatment for adolescent idiopathic scoliosis: A systematic review of the literature	Systematic review	Spine deformity (1.11)	<i>Padhye et al</i> <sup>13</sup>	AIS patients	Thoracoscopic anterior instrumentation and fusion	None	Advantages include less invasive, excellent curve correction, few levels fused, good satisfaction, and no long-term effect on pulmonary function. Drawbacks are, increased operative time and incidence of pulmonary complications.
8. Magnetic growth modulation in orthopaedic and spine surgery	Retrospective study	Journal of Orthopedics (1.463)	<i>Eltorai et al</i> <sup>14</sup>	Patients with the magnetically controlled growing rod system (MCGR)	The magnetically controlled growing rod system (MCGR) relative to traditional growing rod system (TGR)	To traditionally growing rod systems	MCGR is promising in that it involves less surgical procedures, shorter hospital stays, and lower long-term cost relative to TGR
9. What's new in paediatric spine growth modulation and implant technology for early-onset Scoliosis?	Systematic Review	Journal of paediatric orthopaedics (2.046)	<i>Wessell et al</i> <sup>15</sup>	Pædiatric age group with early onset scoliosis	Spine growth modulation	None	Summarizes the recently published literature regarding growth-friendly spinal implants, the status of their Food and Drug Administration approval labelling as well as the indications, applications, and complications associated with their implementation.

Title	Type of study	Publisher/Journal (Impact Factor)	Authors	Patients	Intervention	Comparison	Outcome Primary Secondary
10. 3D correction over 2years with anterior vertebral body growth modulation: A finite element analysis of screw positioning, cable tensioning and postoperative functional activities	Retrospective study	Clinical Biomechanics (2.248)	<i>Cobetto et al</i> <sup>16</sup>	Scoliosis cases	Anterior vertebral body growth modulation	Cable tensioning and screw positioning	Biomechanical possibility to adjust the fusionless instrumentation parameters to improve correction in frontal and sagittal planes, but not in the transverse plane. The convex side stresses increase in the supine position may suggest that growth modulation could be accentuated during nighttime.
11. Biomechanical simulations of costo-vertebral and anterior vertebral body tethers for the fusionless treatment of paediatric Scoliosis	Retrospective study	Journal of orthopaedic research (3.14)	<i>Aubin et al</i> <sup>17</sup>	Adolescent scoliosis patients	Fusionless treatment	CV and ANT	Biomechanical study captured the differences between a CV and ANT tether and indicated the variability arising from the patient-specific characteristics.
12. Growth tethering devices for idiopathic Scoliosis	Narrative review	Expert review of medical devices (1.784)	<i>Courvoisier et al</i> <sup>18</sup>	Idiopathic scoliosis	Growth tethering	None	This review discusses the recent developments in the field of spinal growth modulation techniques and discusses the pros and cons of the medical devices used in this indication.
13. Anterior vertebral body tethering for idiopathic Scoliosis: Two-year results	Retrospective review.	Spine (2.078)	<i>Samdani et al</i> <sup>19</sup>	Patients who underwent anterior VBT with 2-year follow-up	Anterior vertebral body tethering	preoperative, intraoperative, and most recent clinical and radiographical data	Anterior VBT is a promising technique for skeletally immature patients with idiopathic scoliosis. This technique can be performed safely and can result in progressive correction.
14. Surgical aspects of spinal growth modulation in Scoliosis correction	Narrative review	Instructional course lectures (0.6)	<i>Jain et al</i> <sup>5</sup>	Scoliosis	Spinal growth modulation		Surgical aspects of growth modulation described
15. Early onset Scoliosis: Modern treatment and results	Systematic review	Journal of paediatric orthopaedics (2.046)	<i>Tis et al</i> <sup>20</sup>	Early onset scoliosis	Modern treatment methods of scoliosis	None	Recent advances have improved the treatment of children with EOS. Treatment continues to be challenging with complication rates higher than treatment of idiopathic scoliosis.
16. Biomechanical comparison of fusion less growth modulation corrective techniques in paediatric Scoliosis	Comparative study	Medical& biological engineering and computing (1.82)	<i>Driscoll et al</i> <sup>21</sup>	Adolescent idiopathic scoliosis	Fusionless growth modulation corrective techniques	Non-instrumented and instrumented models	Initial implant compression achieved during instrumentation provided a significant influence on initial and long-term spinal profiles. The developed FEM provides an effective platform with which to explore, critique, and enhance fusionless growth-sparing techniques.

Title	Type of study	Publisher/Journal (Impact Factor)	Authors	Patients	Intervention	Comparison	Outcome Primary Secondary
17. Biomechanical analysis and modelling of different vertebral growth patterns in adolescent idiopathic Scoliosis and healthy subjects	Retrospective study	Scoliosis (0.89)	Shi et al <sup>22</sup>	Adolescent idiopathic scoliosis		Healthy subjects	Results from this analysis suggest that accelerated growth profiles may encourage supplementary scoliotic progression and, thus, may pose as a progressive risk factor.
18. Non-fusion treatment of adolescent idiopathic Scoliosis by growth modulation and remodelling	Retrospective study	Journal of Paediatric Orthopædics (2.046)	Aronsson et al <sup>4</sup>	Patients with AIS		Non fusion treatment	A brace that applies the appropriate loading and is worn as prescribed may dramatically improve the results of brace treatment. A procedure using external fixation or adjustable anterolateral tethering may achieve a non-fusion correction of AIS.

Out of 18 articles we found three Systematic reviews 13 15 20, one Meta-analysis study 13, five Narrative Reviews<sup>5 7 8 10 18</sup>, nine Retrospective studies<sup>4 9 11 12 14 16 17 19 22</sup> and one Comparative study<sup>21</sup>.

The final 18 articles were critically analysed with the help of the data extraction form and individual articles.

**Discussion**

Managing scoliosis has been a challenging problem in orthopaedics. Despite many research studies over the years the true understanding of the pathogenic factors leading to different onsets and progression of the disease all remains unclear. However, with modern advances in growth modulation techniques in managing idiopathic scoliosis has been published extensively. The results of our final 18 articles following the detailed systematic review have revealed following points. Each has been summarised below.

Article published by Cheung et al mentions major developments in surgical techniques that include robotic assisted pedicle screw placement, vertebral body tethering, vertebral body stapling magnetically controlled growing rods, and sublaminar polyester bands. All these are considered as fusionless growth modulation

techniques.<sup>7</sup> In 2019 Trobisch et al review article describing an innovative non-fusion option of scoliosis correction by insertion of segmental pedicle screws with flexible polyethylene cord anteriorly. In this review short term results of hundred patients were studied. Short term results concluded with over correction and cord rupture mentioned as noted complications.<sup>8</sup>

Shilla procedure was designed to manage early onset scoliosis in patients with growing spines.<sup>23</sup> Wilkinson et al found out from a retrospective review conducted with 21 patients 9 which was a single centre review demonstrated that apex of the primary curve shifts and most patients develop secondary curves following the procedure.

Anterior vertebral growth modulation (AVBGM) in adolescents with idiopathic scoliosis is described as a minimally invasive technique that gradually correct the deformity while preserving some lumbar mobility. However the patient selection for this procedure and predicting the outcome has been challenging. Mandel et al developed a 3D reconstruction computer model helping to answer this question with equal results to existing biomechanical models.<sup>10</sup> Newton et al in a retrospective look into anterior spinal growth tethering (ASGT) in patients with skeletally immature thoracic scoliosis found out that



scoliosis could be corrected while maintaining spinal flexibility and natural growth of the spine. This is a promising alternative technique compared to a rigid fixation for the skeletally immature spine.<sup>11</sup> Scoliosis vertebral growth plate histomorphometry was studied by Bylski-Austrow et al helping us to understand better the bio mechanics of the curve.<sup>12</sup>

Anterior instrumentation and usage of thoracoscopic procedures in managing AIS as compared to childhood scoliosis has been gaining popularity over last decade. In a systematic review conducted by Padhye et al using multi database searches a total of thirteen studies were identified. In a total of 530 patients thoracoscopic procedure was found to have an excellent curve correction, high patient satisfaction and no long-term effect on pulmonary function over the open procedure. However, longer surgical time and intra-operative pulmonary complications were higher in the thoracoscopic procedure.<sup>13</sup>

Distraction based techniques in managing early onset scoliosis (EOS) has been practiced widely. However Magnetically controlled growing rod systems (MCGR) has been relatively new as compared to the traditional growing rod systems (TGR). Eltoral et al reports promising results of MCGR as compared to the current gold standard of TGR in managing EOS.<sup>14</sup>

In managing EOS or paediatric scoliosis various newer growth modulation implants<sup>15</sup>, 3D correction devices with anterior vertebral growth modulation<sup>16</sup> and biomechanical simulation of costo-vertebral and anterior vertebral tethers attempting fusionless treatment<sup>17</sup> have all been tried with promising results.

The final analysis, had three more articles dealing with EOS or paediatric scoliosis. Systematic review by Tis et al describes the modern growth modulation models in comparison to classic rods used in surgery.<sup>20</sup> Driscoll et al published a biomechanical comparison of fusionless growth modulation surgical techniques in paediatric scoliosis. This increasing gives supportive

evidence towards using growth modulation principle to develop instrumentations.<sup>21</sup> Surgical aspects of growth modulation in EOS are well described by Jain et al in an article published in Instructional course lectures.<sup>5</sup>

Out of the eighteen articles analysed in the final selection we found three articles dealing with Adolescent idiopathic scoliosis (AIS) and using growth modulation techniques in managing these. In principle, growth modulation works better with paediatric scoliosis with flexible and growing spines that can be modulated. One study that looks at 2 year results of anterior vertebral body tethering in AIS shows promising results. However, even in this, patients were in an early adolescent group with a mean age of 12 years.<sup>19</sup> Biomechanical analysis on AIS studied by Shi et al<sup>22</sup>, a study similar to the one done by Driscoll et al<sup>21</sup> on paediatric scoliosis shows that abnormal growth profiles seen in AIS are a potential risk factor encouraging supplementary curve progression.

Bracing and non-operative techniques have been used relatively less in modern day practice due to poor patient compliance. However, specific braces designed and used in the principles of growth modulations, in managing AIS have been reported by a study done by Aronsson et al. In that they conclude a brace that applies proper loading and worn in the prescribed manner will equally improve the correction of scoliosis.<sup>4</sup>

This systematic review tries to evaluate the secondary evidence available within this context of managing scoliosis using growth modulation.

Our review had some limitations. We only used a single database (PubMed) for the search other sources such as books, conference proceedings, unpublished data, lectures and ongoing research were not included. Our language settings were limited to, articles published only in English language.

Using growth modulation in managing surgical correction of scoliosis appears to be

effective as traditional correction. With the development of modern instrumentation as mentioned in many studies above, we found out that high level of success rates can be achieved. The clear advantages of this remain that, gradual correction of the deformity while maintaining spinal growth and slow correction of soft tissues

around, making it more compliant to patient and achieving physiological and functional correction. The downside of this seems that limitation in achieving full cosmetic correction, less success rates in AIS. This works best in younger age group where there is further spinal growth namely the EOS.

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## Article 4

# Association between osteoporotic fractures and statin use – A review of literature and results of a preliminary case control study done at National Hospital of Sri Lanka.

## Abstract

Osteoporosis and related fractures are a major health problem all over the world. Several studies have shown that statin use is associated with a reduced risk for osteoporotic fractures among older population.

A case control study was conducted among elderly patients presenting to NHSL accident service. A hundred patients aged more than 60 years who presented with newly diagnosed neck of femur, distal radial and vertebral body wedge fractures following minor trauma were recruited from April 2021 to July 2021. Patients who had previous osteoporotic fractures and previous hip, distal radius and spine surgery were excluded from the study. An additional group of 100 patients who were matched for age and sex presenting to the same unit following minor trauma without osteoporotic fractures were selected as the control group with the same exclusion criteria. Use of statins for more than 4 months and other sociodemographic and lifestyle factors were compared among the two groups.

As no other independent variable was found to be significantly related to osteoporotic fractures between the two groups, univariable analysis was done. The odds of current statin use in cases with osteoporotic fracture (45.2%) were lower than the odds of current statin use (54.8%) in subjects without osteoporotic fractures adjusted OR 0.74, 95% CI 0.42, 1.32) ( $p=0.304$ ).

According to our results there might be a protective effect in statin use against osteoporotic fractures although a statistical significance was not demonstrated. However further studies using larger sample size and randomized control studies are suggested to ascertain a statistically significant result.

**Key words** – osteoporosis, osteoporotic fractures, statins

## Introduction

Osteoporotic fractures or fragility fractures are a major health problem having a considerable socio-economic burden all over the world [1]. It affects the elderly population by increasing their morbidity and mortality while diminishing the quality

of life. It affects a country's economy as the financial burden incurred on the health system to manage these fractures are considerably high.

In Sri Lanka with the increase of the elderly population the social and economic burden of osteoporosis

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related fractures are expected to rise. Due to its significance, prevention of these injuries is as important as finding out novel treatment strategies to manage them.

The osteoprotective effects of 3-hydroxy-3-methylglutaryl coenzyme A reductase inhibitors (statins) are currently well established in in vitro and animal studies. However only a handful of clinical studies have been done to establish its effect in preventing osteoporotic fractures of the elderly.

## Literature Review

Osteoporosis affects 75 million people in Japan, Europe and USA [2]. Epidemiological data with regard to incidence and prevalence of osteoporotic fractures in Sri Lanka are lacking in published literature.

Osteoporosis is defined as a general disorder of the skeleton characterised by low bone mass and deterioration in the microarchitecture of the bone tissue, which is translated into a deterioration of bone resistance predisposing to fractures [3].

According to the definition, the key clinical fact is fragility fracture. Therefore, the presence of osteoporosis without fracture makes diagnosis difficult. Thus, the diagnosis is based on the confirmation of low bone mineral density (BMD) on dual-energy x-ray absorptiometry (DEXA) scan. Therefore, in 1994 the WHO agreed on an operative definition based on cut-off points of BMD for postmenopausal women. The normal level for BMD was set at a value higher than -1 standard deviation (SD) relative to the average for young adults. For osteopenia, values of BMD between -1 and -2.5 SD was set. Osteoporosis was defined to be set at values of BMD lower than -2.5 SD. Established osteoporosis was defined when, along with these conditions, were associated one or more osteoporotic fractures [4]. It has been recommended that the same cut-off values be used for osteoporosis in males [5]. Even though

BMD is the most quantifiable risk factor of future fracture, many other clinical risk factors come in to play with regard to the occurrence of fractures. As a solution, WHO recently introduced the FRAX algorithm to help clinicians in therapeutic decision making. FRAX is a web-based calculator which, apart from BMD, accommodates multiple clinical risk factors in estimating fracture risk. Since its implementation, FRAX has undergone many alterations and many country-specific FRAX models have been developed. (<http://www.shef.ac.uk/FRAX>).

In accordance with the WHO criteria, the estimated prevalence of osteoporosis in white women over 50 years of age is 15% when one of the three usual locations (spine, hip or wrists) is considered, and 30% when measured in all of them [6]. The prevalence increases with age from 15% for the period between 50 and 59 years of age, up to more than 80% in ages over 80 years [7]. In males, the prevalence of osteoporosis is lower, 8% according to the NHANES study [8].

Approximately 1.6 million hip fractures (neck of femur) annually occur worldwide and this figure is estimated to reach 4.5-6.3 million by 2050 [9, 10]. According to the International Osteoporosis Foundation, the incidence of neck of femur (NOF) fractures in Sri Lanka is estimated to rise from the 2006 figure of nearly 2700 to 4900 in 2020 and 6900 in 2041. However according to the growth rate of elderly sector of the population these figures could become higher [11]. Fracture NOF is the most feared osteoporosis-related fracture owing to the financial burden it incurs and the associated mortality and morbidity. It is estimated that the mortality of NOF fractures during the first year is 20-25% [12]. The increased mortality could persist up to five years after a NOF fracture. It also has profound effects on physical independence. Nearly 40% of hip fracture survivors have walking disability while 60% require assistance to maintain day to day physical activities [13]. Furthermore, a third of NOF

fracture patients are totally physically dependent or require nursing home placement

at one year following fracture [14]. Vertebral fractures present with its own set of characteristics different from NOF fractures. Only one third of them are symptomatic [15]. Therefore, most of the vertebral fractures are detected as an incidental finding. They are also associated with acute and chronic backache, loss of mobility and functions. Furthermore, they lead to more vertebral fractures and non-vertebral fractures in later years [16, 17]. Forearm fractures behave different to other osteoporotic fractures. It lacks the classical exponential rise with advancing age [18] seen with typical osteoporosis-related fractures such as NOF and vertebrae. They also tend to occur in relatively young people [19]. Although no increased mortality is seen following distal forearm fractures, increased incidence of pain and numbness of affected hand is reported.

Most preventative and curative drugs currently used for osteoporosis such as raloxifene, denosumab, bisphosphonates, and calcitonin work by the anti-resorptive mechanism. [20–23] Alendronate and risedronate are the main oral bisphosphonates used in Sri Lanka to treat patients with osteoporosis and a high fracture risk. Zoledronic acid is becoming popular due to its reduced frequency of administration. The inconvenience of injections and prohibitive cost make teriparatide a reserve drug to treat osteoporosis. Upper gastro-intestinal adverse events associated with oral bisphosphonates are the most common side effects seen among patients. Myalgia, bone pain and arthralgia are also unwanted side effects. They can be severe enough to result in discontinuation of oral bisphosphonates in some people [24].

The 3-hydroxy-3-methylglutaryl coenzyme A reductase inhibitors (statins) are widely used as a mainstay in preventing and treating cardiovascular disease (CVD). According to recently published studies they also appear to be potentially promising drugs for osteoporosis. The mechanisms of statin's effects on the bone have been examined

by a number of researchers. The current literature agrees that the effects of statins on the bone may involve a number of mechanisms including proliferation, differentiation and protection of osteoblasts while reducing osteoclastogenesis [25]. As a result, statins which are both anti-resorptive and anabolic agents may play a major role in the clinical management of osteoporosis.

To that end several case-control studies have been published showing that statin use is associated with a reduced risk for osteoporotic fractures among older population [26-28]. Other studies have concluded that bone mass is higher among patients taking statins [29, 30]

After analysing results of 4 large prospective studies [31-34] and cumulative Meta-analysis of observational Studies and controlled trials published until 2002, Bauer et al concluded that statins were associated with a consistent and clinically meaningful reduction in hip and vertebral fractures. They also suggested the necessity of carefully controlled specifically designed Clinical trials to test the effects of statins on skeletal metabolism [35] A meta-analysis published by An et al in 2017 also indicates that statin treatment could be associated with a decreased risk of overall fractures and hip fractures with an increased BMD at the hip and lumbar spine. It also showed that statin treatment may have a greater effect on males than females [36]. A study conducted in Australia on a medical population showed a dose dependant relationship between the diagnosis of osteoporosis and statins [37] In a population-based case control study published in 2017, Cheng et al concluded that the odds of current statin use in cases with hip fracture were lower than the odds of current statin use in subjects without hip fracture in elderly in Taiwan. [38]. The most recently published population-based study we could find was also done in Taiwan by Chen et al among patients with COPD. They demonstrated a beneficial effect of statins in patients with COPD against the occurrence of NOF fractures at 10 years follow

up [39]. Clinical studies done in south asian cohorts in this regard have been lacking so far and we were not able to find any studies done in Sri Lanka.

The Accident & Orthopaedic Service of the National Hospital of Sri Lanka being the Level 1 Trauma Care Centre in Sri Lanka treats about 300 patients per day, averaging about 100,000 patients per year. About 30,000 patients are treated as in ward [40]. Out of that a significant proportion of patients belong to the elderly group presenting with low energy fragility fractures. From our study we hoped to establish the association between statin use and osteoporotic fractures when it comes to a clinical setting among patients presenting to a trauma unit.

## Materials and Methods

Patients above the age of 60 years who present to accident service NHSL within 48hrs of low energy trauma having clinically and radiologically confirmed fragility fractures were recruited to the study starting from April 2021 to July 2021. These included necks of femur fractures, distal radial fractures and vertebral body wedge fractures. The patients who have previously sustained above fractures, those with pathological fractures, those diagnosed with rheumatoid arthritis and those who had undergone previous surgery in the hip radius and spine were excluded from the study. A hundred such cases were collected consecutively. Age and sex matched patients who presented to NHSL accident service with low energy trauma with no clinical or radiological evidence of osteoporotic fractures were recruited using the same inclusion and exclusion criteria until 100 controls were collected. Age was matched by selecting controls within 5 years of the year of birth of the cases. Data collection was done using an interviewer administered questionnaire.

Ethical approval was taken for this study from the Ethical review committee at NHSL (AAJ/ETH/COM//2021/MAR). Statistical analysis was performed through IBM SPSS 23 statistics package.

## Results

The study group included 200 patients (100 cases and 100 controls.) Among the cases and controls 69 were females and 31 were males in each group. The cases had a mean age of 72.86 years with a range of 60 - 92 years (SD – 9.18) The mean age of the control group was 71.06 years with an age range of 60 – 96 years. (SD – 8.49) Among the study participants 69% were females and 31% were males. Among the patients with fragility fractures fulfilling the inclusion criteria, 92% had NOF fractures. Distal radius fractures and vertebral body wedge fractures that fulfilled the criteria were 6% and 2% respectively.

Among the 33 cases who were currently on statins for more than 4 months, all of them were on atorvastatin. Among the 40 controls who were on statins 35 was on Atorvastatin and while 3 subjects were on simvastatin and 2 were on Rosuvastatin.

We compared the distributions of the sociodemographic characteristics, lifestyle factors and comorbidities between osteoporotic fracture cases and controls using the Chi-square test for categorized variables. Student t test was used to examine the difference of mean age between fracture cases and controls (tables 1 and 2). No statistically significant difference was observed among cases and controls with regard to the above. ( $p < 0.05$ ). As no other independent variable was found to be significantly related to osteoporotic fractures in the univariable analysis, we did not perform the multivariable logistic regression model. The univariable logistic regression showed that the odds of current statin use in cases with osteoporotic fracture (45.2%) were lower than the odds of current statin use (54.8%) in subjects without hip fracture (adjusted OR 0.74, 95% CI 0.42, 1.32) ( $p = 0.304$ ). Our results suggest that there may be a negative association between statin use and osteoporotic fractures (i.e. statins may have a protective affect against osteoporotic fractures) in the studied population although a statistical significance was not demonstrated. ( $p < 0.05$ )

	Overall	Cases	Control	P value	Test
Mean Age	71.96 (60-96) Median 70.00	72.86 (60-92)	71.06 (60-96)	0.152	Independent sample t test
<b>Marital status</b>					
Married	192 (96.0%)	96 (96.00%)	96 (96.00%)	0.565	Chi Square
Single	7 (3.5%)	3 (3.00%)	4 (4.0%)		
Separated/Widowed	1 (0.5%)	1 (1.00%)	0		
<b>Functional Status</b>					
Independent	175 (87.5%)	92 (92.0%)	83 (83.0%)	0.054	Chi Square
Dependent	25 (12.5%)	8 (8.0%)	17 (17.0%)		
<b>Occupational Status</b>					
Working	38 (19.0%)	20 (20.0%)	18 (18.0%)	0.843	Chi Square
Retired	74 (37.0%)	38 (38.0%)	36 (36.0%)		
Unemployed	88 (44.0%)	42 (42.0%)	46 (46.0%)		
<b>Physical Activity Level</b>					
Mild	38 (19.0%)	16 (16.0%)	23 (23.0%)	0.364	Chi Square
Moderate	64 (32.0%)	34 (34.0%)	30 (30.0%)		
High	97 (49.0%)	50 (50.0%)	47 (47.0%)		
<b>Smoking Status</b>					
Smoking	48 (24.0%)	22 (22.0%)	26 (26.0%)	0.427	Chi Square
Non-Smoking	152 (76.0%)	78 (78.0%)	74 (74.0%)		
<b>Alcohol consumption</b>					
Yes	46 (23.0%)	24 (24.0%)	22 (22.0%)	0.737	Chi Square
No	154 (77.0%)	76 (76.0%)	78 (78.0%)		

**Table 1** – Analysis of the association between demographic and lifestyle factors among cases and controls

	Overall	Cases	Control	P value	Test
<b>Diabetes Mellitus</b>					
Yes	98 (49.0%)	50 (50.0%)	48 (48.0%)	0.777	Chi Square
No	102 (51.0%)	50 (50.0%)	52 (52.0%)		
<b>Hypertension</b>					
Yes	104 (52.0%)	46 (46.0%)	58 (58.0%)	0.103	Chi Square
No	96 (48.0%)	54 (54.0%)	42 (42.0%)		
<b>Ischaemic Heart Disease</b>					
Yes	32	14 (14.0%)	18 (18.0%)	0.440	Chi Square
No	168	86 (86.0%)	82 (82.0%)		
<b>Dyslipidaemia</b>					
Yes	57 (28.5%)	30 (30.0%)	27 (27.0%)	0.638	Chi Square
No	143 (71.5%)	70 (70.0%)	73 (73.0%)		
<b>Cerebrovascular Disease</b>					
Yes	12 (6.0%)	4 (4.0%)	8	0.373	Chi Square
No	188 (94.0%)	96 (96.0%)	92		

**Table 2** – Analysis of the association between co morbidities among cases and controls



	Value	95% confidence interval	
		Lower	Upper
Odds Ratio for Cases or controls (Cases / Controls)	0.739	0.415	1.317
For cohort Usage of statin = Yes	0.825	0.571	1.192
For cohort Usage of statin = No	1.117	0.904	1.379
N of valid cases	200	0.415	1.317

**Table 3** – Risk estimate for cases and controls for statin use

## Discussion

The main conclusion of the present study is that oral statin use might have a protective effect against osteoporotic fractures. This is in keeping with all the previous published studies done in this regard. However, all previous studies had been done as population-based cohort or case control studies. According to our knowledge this is the only single centre case control study done regarding the association between statins and osteoporotic fractures. One of the main drawbacks of population-based case control studies is the potential explanation of its results by the healthy drug user effect. The population receiving preventive oral statins for the cardiovascular disease or dyslipidaemia may exhibit certain behaviours that put them at lower risk of osteoporotic fractures. This may include better health insight and help-seeking behaviour which put them at a lower risk of sustaining falls and other minor trauma leading to fractures. Since both our cases and controls were recruited from among patients presenting to a trauma centre with minor trauma, healthy drug user effect is minimised. DEXA scans were not performed on our patients to confirm the osteoporosis unlike other studies due to the lack of facilities. Collecting data regarding osteoporotic fractures is difficult in the Sri Lankan population as we lack a functional database or a registry of patients who are affected.

So far, we are lacking a nationwide program for the prevention of osteoporotic fractures. Also, DEXA scans are not routinely performed as a means of diagnosing osteoporosis due to the unavailability of scan machines and

the cost. Consequently, in most of these patients the first indication of established osteoporosis maybe an occurrence of a low energy fragility fracture. Therefore, within the framework of the Sri Lankan health system with its financial and practical constraints, the occurrence of fragility fractures in the elderly population can be used as an indirect indication of the presence of osteoporosis.

Statins are widely used in the Sri Lanka for the treatment of cardiovascular and cerebrovascular diseases as well as dyslipidaemia. As the above diseases are prevalent in the same age group where osteoporotic fracture risk is increased, the osteoprotective effects of statins may be a blessing in disguise for these patients. If these effects are properly established from extensive studies, statins may in fact be indicated in the future in its own right for the prophylaxis and treatment of osteoporotic fractures.

One of the main limitations of this study compared to other published studies is the small number of cases and controls. This study was conducted during the Covid-19 pandemic. Therefore, the number of cases which could be obtained in the stipulated time period was difficult. Also, the study was conducted only at NHSL. A larger sample size and a more statistically significant result could have been obtained had the study been a multicentre one.

We suggest further population based observational and randomised studies using larger samples to establish the connection between statin use and osteoporotic fractures. Multicentre studies could be conducted in

trauma centers using a larger population with the same methodology. It should also be emphasised regarding the necessity of creating a patient database with regard to osteoporotic fractures at institutional and national level in Sri Lanka which will make the data accessible to researchers

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## Article 5

# Pattern of second osteoporotic fractures. A descriptive study in two tertiary care centers in Sri Lanka

### Abstract

**Introduction:** Second osteoporotic fractures have higher morbidity and mortality than prior osteoporotic fractures.

**Materials and methods:** A retrospective case series of patients admitted to two tertiary care hospitals with second osteoporotic fractures from April 2020 to March 2021 assessed demography, time since the first osteoporotic fracture, fracture locations, comorbidities, menopausal age, predisposing drugs and habits, parental history, fall risk, BMI, family support and treatment.

**Results:** Fifty-four patients were studied. Forty-nine (90.7%) were females, Mean age was 75.8 years (57-95). Mean time since the first fracture was 3.67 years (3/12-12). Twenty-six (48.1% p 0.00) had second fracture within 2 years since first osteoporotic fracture. Major osteoporotic sites involved in 79.6% of first fractures and 85.1% of second fractures. Proximal femur was the predominantly involved major site in first (23/43 p 0.00007) and second (35/46 p 0.00) fractures. Females who had premature/early menopause were significantly associated with the second fracture before the age of 75 years (10/15 p 0.03) and with non-major site involvement (5/15 p 0.035). Forty-nine (90.7%) patients had fall risk. Forty-seven (87.03%) patients lived with their family. Only 2/54 had DEXA. Patients who were aware of their condition had better compliance for supplements (9/12 p 0.0003). Anti-osteoporotic agent was used by 3/54 patients.

**Discussion and Conclusion:** Female gender, premature/early menopause and prior proximal femur fracture are the main risk factors for second osteoporotic fracture. Following all low energy fractures, immediate osteoporosis diagnostic work up with commencement of anti-osteoporotic regime accordingly and modification of fall risk are recommended to reduce the imminent risk of second osteoporotic fracture occurrence

**Keywords:** Prior osteoporotic fracture; Second osteoporotic fracture; Osteoporosis; Proximal femur fracture; Low energy fracture

### Introduction

Osteoporotic fractures are the main complication of osteoporosis which is a chronic progressive systemic skeletal disease, characterized by deteriorating bone mineral density and micro-architecture.

Osteoporotic fractures are also called as fragility fractures. These fractures are low energy fractures which occur in inherently weak bones due to a fall from standing height or less. These fractures imply the diagnosis of osteoporosis regardless of a t score of  $\leq 2.5$  in a dual –energy

x ray absorptiometry scan (DEXA) which is a standard investigation to diagnose osteoporosis.

NICE guideline and guidelines of National Osteoporosis Foundation mention proximal femur, spine and distal radius as most commonly affected sites by osteoporotic fracture. According to World Health Organization (WHO) proximal humerus also is a major site of osteoporotic fracture. In addition to these sites, pelvic bone, sacrum, ribs, distal humerus, distal femur and ankle are also affected by osteoporotic fractures due to low energy trauma. These are called minor osteoporotic fracture sites [16]. These fractures have their own patient characteristics, morbidity and mortality.

Prior osteoporotic fracture is an indicator of subsequent osteoporotic fracture. Morbidity, mortality and socio-economic burden on patient, family and health care are higher in second osteoporotic fracture than prior osteoporotic fracture. The elderly population is at high risk to sustain osteoporotic fractures. According to the population prediction for Sri Lanka, the population of aged more than 50 years will rise from 24% in 2013 to 38% in 2050. [12]. Therefore, preventive measures are mandatory to reduce the incidence of second osteoporotic fracture.

Osteoporosis is not recognized as a major health issue by health authorities of Sri Lankan government. In Sri Lanka access to the DEXA scan is limited to diagnose osteoporosis (0.1 DEXA scan machine per 1 million people) [12]. The knowledge on factors associated with second fragility fracture will be helpful for the clinical diagnosis of osteoporosis and commencement of early bisphosphonate therapy.

## Materials and Methods

To describe the associated factors and clinical characteristics of second osteoporotic fractures, a retrospective case series study was carried out from 01st April 2020 to 31st March 2021 among the patients admitted with second osteoporotic fracture to accident service wards and orthopaedic wards in Teaching Hospital-Jaffna and National Hospital of Sri Lanka

Patients with a fracture due to a fall from standing height with history of prior fracture due to a fall from standing height were included in the study. Patients with the age less than 40-year-old at the time of first osteoporotic fracture and those with peri-prosthetic or peri-implant fracture as the first or second fracture, malignancy with known metastasis to bone or femoral neck t score > 2.5 in DEXA scan which was done after first fracture were excluded from the study.

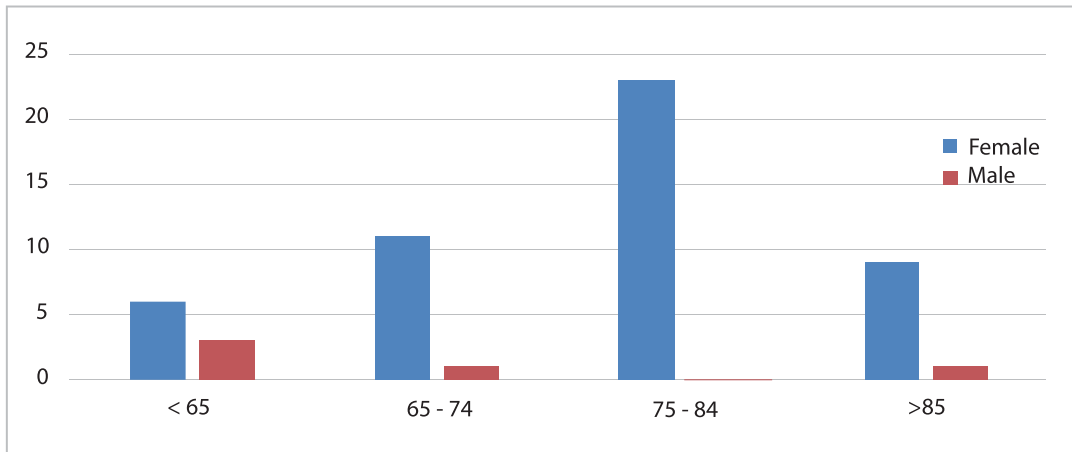
Consecutive sampling method was used. Interviewer administered questionnaire containing patient demography and study variables (Annexure), Bed Head Tickets (BHTs), clinic books and diagnosis cards were used as study instruments. Demography, time since the first osteoporotic fracture, fracture locations, mode of management of prior fracture, comorbidities, menopausal age, predisposing drugs and habits, parental history, fall risk, BMI, family support and treatment were assessed

Data entry and analysis of descriptive statistics were done by using SPSS version 21. Frequency and proportions were calculated for categorical variables. Means were calculated for continuous variables. For both type of variables ranges, maximum and minimum were reported. To compare the groups of categorical variables Pearson's chi-square test and Fisher's exact test were used. To compare the groups of continuous variables t-tests was used. A p value of less than 0.05 was considered as a statistically significant finding.

Ethical clearance was obtained from Ethics Review Committee, Post Graduate Institute of Medicine, University of Colombo and Ethics Review Committee, National Hospital of Sri Lanka.

## Results

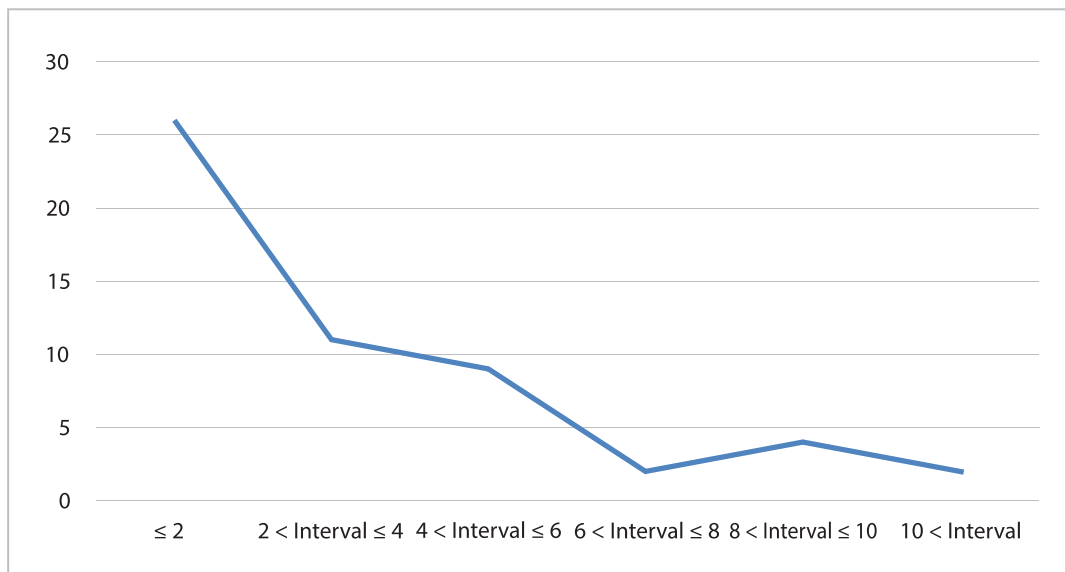
Fifty-four patients with second osteoporotic fractures were studied. Forty-nine (90.7%) were females (p 0.000). Mean age was 75.8 years (Range 57-95). Twenty-three (42.6%) patients were in the 75-84 age group.



**Figure 1 – Age and gender of study population**

The mean time interval between first and second osteoporotic fractures was 3.67 years (Range 3/12-12). The risk of getting second osteoporotic fracture is higher within short

intervals than long intervals. Twenty-six (48.1%) patients had the second osteoporotic fracture within 2-year interval since first osteoporotic fracture (p 0.000).



**Figure 2 – Interval between first and second osteoporotic fractures**

Proximal femur was the predominant site involved in the second osteoporotic fracture. Thirty-five (64.8%) patient had proximal femur fracture as the second osteoporotic fracture. Major osteoporotic sites were involved in 46 (85.2%) second osteoporotic fractures.

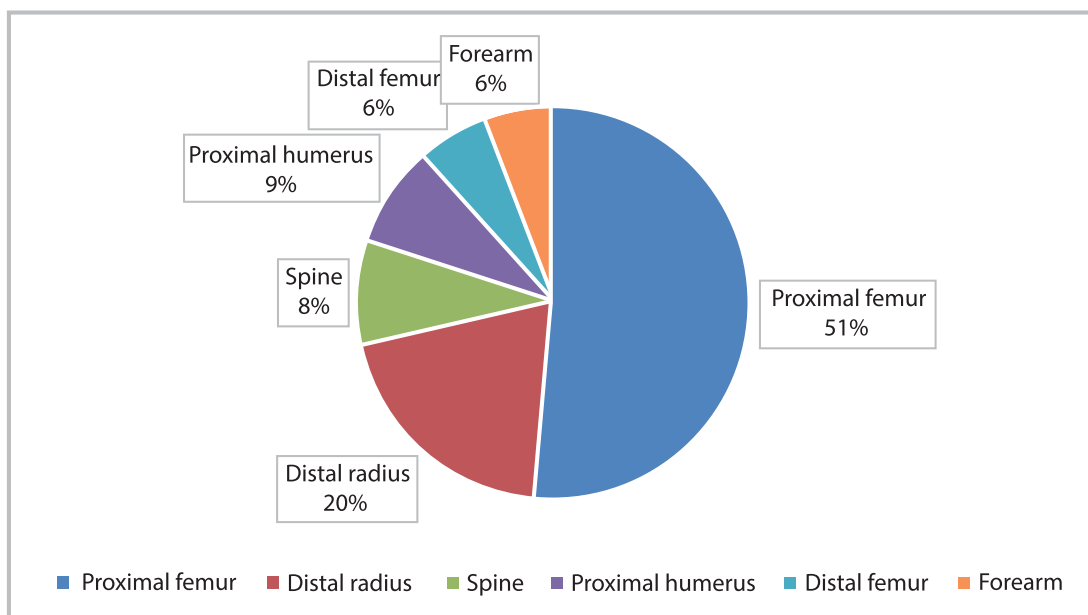
Forty-three (79.6%) patients had major osteoporotic site involvement in the first osteoporotic fracture. Out of the first major osteoporotic site fractures, proximal femur was involved significantly (23/43 p 0.0001).

Fracture site	Number of patients	Percentage
<b>Second osteoporotic fracture</b>		
<b>Major osteoporotic sites</b>	<b>46</b>	<b>85.2%</b>
Proximal femur	35	64.8%

Fracture site	Number of patients	Percentage
Spine	01	1.9%
Distal radius	06	11.1%
Proximal humerus	04	7.4%
<b>Minor osteoporotic sites</b>	<b>05</b>	<b>9.2%</b>
Ankle	03	
Distal femur	02	
<b>Others</b>	<b>03</b>	<b>5.6%</b>
Tibial plateu	02	
Femur shaft	01	
<b>First osteoporotic fracture</b>		
<b>Major osteoporotic sites</b>	<b>43</b>	<b>79.6%</b>
Proximal femur	23	42.6%
Spine	06	11.1%
Distal radius	10	18.5%
Proximal humerus	04	7.4%
<b>Minor osteoporotic sites</b>	<b>08</b>	<b>14.8%</b>
Ankle	02	
Distal femur	03	
Distal humerus	01	
Pelvis	02	
<b>Others</b>	<b>03</b>	<b>5.6%</b>
Tibial shaft	01	
Forearm	02	

**Table 1** – Sites of first and second osteoporotic fractures

Second proximal femur fracture was preceded by proximal femur (18/35), distal radius (7/35), spine (3/35), proximal humerus (3/35), distal femur (2/35) and forearm bone (2/35) fractures.



**Figure 3** – Distribution of first osteoporotic fractures among second osteoporotic fracture which involved proximal femur.



Forty-four females were able to provide the age at menopause. Mean age at menopause was 46.3 years (Range 37-60). Fifteen (15/44) patients had premature or early menopause (Below the age of 45 years). Mean ages of second osteoporotic fracture were 70.3 and 78.2 years for premature or early menopausal age group and normal menopausal age group respectively. Occurrence of two osteoporotic fractures before the age of 75 years was significantly associated (p 0.03) with premature or early menopause patients (5/15) compared to normal menopausal age group (9/29). Significant association (p 0.035) was found between the involvement of non-major osteoporotic sites in second fracture

and premature or early menopausal age (5/15) compared to normal menopausal age (2/29).

Males with second osteoporotic fractures had significant association with smoking (3/5 p 0.0004) and alcoholism (2/5 p 0.007) compared to females (00/49).

Comorbidities, drug history, parental history, mode of management of prior osteoporotic fracture, history of fall in the interval between first and second osteoporotic fractures, fall related individual risk factors and poor family support did not show any statistically significant association with the second osteoporotic fractures.

Associated factor	Frequency
<b>Comorbidities that can cause bone loss</b>	<b>33(61.1%)</b>
Hypertension	22
Diabetis Mellitus Type 2	15
Bronchial Asthma	03
Hyperthyroidism	01
Dementia	02
Parkinsonism	01
<b>No comorbidities that can cause bone loss</b>	<b>21 (38.9%)</b>
<b>On Steroids for more than 3 months</b>	<b>03 (5.6%)</b>
<b>Not on steroids for more than 3 months</b>	<b>51 (94.4%)</b>
<b>Use of Medications that can induce osteoporosis</b>	<b>25 (46.3%)</b>
Antihypertensives	22
Thyroxin	02
Selective serotonin reuptake inhibitor	01
Protein pump inhibitor	03
<b>No usage of Medications that can induce osteoporosis</b>	<b>29 (53.7%)</b>
<b>Parental history</b>	<b>45 (88.2%)</b>
<b>No parental history</b>	<b>06 (11.8%)</b>
<b>Low BMI (kg/m2) &lt; 18.5</b>	<b>10 (18.52%)</b>
<b>Normal BMI 18.5-22.9</b>	<b>26 (48.15%)</b>
<b>High BMI ≥ 23</b>	<b>18 (33.33%)</b>
<b>History of fall in between first and second osteoporotic fractures</b>	<b>26 (48.15%)</b>
<b>No History of fall in between first and second osteoporotic fractures</b>	<b>28 (51.85%)</b>

Associated factor	Frequency
<b>Fall related risk factors</b>	<b>49 (90.7%)</b>
Assistance or supervision for mobility	30
Unsteady gait	24
Visual impairment	23
Auditory impairment	13
Urinary or fecal incontinence, urgency or frequency	24
<b>No fall related risk factors</b>	<b>5 (9.3%)</b>
<b>Operative management for first osteoporotic fracture</b>	<b>25 (46.3%)</b>
<b>Non operative management for first osteoporotic fracture</b>	<b>29 (53.7%)</b>
<b>Living alone</b>	<b>04 ( 7.4%)</b>
<b>Living with the caregiver who is not a family member</b>	<b>03 (5.5%)</b>
<b>Living with the spouse or a family members</b>	<b>47 (87.1%)</b>

**Table 2** – Associated factors with second osteoporotic fractures

Twelve (12/54, 22.2%) patients were told by a doctor that they have osteoporosis. Two patients (2/12) had DEXA within last 2 years. They (2/2) were found to have T value  $\leq -2.5$ .

Eleven (20.4%) patients had calcium and vitamin D supplements. Five (9.3%) patients had calcium supplement alone. Out of 12 patients who were told by a doctor that they have osteoporosis, nine (9/12) had calcium and/or vitamin D. Out of 42 patients who were not aware about their condition regarding osteoporosis, three (3/42) patients had calcium and/or vitamin D supplement. Taking calcium and/or vitamin D had significant association with the knowledge that they have osteoporosis, compared to not knowing whether or not having osteoporosis ( $p = 0.0003$ ). Three (3/54) patients were on alendronate for last one year. Two of them had DEXA before starting alendronate and one patient was started on alendronate following a major osteoporotic fracture.

## Discussion

Our study revealed the approximate ratio between males and females was 1:9. According to the literature the risk of sustaining a second osteoporotic fracture was slightly greater in males than in females [3,14]. However higher mortality rate among males following first

osteoporotic fracture and higher fall related risk and earlier beginning of bone resorption process among females were possible causes leading to higher frequency of females than males in our study group. A study to compare the mortality rate of Sri Lankan males and females following first osteoporotic fracture is recommended for future directions.

Wang L et al found mean ages of second osteoporotic fracture were 74.1 and 75.1 among males and females respectively. In their study 68% of patients with second osteoporotic fractures were over 70 years [15]. Our study revealed the mean age at second osteoporotic fracture was 75.8 years and 61.1% of the study population were equal or over 75 years. With increasing age, above mentioned middle old and oldest old age category patients were likely to suffer with lack of full recovery, compromised healing and infection following second osteoporotic fracture.

Many studies regarding second osteoporotic fracture highlighted the imminent risk of second osteoporotic fractures following the first osteoporotic fracture [1,2]. In our study also significant number of patients had equal or less than 2-year interval between first and second osteoporotic fractures. This finding emphasized the requirement of immediate work up related to osteoporosis and starting the pharmacological management early. Mean interval between first

and second osteoporotic fractures in our study group was 3.67 years which was an adequate period to implement necessary preventive measures to reduce second fracture rate. This finding was similar to a Chinese analysis by Ruan W D et al on the risk factors of second osteoporotic fractures [13].

Recent studies or articles emphasized on the clinical definition of an osteoporotic fracture rather than the definition based on Bone Mineral Density. A Fracture due to a low energy trauma (fall from standing height or less) in patients over 50 years was defined as an osteoporotic fracture. Though we added the patients over 40 years in the inclusion criteria, age of our study population was distributed from 57 years to 95 years.

In our study spinal fracture was not the commonest osteoporotic fracture. Proximal femur fracture was the commonest first osteoporotic fracture. Distal radial fracture and minor osteoporotic site fractures were second and third respectively. A Canadian study by Adachchi J D et al also had similar findings [17]. Asymptomatic nature of spinal fractures was a possible cause for the reduction of spinal fracture cases.

The proximal femur fracture was the most common second osteoporotic fracture not only in our study, Choi J Y et al and Adachchi J D et al also had the same finding [17, 18]. Being the second osteoporotic fracture and involvement in the hip region usually result in highest morbidity and mortality. Majority of second osteoporotic fractures involving the proximal femur were preceded by prior proximal femur osteoporotic fractures of contralateral side in our study. Second osteoporotic fractures of spinal column and majority of distal radial bones did not show this kind of association with different level or contralateral side respectively. This was contrary to the finding of Choi J Y et al and Adachchi J D et al.

Routine risk factors for osteoporosis except the menopausal age in females and habits of males did not show any statistically significant

association with second osteoporotic fractures in our study. Failure of pharmacotherapy as a main cause for the second osteoporotic fracture was supported by the insignificant association of those risk factors too.

Findings related to premature and early menopausal females emphasized the importance of extra precautions to prevent second osteoporotic fractures among them. Involvement of non-major osteoporotic sites in this category was an important finding not to neglect immediate diagnostic work up and early pharmacotherapy, even in the absence of traditional major osteoporotic fractures.

Our study found lack of standard diagnostic work up, poor follow up and poor compliance to pharmacotherapy even after sustaining major osteoporotic fractures. In developed countries also unsatisfactory work up and treatment were not uncommon problems. An US study by Balasubramanian A et al and a Swiss study by Morell S et al also revealed the lack of adherence to the standard diagnostic work up and to the pharmacotherapy [19,20]

### **Strengths and limitations**

Our study analyzed not only the traditional major osteoporotic fractures, but other sites which contribute to the increasing number of fractures and socio-economic burden also were analyzed. Maximum effort was undertaken to avoid the impact of all three waves of Covid 19 pandemic in Sri Lanka in data collection process.

Retrospective study design, convenience sampling method and mainly being a questionnaire based study were some noticeable weaknesses in the study design. Asymptomatic nature of spinal fracture could have underestimated the frequency of spinal fractures. To make sure reaching adequate sample size two hospitals - a national hospital and a teaching hospital in the country were included in the study. Differences in various aspects between these two hospitals were not considered as a study variable.

## Conclusion and recommendations

Female gender and prior proximal femur fracture were the main non-modifiable risk factors leading to the occurrence of the second osteoporotic fracture. Smoking and alcoholism in males and premature and early menopausal age in females had significant influence on characteristics of second osteoporotic fractures. Imminent risk of second osteoporotic fracture following prior osteoporotic fracture was high. Following an osteoporotic fracture, poor quality in diagnostic work up and pharmacotherapy were noted. Immediate diagnostic work up of osteoporosis including DEXA and commencement

of anti-osteoporotic therapy accordingly with adequate health education and fall prevention strategies following first osteoporotic fracture are recommended to reduce the second osteoporotic fracture occurrence in the future. Establishment of community osteoporosis screening services in Sri Lanka and free supply of calcium and vitamin D supplements and anti-osteoporotic therapy accordingly are cost effective preventive measures to reduce the socio-economic burden of osteoporotic fractures. Studies regarding factors influencing the diagnostic work up and pharmacotherapy for osteoporosis in Sri Lanka are recommended for future directions.

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**ANNEXURE**

Please place the check mark (√) in the appropriate box or when relevant specify the answer.

1. Current fracture (Second osteoporotic fracture)

Proximal femur	<input type="checkbox"/>	Distal humerus	<input type="checkbox"/>
Spine	<input type="checkbox"/>	Ankle (malleolar)	<input type="checkbox"/>
Distal radius	<input type="checkbox"/>	Distal femur	<input type="checkbox"/>
Proximal humerus	<input type="checkbox"/>	Pelvic bone including acetabulum	<input type="checkbox"/>

Others .....

Diagnosis (Please refer BHT and Images).....

2. Prior fracture (First osteoporotic fracture)

Proximal femur	<input type="checkbox"/>	Distal humerus	<input type="checkbox"/>
Spine	<input type="checkbox"/>	Ankle (malleolar)	<input type="checkbox"/>
Distal radius	<input type="checkbox"/>	Distal femur	<input type="checkbox"/>
Proximal humerus	<input type="checkbox"/>	Pelvic bone including acetabulum	<input type="checkbox"/>

Others .....

Diagnosis (Please refer diagnosis card, clinic book, BHT and images) .....

3. How was the prior fracture managed?

Operative Management		Non-operative management	
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Specify: .....(Please refer diagnosis card and clinic book)

4. What is the interval between first and second fractures?

.....

5. Did your father or mother sustain a fracture following a fall after the age of 40 years?

Yes  No

6. If yes, what bone was fractured?

Hip	<input type="checkbox"/>
Spine	<input type="checkbox"/>
Wrist	<input type="checkbox"/>

Others .....

7. Have you been diagnosed with any of the following conditions?

Condition	Yes	No	Condition	Yes	No
Rheumatoid Arthritis			Hyperparathyroidism		
Type 1 Diabetes Mellitus			Osteogenesis Imperfecta		
Type 2 Diabetes Mellitus			Menopause before 45 years		
Hyperthyroidism			Hypogonadism		
Chronic Kidney Disease			Chronic liver disease		
Crohn's disease/Ulcerative colitis			Celiac disease		
Parkinsonism			Dementia		

Other comorbidities .....

(Please refer BHT and clinic books)

8. Are you on any of the following medication before this fracture occurring?

Medication	Yes	No	Medication	Yes	No
Antidepressant- SSRI			Warfarin		
Anti-seizure medication			Heparin		
Anti-acid medication-PPI			Thyroxin		
Anti-hypertensives			Diuretics		
Opioids			Hypnotics		
Psychotropic drugs			Laxatives		

(Please refer BHT and clinic books)

9. Are you on steroids for more than 3 months?

Yes  No

10. Do you smoke?

I never smoked   
 I smoked in the past   
 I smoke currently

11. Do you consume more than 14 units of alcohol per week?

(6 pints of 4% beer or 7 glasses of 11.5% wine or 14 single shots of 40% alcohol)

Yes  No  Don't know

12. Have you fallen in the interval between first and second osteoporotic fractures beside the fall that led to second osteoporotic fracture?

Yes  No  Don't know

13. During the interval between first and second osteoporotic fractures

	Yes	No
Did you require assistance or supervision for mobility?	<input type="checkbox"/>	<input type="checkbox"/>
Did you have unsteady gait?	<input type="checkbox"/>	<input type="checkbox"/>
Did you have visual impairment?	<input type="checkbox"/>	<input type="checkbox"/>
Did you have auditory impairment?	<input type="checkbox"/>	<input type="checkbox"/>
Did you have urinary or faecal incontinence, urgency or frequency?	<input type="checkbox"/>	<input type="checkbox"/>

14. Have you ever been told by a doctor as you have osteoporosis?

Yes  No

15. Have you ever had DEXA scan test?

Yes, I had less than 2 year ago   
 Yes, I had more than 2 year ago   
 No, I have never had   
 Don't know

(Please refer clinic books)

16. What are the results of DEXA scan?

I have osteoporosis   
 I have osteopenia   
 I have low bone mineral density in one area   
 I have normal or highbone mineral density   
 Don't know

(Please refer clinic books)

17. Do you take following supplements?

Medication	Yes	No
Calcium		
Vitamin D		

18. Do you take medication for osteoporosis?

- Yes, doctor has prescribed
- No, doctor has not prescribed
- No, doctor said that I cannot take medication for osteoporosis
- Don't know


(Please refer BHT, Clinic book and diagnosis card)

19. Have you been prescribed any of the following drugs?

Medication	Yes	No	Medication	Yes	No
Alendronate			Denosumab		
Ibandronic acid			Raloxifene		
Zoledronic acid			Teriparatide		
Pamidronate			Hormone Replacement Therapy		

Others: .....

(Please refer clinic book, diagnosis card and BHT)

20. Do you take medications for osteoporosis?

- Yes, I take exactly as prescribed
- Yes, I take, but I forget sometimes
- Yes, I take, but I skip sometimes
- No, I don't take
- Don't know


21. If you don't take medication for osteoporosis, state the reason.

Side effects		Doctor suggested not to take it	
Too expensive		Forgot to take	
Already too many medications		Decided not to take any medication	
Not decided whether to take or not		Prescribed medication finished	

22. How long have you been on medication for osteoporosis?

Less than 6 months	
6-12 months	
More than 1 year	
Don't know	



## 23. Gender

Male	
Female	

## 24. What is your year of birth and age?

Year of birth	1	9			Age		
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## 25. Do you live

Alone	
With the spouse or a family members	
With the caregiver who is not a family member	
Living in elders' home	

## 26.

Weight	kg	Height	m	BMI	kgm <sup>-2</sup>
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## Article 6

# CARING FOR A CHILD WITH CLUB FOOT IMPACT ON FAMILY: A CROSS SECTIONAL STUDY

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**Key Words:** Clubfoot, Impact on family, financial burden, Coping strategies

## Abstract

### Background

Clubfoot is a treatable abnormality that can be managed with early interventions. Parental support is important for the compliance of the management and to achieve good outcome. Identifying and quantifying the family impact due to this condition is important when addressing the caregivers of these children. The aim of this study was to determine the financial, familial social impact, personal strain, and coping strategies of parents of a child with CTEV and explore any associated factors.

### Method

A descriptive cross-sectional study was carried out on 48 families presenting to the club foot clinic at Lady Ridgeway Hospital, Sri Lanka. A self-administered questionnaire with the Impact to Family Score and selected demographics was filled out by the selected participants.

### Results

Mean age of the children in the study sample was 21.4 months. 65.96% of the sample were families where only one parent was working and 64.6% of the total sample were living with extended families. The perceived mean total impact was 53.9 (SD=11.59) (range 34 to 78) and implying to be significantly lower than a USA study measuring impact for other chronic illnesses in children (M=48.03, SD=8.2,  $t(47)=3.53$ ,  $p<0.01$ ). Total impact in only one parent working (M=50.42, SD=9.80) implied to be significantly higher than both parents working (M=59.88, SD=12.51)  $t(45)=-2.87$ ,  $p<0.01$ .

### Conclusion

Except for personal strain domain, all other domains of the family impact are perceived less when caring for a child with clubfoot than when caring for a child with chronic medical illness. Instances with only one working parent, and those living with extended family were identified to have a higher perceived impact when compared to their counterpart groups. Coping strategies of the parents were more favourable within nuclear families. This can be taken as a baseline study which demonstrates caregiver and family burden when caring for a child with club foot.

## Introduction

Congenital talipes equinovarus (CTEV), commonly known as club foot is the most common congenital orthopaedic deformity encountered in the lower limbs [1]. Usually, the diagnosis is made soon after birth, but it can be detected by ultrasound scan from the third trimester of the gestation. This deformity affects males more than females and can occur unilaterally or bilaterally. Globally, prevalence of clubfoot is between 0.6 to 1.5 live births and about 80% of all the babies with clubfoot are from low- and middle-income countries [2]. When considering the South Asian region, clubfoot prevalence in India is 0.9 per 1000 live births [2] but no documented data on prevalence was available for in Sri Lanka. However, for Sri Lanka, The male to female ratio of clubfoot was identified to be 2.7:1, and bilateral deformity was seen in 48% of the cases[3].

Serial casting and manipulations followed by bracing, which was introduced and popularised by Dr Ignacio Ponseti, is the widely practiced treatment modality for clubfoot in the current practice [4]. This technique is based on addressing to the patho-anatomy of clubfoot. In Dr Ponseti's technique, the casts are numbered one to six and the first two casts are applied with the forefoot supinated, to bring it into alignment with the hind foot [5]. Then the third cast is applied using the head of the talus as the fulcrum point keeping simultaneous counterpressure over that with the forefoot abducted. In the fourth cast, the forefoot is further abducted. Usually, before the fifth cast, the degree of dorsiflexion is assessed and if dorsiflexion is not possible beyond neutral, then a percutaneous Achilles tenotomy is performed. The tenotomy, if required, is done under local anaesthesia as an outpatient procedure. However, in most Sri Lankan centres it is done under general anaesthesia in an operation theatre setting. The casts before the tenotomy are changed at weekly intervals while the cast after the tenotomy is removed at the end of three weeks. Following the removal of the last cast, irrespective of whether

a tenotomy is done or not, the patient is placed in a modified Foot Abduction Orthosis (FAO). This FAO is used for 23 h a day in the initial four to six months and subsequently during bed time up to three to four years of age[6]. The average number of casts with the Dr. Ponseti technique is only 5.4. According to Dr. Ponseti, the clubfoot usually recurs until four years of age and parents should be warned of this possibility [5].

In Sri Lanka, care for children with club foot is currently offered free of charge at all National hospitals, teaching hospitals and district general hospitals, where a Consultant Orthopaedic Surgeon is on duty. Orthopaedic unit 1 of Lady Ridgeway Hospital was the only specialized paediatric orthopaedic unit established in Sri Lanka up until 2019. Currently in this unit the Sri Lanka Clubfoot Program, in collaboration with the International Clubfoot Registry offers services for babies with clubfoot. It offers braces, continuous support, follow up and educational materials for the caregivers of babies with clubfoot free of charge.

Success rate of Ponseti casting rates 80 % - 94% in the current literature [7, 8]. However, the recurrence rate is around 20% and if not treated or neglected the foot will undergo complete equines and varus deformity hindering normal ambulation and requiring complex deformity correction surgeries with variable outcomes [9].

In this context, understanding of the caregivers about the long course of treatment, the nature of the treatment, the necessity to attend to the clubfoot clinic on exact given dates to replace casts and after serial casting, making the child wear the abduction foot orthosis for 23 hours a day for several months and essential night time wearing up until 3-4 years of age is utmost important to prevent recurrence of the disease and to get a better outcome. Nevertheless, this is a challenging task for the child and as well as for the caregivers.

Knowledge of any chronic ailment or a visible deformity of a new-born is distressing to any family. Caring for a child with such a condition can affect the family dynamics. Along the course, the special care that the family and parents should give to the child with clubfoot may affect the entire family. Certain aspects of this impact can be measured in terms of personal strain to the parents and caregivers, financial burden, effect on social interactions. This special context will eventually create unique coping strategies for the caregivers of the affected baby and the family.

Caregiver support is extremely important to achieve a successful outcome in the clubfoot treatment. If the family support is minimal, the child's condition can worsen, and it further imposes a negative impact on the family, and this may go on as a vicious cycle. Identifying the factors determining optimal family support and assessing the impact this condition has on the family can be considered a first step to plan methods of improving compliance. Suitable measures can then be tailor-made for the families affected by clubfoot. Furthermore, quantifying this impact into a numerical figure where we it can be compared with the impact caused by other diseases also allows better understanding and documentation.

The main objective of this study is to quantify the family burden when caring for a child with club foot and to quantify other aspects such as financial burden, familial and social burden, personal strain, and the coping/mastery mechanisms the parents or caregivers develop due to the child's health condition. The secondary objective is to explore any relationship between family burden and selected demographic factors.

## Methodology

A descriptive cross-sectional study was carried out on 50 families accompanying their children with clubfoot deformity to Orthopaedic unit 1 of Lady Ridgeway Hospital for children (LRH), Sri Lanka. The sample was randomly selected using every 3rd patient attending the

clinic. The clinic is held once a week and the sample of 50 was collected from June 2022 to August 2022. The questionnaire comprising the translated IFS scale and the demographic data extraction sheet was given to the families to complete by themselves during the waiting time and was collected just prior to leaving the clinic.

Impact to family scale (IFS) is a 27-item questionnaire with Likert scoring which measures the impact the child's health condition has on the family. This tool assesses financial hardship, work changes, travel, social interactions with friends, social interactions with family, family dynamics, finding caregivers, stigmatization, opportunity costs, worry, fatigue, depression, marital or family strife, grades, and school impact. These components are re-categorized to four areas of family impact. Namely, economic impact on family, social impact, familial impact, and personal strain/coping. All these areas together is considered the overall family impact. Lower scores indicate a greater negative impact or disability on the family.

This scale was originally developed to assess the family burden of chronic childhood illnesses by Prof. R. E. Stein and her team [10]. This scale was used in Paediatric Ambulatory Care Treatment Study in 1984, and this study is the hallmark of quantifying the family impact and other subscales using this study instrument.[11]

Mininder S. Kocher, et al (2022) used this for their study on Impact on Family Functioning of Immediate Spica Casting for Paediatric Femur Fractures: An Ecological Study, which revealed spica cast for femur fractures imposes greater overall impact on the family than taking care of a child with chronic medical illness. [12].

The original study instrument (IFS) was translated to Sinhala following standard protocols of forward and backward translation by sworn translators. It was finally checked by the original author and approved to be used in the study. The Sinhala version of the impact to family scale

questionnaire is yet to be validated (validation process underway by the same author of this article) to the Sri Lankan context.

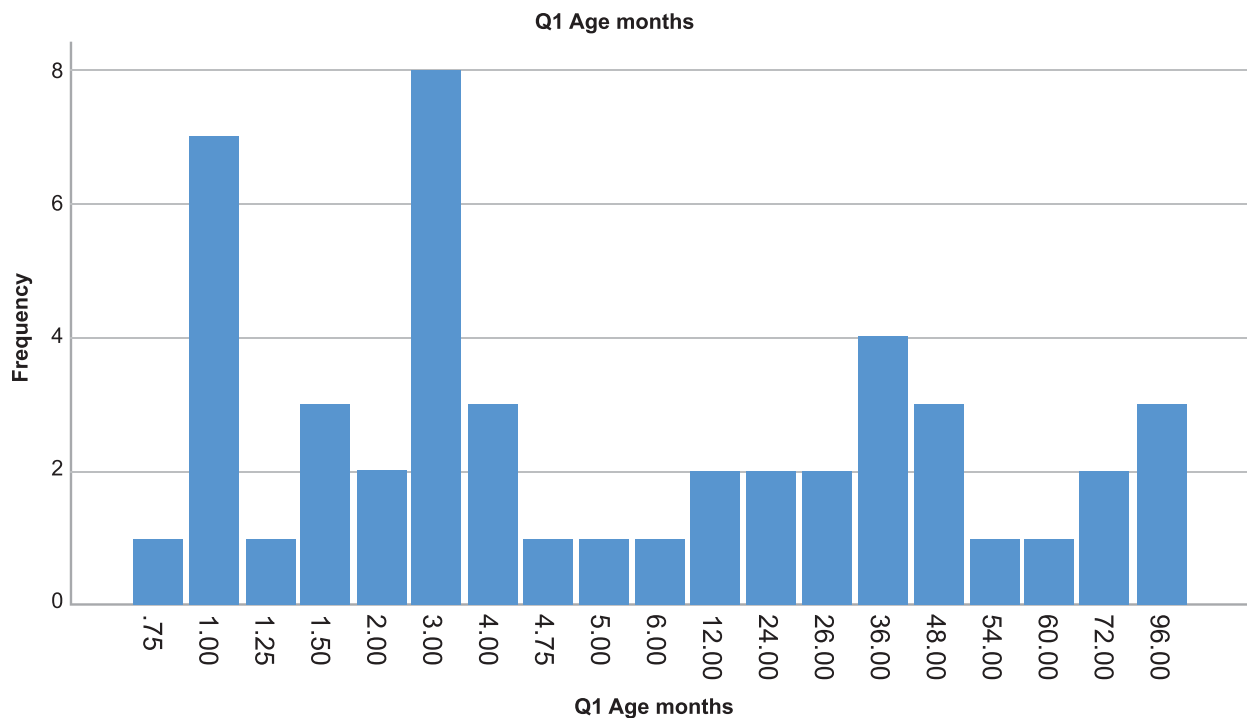
Statistical analysis was performed using SPSS 25.0. Overall mean scores and standard deviations were determined for overall scores and subscales. Normalcy of data was confirmed using the Shapiro Wilk test. Inter-group comparisons were made using the independent samples t-test and ANOVA or their nonparametric equivalents.

**Results**

**Cohort Characteristics**

Of the 50 families the questionnaire was given to only 48 families returned after proper completion (response rate: 96%).

Mean age of the child with clubfoot was 21.4 months (range, 3 weeks to 8 years). Figure 1 demonstrates the distribution which was not normally distributed ( $W(47) = 0.74$   $p < 0.001$ ).



**Figure 1 - Histogram showing distribution of age**

**Characteristics of illness**

According to Table 1, 91.6% (n=44) of the sample was diagnosed at birth, and only one was diagnosed at 3 years of age. The mean time from diagnosis of the illness to completion

of the survey was 4.26 years (51.17 months). However, the mean treatment duration was 1.58 years (18.99 months). None of the illness related statistics measured on a continuous scale were normally distributed as the Shapiro-Wilk statistic was significant in all cases.

**Table 1 - Descriptive statistics of the illness**

Characteristic	N (%)	Mean (SD)	Range	Shapiro Wilk statistic
Age of Diagnosis (days)	48 (100)	28.02 (184.12)	1-1277 days	0.13***
1	44 (91.7)			
7	2 (4.2)			
10	1 (2.1)			
1277	1 (2.1)			

Characteristic	N (%)	Mean (SD)	Range	Shapiro Wilk statistic
Laterality				
Unilateral	10 (20.83)			
Bilateral	38 (79.17)			
Recurrence				
Yes	6 (12.5)			
No	42 (87.5)			
No of episodes	48 (100)	1.21 (0.58)	1- 4	0.38***
1	41 (85.4)			
2	5 (10.4)			
3	1 (2.1)			
4	1 (2.1)			
Period of illness (months)	48 (100)	51.17 (66.36)	1.79 - 239.92	0.75***
Treatment duration (months)	48 (100)	18.99 (26.29)	0.03 - 96.00	0.73***
Defaulted treatment				
No	10 (20.8)			
Yes	9 (18.8)			
No data	29 (60.4)			
Cast Number				
1	6 (12.5)			
3	9 (18.8)			
4	7 (14.6)			
5	8 (16.7)			
6	3 (6.3)			
Brace	15 (31.3)			

\*\*\*p<0.001

### Characteristics of Family

The family characteristics of the sample are summarized in Table 2. One child is cared for by the grandparents since the mother is working abroad to support the family and the father is separated. This entry was removed during some inferential analysis in some further analyses and the total sample shows n=47. This entry was not included where the descriptive statistics of occupation are

shown. In all other families where only one parent was working it was always the father.

The maternal and paternal ages were normally distributed. Data was collected on other children of the family suffering with clubfoot. Of the seven families that had more than one child none of the other children had club foot. All parents in the sample had received secondary education or above.

**Table 2 - Characteristics of family**

Characteristic	N (%)	Mean (SD)	Range	Shapiro Wilk statistic
Mother's age	48 (100)	30.02 (4.76)	22- 44 years	0.96a
Mother's education				
Up to Grade 10	9 (18.8)			
Up to Advanced level	25 (52,1)			
Diploma and above	14 (29.2)			
Father's age	47 (100)	33.19 (4.98)	25 – 47 years	0.96a
Father's education				
Up to Grade 10	5 (10.4)			
Upto Advanced level	19 (39.6)			
Diploma and above	23 (47.9)			
Family Type				
Nuclear	17 (35.4)			
Extended	31 (64.6)			
Other children				
No	41 (85.4)			
1 other child	4 (8.3)			
2 other children	2 (4.2)			
3 other children	1 (2.1)			
Parental occupation				
Both parents working	16 (34.04)			
Only father working	31 (65.96)			

<sup>a</sup> – The Shapiro Wilk statistic was not statistically significant; hence these were normally distributed.

### Other demographics

The distance from the hometown to the hospital was less than 50km for 58.3% of the sample (n=28), and an equal number (n=10) belonged to each 50-100km and >100km categories.

Overall satisfaction of the family regarding the service received at Lady Ridgeway Hospital for Children was, very poor n=1, (2.1%), neither good bad n=1 (2.1%), good n=9, (18.8%), very good n=18 (37.5%), extremely satisfied n=19 (39.6%).

The main objective of this study was to quantify the family burden when caring for a

child with club foot and to quantify the subscales namely, financial burden, familial and social burden, personal strain, and the coping/mastery mechanisms the parents or caregivers develop due to the child's health condition.

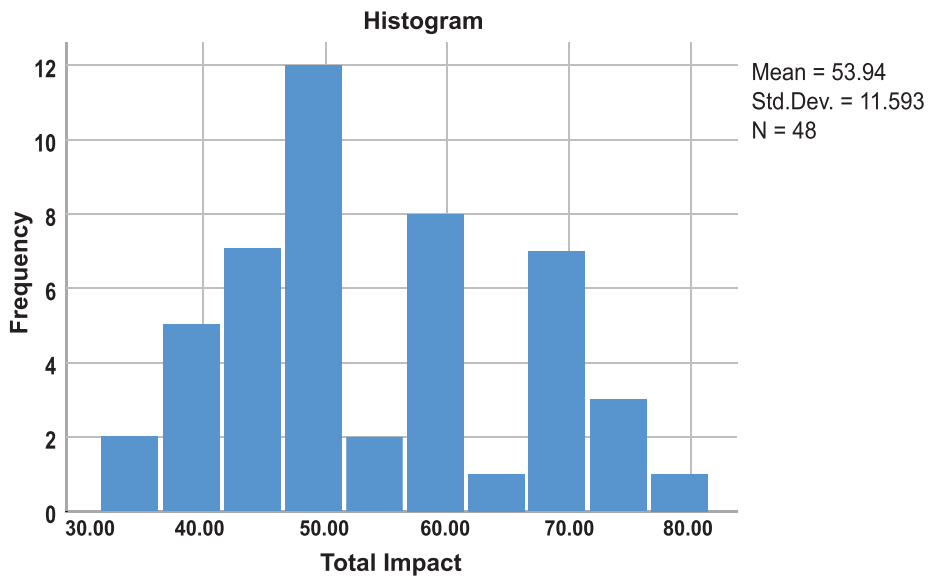
Table 3 shows the descriptive statistics of the 4 subscales of the IFS and the total IFS score. The main IFS scores and all the other sub scores except the Family and Social impact score, were normally distributed. The total mean score of 53.94 ( $\pm 11.59$ ) is significantly higher than the total mean score obtained in the PACTS study (M=48.3, SD = 8.2) ( $t(47) = 3.53, p < 0.01$ ).

**Table 3** - Descriptive statistics of the total IFS score and four subscales

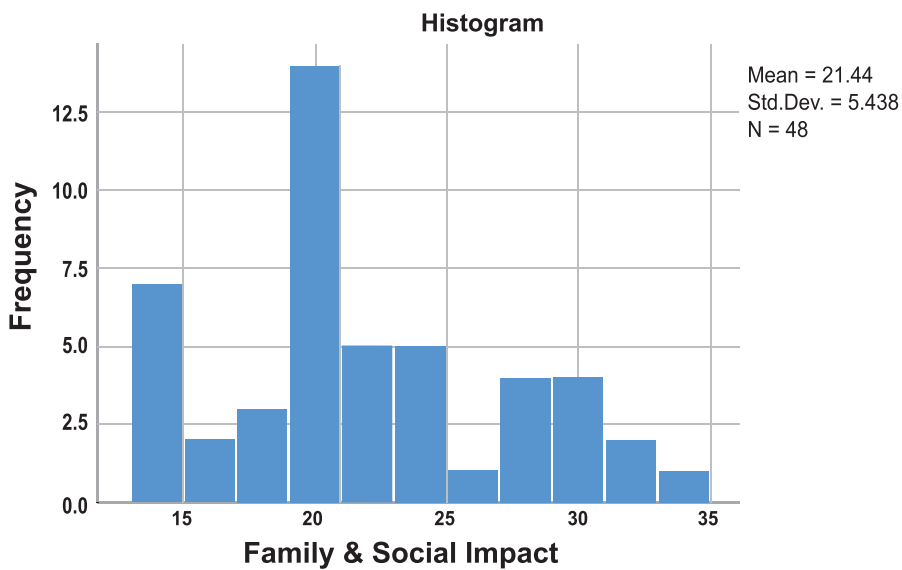
	N	Maximum possible score	Minimum	Maximum	Mean	Std. Deviation	Shapiro-Wilk (df=48)
Financial	48	16	4	16	9.23	3.08	0.97
Family & Social Impact	48	36	14	34	21.44	5.44	0.93*
Personal Strain	48	24	7	22	14.15	4.31	0.95
Coping/ Mastery	48	20	5	13	9.12	1.81	0.97
Total Impact	48	96	34	78	53.94	11.59	0.96

\*p<0.05

The histograms showing the distributions of the total IFS scores and the four subscales are displayed in figures 2 to 6.

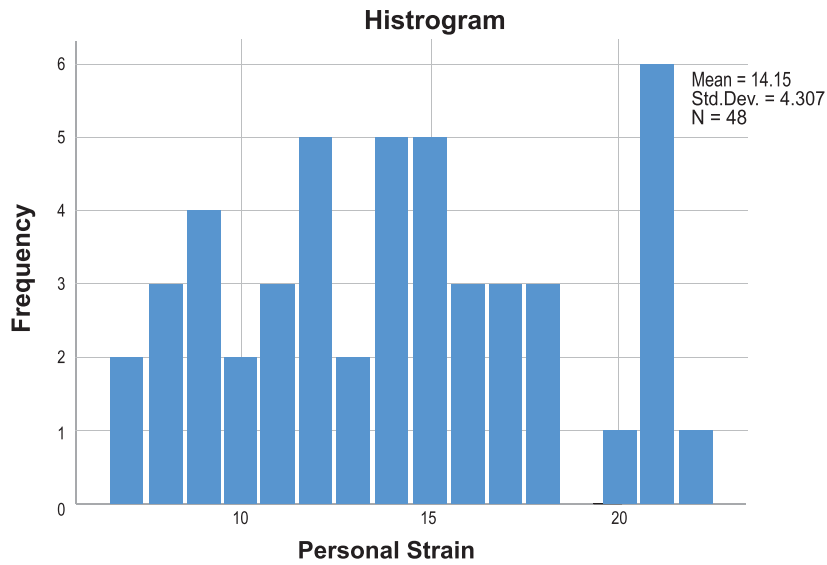


**Figure 2** - Histogram of Total IFS scores

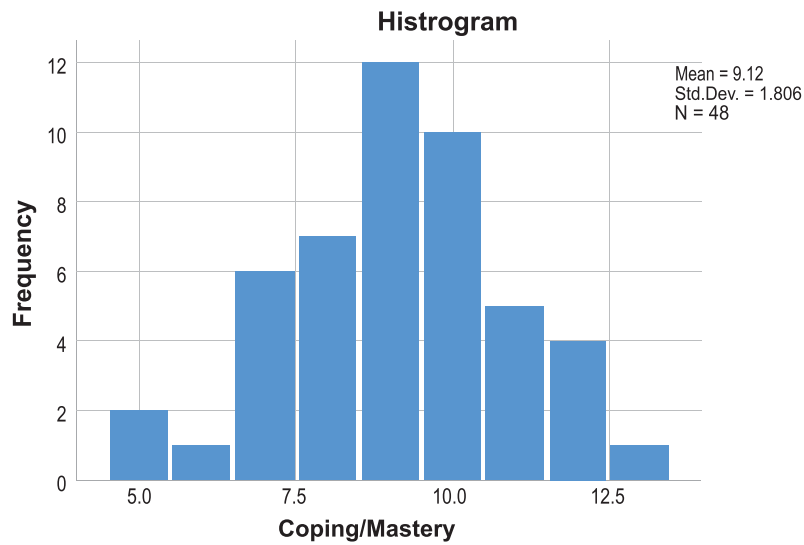


**Figure 3** - Histogram of Family and Social impact distribution

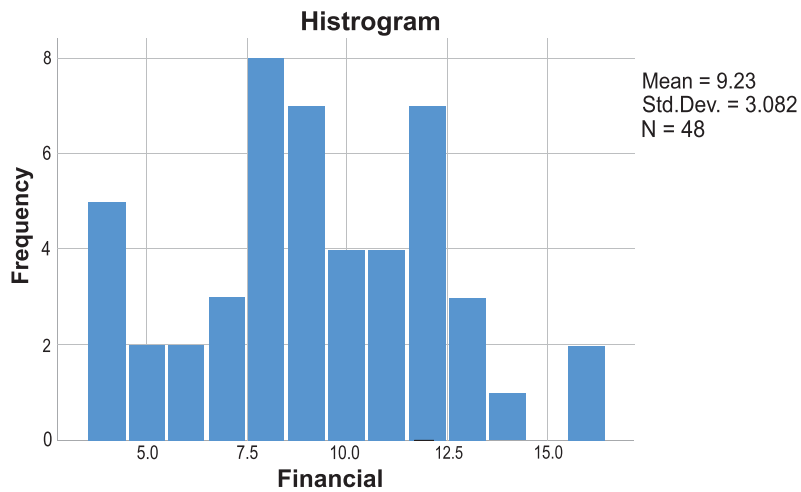




**Figure 4 - Histogram of Personal strain**



**Figure 5 - Histogram of coping/ mastery subscale**



**Figure 6 - Histogram of Financial subscale**

Table 4 compares all the subscales of the IFS score with the means obtained from the PACTS study. Only the personal strain in the study sample was found to be significantly lower than the mean of the PACTS study implying that the personal strain was higher in the study

population caring for a child with club foot. All other domains had mean values higher than that of the PACTS study, and the mean differences were significantly higher in all domains except in family and social impact domain.

**Table 4 - Comparison of means against the PACTS study**

	Sample Mean	PACTS scores mean (SD)	T (df=47)
Financial	9.23	7.7 (1.77)	3.44**
Family & Social Impact	21.44	20.8 (4.13)	0.81
Personal Strain	14.15	25.45 (4.83)	-18.18***
Coping/ Mastery	9.12	7.9 (1.55)	4.7***
Total Impact	53.94	48.03 (8.2)	3.53**

\*\*p<0.01  
\*\*\*p<0.001

## Objective 2

The secondary objective was to explore any relationship between this quantified burden and selected demographic factors.

*Analyses of associations between total IFS score and subscales with the continuous variables*

Only the maternal age showed a statistically significant weak negative correlation with the

financial subscale. ( $r=-0.29$ ,  $p<0.05$ ). This implies that with advancing maternal age the financial burden is perceived more.

No other illness related, or family related continuous variable showed a statistically significant association with the total IFS score or any of its subscales.

**Table 5 - Correlation Coefficient of the subscales and other demographics with scalar variables**

	Period of illness	Age months	Mother Age	Father Age	Siblings	No of episodes
Financial	-0.02	-0.04	-0.29*a	-0.23 a	-0.15	0.00
Family & Social Impact	-0.18	-0.18	-0.07	-0.08	0.04	0.13
Personal Strain	-0.05	-0.06	-0.10 a	-0.14 a	0.16	0.13
Coping/ Mastery	-0.08	-0.1	0.03 a	-0.12 a	-0.18	-0.13
Total Impact	-0.09	-0.10	-0.15 a	-0.19 a	0.00	0.11

\*p<0.05

<sup>a</sup>-Pearson Correlation was used since both corresponding scales were normally distributed

## Parental employment and impact to family

The entry filled by the grand parents in which both parents were deceased was removed from the analysis where the impact of parental employment was assessed.

According to Table 6 the total impact score in the group where only one parent is working is

significantly lower in the group where only one parent is working hence the perceived burden in this group is higher compared to the group where both parents are working. Same was observed in all domains but the difference was not significant in the personal strain and coping/ mastery domains.

**Table 6 - Mean differences of the burden among categories of parent occupation**

	<b>Only one working (N=31)</b>	<b>Both parents working (N=16)</b>	<b>Statistic of Mean difference (df=45)</b>
	Mean (SD)	Mean (SD)	
Financial	8.35 (2.37)	10.81 (3.75)	-2.749**
Family & Social Impact	20.06 (4.7)	23.75 (6.07)	148.5a*
Personal Strain	13.26 (4.04)	15.63 (4.53)	-1.83
Coping/ Mastery	8.74 (1.73)	9.69 (1.74)	-1.77
Total Impact	50.42 (9.70)	59.88 (12.51)	-2.87**

### Living with an extended family versus nuclear family

According to Table 7 the mean for coping/mastery domain within those in nuclear families (M=9.88,SD=1.17) is significantly higher than the mean coping/mastery domain of those in extended

families(M=8.71, SD=1.97). Equal variances were not assumed in the comparison of means for the two groups within this sub scale (F=5.89, p<0.05) t (45.64) = 2.59, p<0.05.

**Table 7 - Comparison of means of IFS scores between nuclear and extended families**

	<b>Nuclear (N=17) Mean (SD)</b>	<b>Extended (N=31) Mean (SD)</b>	<b>Statistic of Mean difference</b>
Financial	8.71(2.89)	9.52(3.19)	-0.87
Family & Social Impact	21.24(4.78)	21.55(5.84)	-0.19
Personal Strain	14.24(4.27)	14.10(4.40)	0.11
Coping/ Mastery	9.88(1.17)	8.71(1.97)	2.59*
Total Impact	54.06(10.80)	53.87(12.18)	0.05

\*p<0.05

There were no consistent significant differences in overall IFS scores or its domains among the subgroups of maternal education, paternal education, or distance from the residence to treating hospital.

### Discussion

In the management of paediatric orthopaedic conditions, the impact treatment has on the entire family is important to consider. Assessment of the family impact overall and the subgroups is important so that the interventions, we made can be tailor-made to each family and to a population.

Unlike in other orthopaedic conditions, like paediatric femur fractures [12] the child's age did not affect significantly over the impact in overall and in sub scales in our population. Hence, it can be assumed that the burden imposed on the family is independent of the child's age in cases of clubfoot.

Impact to family scale numbers is discussed in detail in the paediatric ambulatory care treatment study (PACTS) done in USA to assess the family impact when caring for children with chronic medical conditions such as asthma, diabetes, renal failure or juvenile rheumatoid arthritis. The total mean score in the PACTS study was (M=48.3, SD = 8.2) [11] was significantly lower than in our study (M= 53.94, SD ±11.59) (t(47) = 3.53, p<0.01). This shows that in the sample of parents taking care of a child with clubfoot and seeking treatment in the pioneer Centre in Sri Lanka, perceives less severe burden to the family when compared to USA sample caring for a child with a chronic illness. The reasons for this can be the availability of free healthcare facility in Sri Lanka and the predictable nature of the illness.

The other subscale values are considered as in table 4, Financial impact, family and social impact and coping domains shows greater scores

than PACTS study, showing in Sri Lankan context when caring for clubfoot children these aspects are not severely affected as in USA when chronic medical illnesses are considered.

However, in personal strain domain our sample mean 14.15 is significantly lower than PACTS mean ( $M=25.45$   $SD=4.83$ )  $t(47)=-18.18$ ,  $p<0.01$ ). This observation is important as it shows the caregivers personal burden is considerably higher. The reasons for this can be lack of teamwork within Sri Lankan family when caring for an ill child and the person, usually the mother of the family is expected to do all the household work and caring for the child on her own in Sri Lankan context. Although data was not directly collected on who filled in the questionnaire, it was almost always handed to the mother of the family. In USA, the cultural background is such that usually the family shares tasks hence one single person getting the burden all the time is minimal.

When considering coping/mastery domain, Sri Lankan families have adopted positive coping strategies. The cultural upbringing of acceptance of the illness and expecting positive outcome while giving continuous good care is seen in Sri Lankan context.

The financial domain shows significantly negative association with increasing maternal age. This means that the financial burden is felt more as the maternal age increases.

The children with both working parents (Figure 3) ( $n=16$ ), total impact mean = 59.88 ( $SD=12.51$ ) and the group with one working parent mean = 50.42 ( $SD=9.70$ )  $t(45)=-2.87$ ,  $p<0.01$ . This was statistically significant. This implies that when a single parent is working the family experiences a severe impact. The reasons could be the parent at home (mother, in our study sample) has to take up the whole burden of caring for the baby as when both parents are working, they have other support to care for the baby so that a "timeout" is received to parents while they are at work and the financial support to the family is more when both parents are working. This observation is backed by the finding that financial impact mean was 10.81 ( $SD=3.75$ ) significantly higher in the children with both working parents than in the group with one working

parent where the financial impact mean was 8.35 ( $SD=2.37$ )  $t(45) = -2.75$ ,  $P<0.01$ . Although not statistically significant, the positive coping strategies are seen when both parents are working than a single parent working.

We explored whether living with extended family, (with parents, siblings, in-laws,) has any difference in the perceived burden felt by the family when compared to living in a nuclear family. Overall impact was not significantly different, but it was noteworthy that in coping and mastery domain, nuclear families have better positive coping strategies than living with extended family. The reasons could be when living alone, the parent and child bond is stronger and each other shares the psychological stress while when living with an extended family, there may be added responsibility of the family towards the other family members and in the Sri Lankan context, shared support and expectations through arranged marriages may add to it[13].

## Limitations

Although the response from the parent/caretaker who accompanied the child to the clinic was recorded as a proxy for the entire family, he or she may not be the only respondent. If the response from both parents could be obtained the results would be more balanced with minimal disturbance to information from lack of communication between parents, unequal perception of the burden and other possible disturbances in the marital relationship.

## Conclusion

In conclusion the perceived impact to the family of a child with clubfoot lies at less than 50% of the maximum impact. However, a considerable impact is felt by the families of these children with clubfoot, and all its subscales. In comparison to studies in other countries on impact due to and other illnesses the perceived personal strain was higher, but the impact of all other aspects was lower in our study sample. Out of the sociodemographic variables lower maternal age and both parents being employed affected positively in some aspects of measured perceived burden, while coping strategies were significantly better when parents lived as nuclear families.

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## Case report 1

# Myositis ossificans of hip: A case report

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### Abstract

### Introduction:

Myositis ossificans is a benign condition in which there is an increased activity of periarticular tissues resulting in intramuscular bone formation. In most of the cases trauma is the leading etiology.

### Case presentation:

A 25 year old male with a history of pain and restricted range of motion of the left hip was admitted for further management. His hip motions were restricted with fixed flexion and abduction. There was a history of highway car accident with severe head injury and associated mid shaft comminuted fracture of left humerus. He was given ICU care for a period of two months. The imaging of left hip revealed mature myositis ossificans. The mass was removed surgically and post operatively the patient was rehabilitated with physiotherapy.

### Conclusion:

Myositis ossificans is a benign heterotrophic ossification of the muscle tissue. It has a prevalence of less than 1 per 1 million. Trauma is the most frequent etiological factor seen in almost 60 -75% of the cases. Non-traumatic myositis ossificans is very rare. Combination of surgical excision with radiotherapy in the treatment of the myositis ossificans may give satisfactory results.

**Key words:** Myositis ossificans, Heterotrophic ossification, Trauma Treatment of Undifferentiated IA

### Introduction:

Myositis ossificans is a benign condition in which there is an increased activity of periarticular tissues resulting in intramuscular bone formation. In most of the cases trauma is the leading etiology. It may affect any location in the body, which are susceptible to trauma. It commonly involves elbow, hip

and wrist. BIOPSY MAY BE REQUIRED IN SOME CASES FOR DIAGNOSIS. THIS PAPER PRESENTS A CASE OF TRAUMATIC HETEROTROPHIC OSSIFICATION OF hip.

### Case description:

A 25 year old male presented with a history of pain and restricted range of motion of left hip was admitted to ward. He had a history

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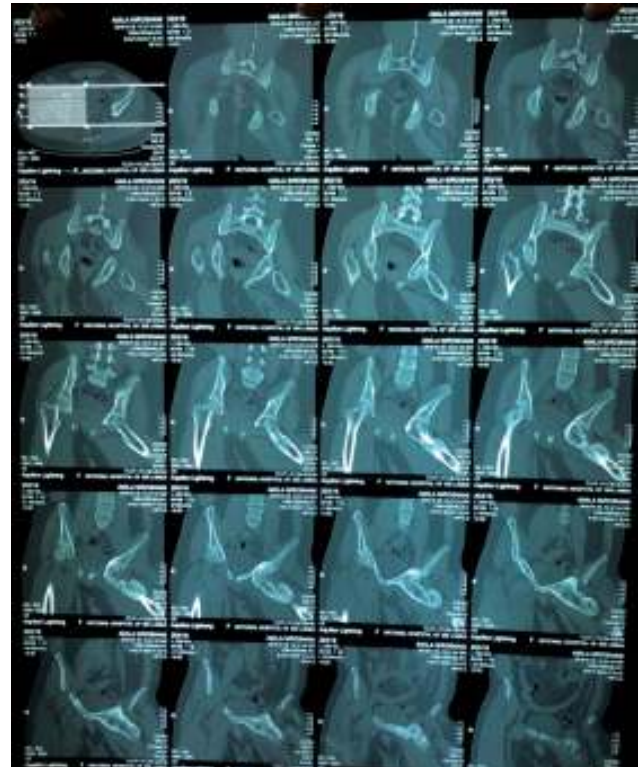
of highway car accident in 2016 march 16th. He was admitted to ICU for ventilation following a severe head injury with depressed skull fracture. He was given treatment in overseas for a period of two months and transferred to national hospital Colombo for further management. He had mid shaft comminuted fracture of left humerus. It was managed conservatively. On admission to national hospital Colombo, he had left hip fixed flexion and abduction deformity. He underwent CT scan of hip and followed by several manipulation under anesthesia was attempted and failed. Hip motion was restricted with fixed 200 of flexion, 150 of external rotation and 100 of abduction. He was able to walk without support with a wide base gait.

Radiographic evaluation of the patient with X-ray revealed a radiopaque mass extending from the anterior border of acetabulum and from the anterolateral part of obturator foramen to lesser trochanter of the femur (figure 1).



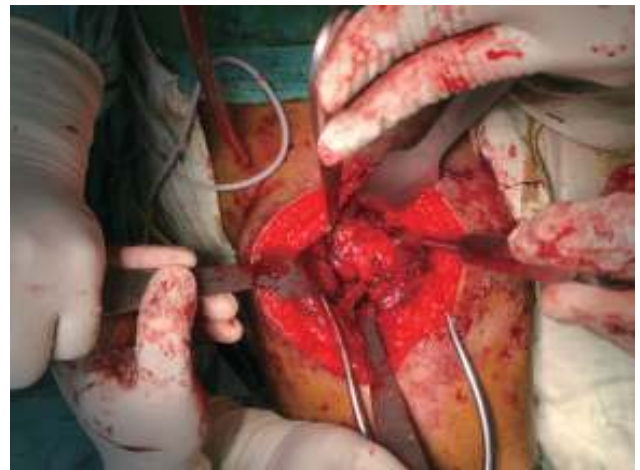
**Figure 1 - X Ray AP pelvis including bilateral hip**

CT scan revealed a mass which was bridging from anterior aspect of acetabulum to lesser trochanter with a large attachment (figure 2). The mass was osseous in character which was located along the iliopsoas muscles in its craniocaudal extension.



**Figure 2 - CT Scan of pelvis and bilateral hip**

The mass was removed surgically by using vertical limb of Smith-Petersen approach (figure 3). Intraoperatively, the mass was seen starting from the anterosuperior edge of the acetabulum and extending with a large attachment part to the anterior and medial aspect of femur on the lesser trochanter level. Furthermore, it was building an osseous bridge from anterior aspect of hip joint that was limiting the range of motion of the hip. After removal of the mass the range of motion of the hip was achieved (figure 4).



**Figure 3 - Excision of myositis - Intra operative image**

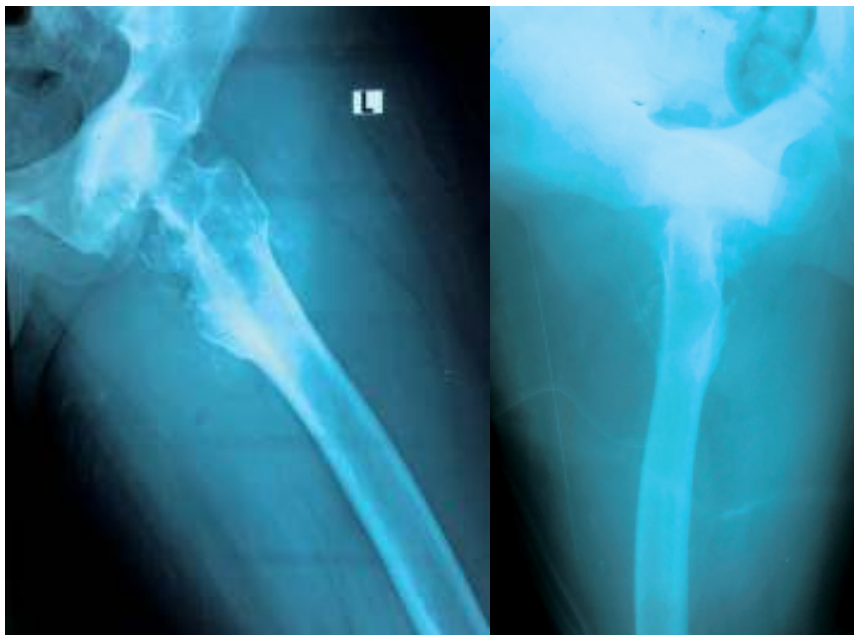
The biopsy of the lesion revealed mature bone tissue conforming our diagnosis of myositis ossificans. Post operatively the patient was treated with indomethazine (NSAID) and early physiotherapy. There was a decreased in the pain and increased in the range of motion post operatively.

**Discussion:**

Myositis ossificans is a benign and well differentiated bone formation of the muscle tissue. It has a prevalence of less than 1/1 million. There is no sexual predominance. Trauma is the most frequent etiological factor seen in almost 60–75% of the cases. It is believed that after a distinguishable trauma there occurs a tissue necrosis or bleeding initiating an uncontrolled vascular and fibroblastic activity resulting with bone formation. Although unproven, some other etiological mechanisms were also hypothesized. One of the theories claims osteoblasts that are freed from periost and trapped in the soft tissues as the provocateur of the myositis ossificans. The other

mechanism is the “ectopic calcification islands” theory which accuses periosteal tissue itself to be displaced into the soft tissues because of the impact of the trauma causing myositis ossificans. Tabes dorsalis, syringomyelia, poliomyelitis, paraplegia, tetanus, and hemophilia may play a role as the underlying pathology. In the presence of such conditions myositis ossificans may occur even passive range of motion exercises is carried out. Burns, infections, and drug abuse are other rare conditions which may cause myositis ossificans.

Nontraumatic myositis ossificans is very rare. Repetitive microtrauma, tissue ischemia, and inflammation were addressed as the causal mechanisms of the nontraumatic myositis ossificans. Myositis ossificans of the hip occurs more frequently in patients experiencing palsy, subdural or epidural bleeding, and hip operation. Fibrodysplasia ossificans progressive is another disease which presents with nontraumatic myositis ossificans.



**Figure 4 - Postoperative X Ray of left hip AP and cross table view**

The pattern of progression in myositis ossificans is pathognomonic by expressing a peripheral to central manner. Histologically collagen producing cells are located in the center and increased osteoblastic activity and immature bone lies in the intermediate zone and lamellar

bone in the peripherally.

Clinically there is a formation of a painful mass at the region of trauma within 7–10 days. Between 10 days and 6 weeks there appear to be irregular osseous fragments in this mass. Cortical



bone production takes place between 6 and 8 weeks. From 10 weeks to 6 months the typical egg shell appearance of central zone is visible. Maturation of the mass takes place between 6 and 8 months and the mass may shrink to some degree. Some lesions decrease in volume and some disappear within 1-2 years.

MRI findings demonstrate heterogeneity due to the histological structure of the myositis ossificans lesions. In the early period of the disease in T2 MRI section there is a dark and nonhomogenous intensity distribution in the central zone. The emergence of a hyperintensive ring around a hypointensive core is the sign of maturation of the mass.

Myositis ossificans is generally self-limited pathology. There is a possibility of the spontaneous regression; thus surgical excision is not the primary choice of treatment by most of the surgeons. Typical lesions may be followed with clinical and radiological observation. Surgical indications

include pain, increasing diameter of the mass, deteriorating local tendon or muscle function, and decreasing functional ability of the patient. Such lesions may be excised after maturation.

Radiotherapy may decrease the diameter of the mass and may increase the maturation of the mass. In the treatment of myositis ossificans, one low dose radiotherapy was performed in many cases and it was seen very effective. Gokkus et al. reported that 24 hours after operation one low dose radiotherapy was effective in their case. In another case report, Pakos et al. showed that radiotherapy treatment with combined indomethacin protocol was an effective treatment in myositis ossificans.

### Conclusion:

Myositis ossificans following trauma is common. Combination of surgical excision with radiotherapy, NSAIDS and physiotherapy in the treatment of the myositis ossificans may give satisfactory results.

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## Case report 2

# Chondrosarcoma of the Proximal femur treated by resection and Total Hip Arthroplasty.

### Introduction

Chondrosarcomas are the second most common primary malignant bone tumors after osteosarcoma, with heterogeneous clinical, radiological, and histological features(1). Chondrosarcoma is an aggressive malignant tumor of bone with pure hyaline cartilage differentiation. It can present in adults from the third to eight decades, peaking between 40 and 70 years of age, and men are affected more often than women(2).

The commoner central and peripheral types constitute the largest subgroups; rarer subtypes include mesenchymal, periosteal, clear cell, and de-differentiated chondrosarcomas. More than 90% of chondrosarcomas are primary [arising from previously normal bone] while secondary chondrosarcoma arises in pre-existing osteosarcoma including Ollier's disease or Maffucci's syndrome(1).

In this case report, we have presented our Sri Lankan experience in managing chondrosarcoma of the proximal femur with extensive wide local excision and Total hip arthroplasty using a diaphyseal fixation implant.

### Case presentation

A 47-year-old female housewife with no co-morbidities and no family history of cancer, presented with a large painful lump involving the right upper thigh and lower gluteal region for a six-month duration. It was progressively increasing in size for the initial two months, however, it shows rapid enlargement in the last four months. She had night pain but he denied any history of trauma, fever, numbness, and weakness of the lower limb.

Physical examination revealed a large elongated [29cm x 16cm] lump without any ulceration in the overlying skin and firm consistency.



**Figure 4 - Shows lump involving in the right gluteal region and upper thigh.**

Biochemical and hematological investigations were within normal limits except ESR 95mm/hr. Her Xray of bilateral hip and proximal femur

anteroposterior view revealed a large, intraosseous, osteolytic lesion with a narrow zone of transition and irregular granular calcifications

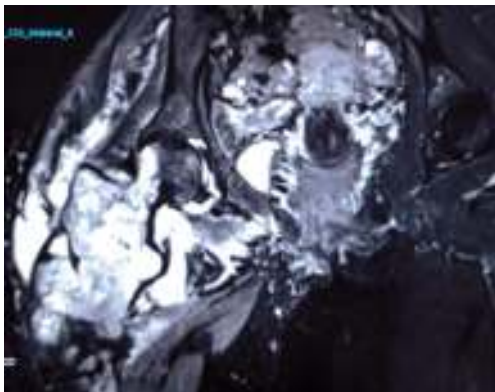
within the matrix described as honeycomb or popcorn sign [fig.2.] contrast-enhanced computer tomography [CECT] of chest, abdomen, and pelvis were not revealed any abnormalities.

CECT of the right thigh revealed destructive bony lesion involved right proximal femoral head, greater trochanter, neck, and proximal shaft. The bony lesion is associated with multiple septated solid and cystic soft tissue mass measured 28cm x 15.5cm x 10cm [fig.3].

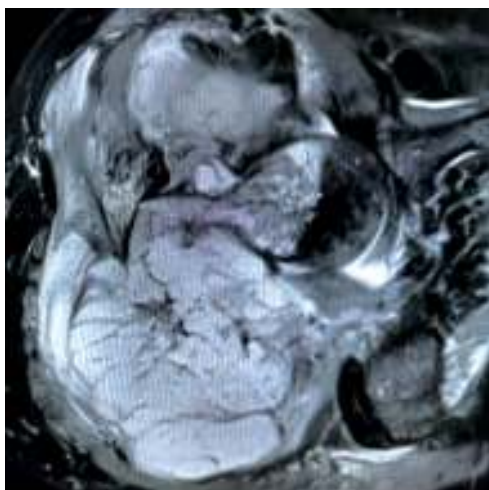
Magnetic resonance image [MRI] of the right thigh revealed a large bony destructive bony lesion arising from the right femur head, neck, and proximal shaft. The tumor is not involved the acetabulum of the pelvis. The tumor extending up to 16cm from the head of the femur. The vascular compartment is displaced anteromedially however no vascular invasion or encasement. [fig.3]



**Fig. 2.** : Shows an anteroposterior view of the bilateral hip joint and proximal femur radiograph, which revealed intraosseous, osteolytic lesion with a narrow zone of transition and irregular granular calcifications within the matrix described as honeycomb or popcorn sign in the right proximal femur.



**Fig 3.** : Shows MRI of right thigh coronal section



**Fig 4.** : Shows MRI of right thigh axial section shows tumor involved the soft tissue around the femur shaft and proximal femur.

Incision biopsy was arranged following a discussion with a multi-disciplinary team, under subarachnoid block. It shows grade 1 chondrosarcoma.

The treatment opted for this patient was wide local excision and total hip arthroplasty using a diaphyseal fixation implant. The indexed surgery was performed by an experienced orthopedic surgeon. Wide local excision of the malignant tumor and the hip reconstruction were carried out in a single session.

The procedure was explained and informed written consent was obtained. She underwent routine preoperative assessment. The patient was given a subarachnoid block and she was placed in the left lateral decubitus position. The posterior approach was used with a long curvilinear incision that reached the soft tissue tumor part. Meticulous soft tissue dissection made, and soft tissue tumor part entered the vastus intermedius and vastus lateralis muscles were resected out owing to tumor infiltration. The muscles that are attached to the proximal femur and midshaft periosteum

are resected out. Wide local excision of tumor performed by dislocation of the femoral head and femur shaft resection at 21cm from the head of the femur according to the preoperative templating.

The acetabulum was prepared routinely to fix the acetabular component. The femoral canal was prepared by reaming to insert the long Wagner stem (Wagner SL revision® hip system, 265mm × and additionally supported by cerclage wires. After the reduction of the femoral component, stability was checked and routine closure done drain kept in.

Postoperative antibiotics and prophylaxis of deep vein thrombosis were given and the intravenous antibiotics were continued for one week postoperatively.

The patient was mobilized in the bed on the first day of post-op. After five days the patient was instructed to touch toe weight-bearing with direct supervision and care of a physiotherapist. The patient was discharged successfully on the 10th day of post-op.



**Figure 5:** shows the intra operative picture of the tumor during excision



**Figure 6:** shows the intra operative view following the wide local excision



**Figure 7:** shows the intra operative view following the hip reconstruction



**Figure 8:** shows the excised tumor with involved soft tissue



**Fig:9 :** shows postoperative anteroposterior radiograph and lateral radiograph of femoral and acetabular component of hip reconstruction.

## Discussion.

Chondrosarcoma is a pure hyaline cartilaginous origin rare malignant tumor of the bone. Chondrosarcoma shows a heterogeneous group of lesions and a wide range of morphological and clinical behavior.

The chondrosarcoma can be classified according to histological type, origin, and site.

Histological:- conventional, clear cell, mesenchymal, dedifferentiated

Origin:- primary or secondary

Site:- central or appendicular

Macroscopic features of chondrosarcoma of a cut section are translucent, bluish-grey glistening cut surfaces with a lobular pattern. There will be areas of myxoid or mucoid material and cystic degenerations and some amounts of calcium deposition. The histology shows lobules of hyaline cartilage are separated by fibrous septae, which shows a permeative growth involving lamellar bone indicative of a rapidly growing malignant tumor. This permeative growth pattern is the differential factor between chondroma and chondrosarcoma(1).

The lobules of chondrosarcoma contain hypercellular, atypical, and hyperchromatic chondrocytes. Necrosis and mitoses can be seen in high-grade chondrosarcoma.

Chondrosarcoma can be divided into grades histologically based on the factors of cellularity, cytological atypia, chromasia, and mitotic features.

Grade 1:- (low-grade) are moderately cellular and nuclei are hyperchromatic and abundant chondroid matrix

Grade 11:- more cellular and nuclear atypia and hyperchromasia, mitoses are present.

Grade 111:- ( high-grade ) hypercellular with nuclear pleomorphism and frequent mitoses.

The histological grading has good guidance for best treatment and clear prognostic value. The survival rate was 89% for Grade 1, 53 % for Grade 11, 38% for Grade 111(3).

The management of chondrosarcoma is a multidisciplinary approach that involves orthopedic surgeons, a radiologist, and Histopathologist. The primary option of the treatment is surgical resection; radiotherapy and chemotherapy have not shown any benefit except for palliative purposes.

After the radiological investigation such as contrast-enhanced computer tomography and magnetic resonance image, the biopsy should be planned to be performed in line with a subsequent resection approach, because these lesions have a definitive potential of recurring in a biopsy track.

The treatment of low-grade tumors depends on the location. The low-grade appendicular tumors can be treated by aggressive curettage plus or minus cement or cryotherapy.

The treatment of grade 11 and grade 111 tumors should undergo surgical resection with wide surgical margins. The intralesional curettage is not recommended because of high rates of local recurrence and metastasis.

The chondrosarcomas are radio and chemo-resistant. Recent studies show the Alendronate has shown anti-proliferative effects on cartilaginous cells(4). Carbon ion therapy has been reserved for non-operable cases or difficult locations. The carbon ion therapy has a high cell mortality rate than photon therapy for the same physical total dose given(5).The recent study shows the effect of selective cox-2 inhibition on chondrosarcoma growth(6).

In this patient, even though the tumor is grade 1, it shows large soft tissue involvement and cortical bone involvement. So we decided to go for wide local excision of the tumor and followed by hip joint reconstruction.

**Conclusion.**

Chondrosarcoma is an aggressive malignant tumor. For many decades, disarticulation of the hip joint was the method of choice for proximal femoral tumors. A great deal of attention has focused on malignant bone tumors. Nowadays proximal femoral tumor resection with total hip arthroplasty with long femoral stem or endoprosthetic replacement is the most commonly used treatment method.

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**Data availability.**

The data used to support the findings of this case study are included in this article.

**Consent.**

Informed written consent was obtained from the patient for publication.

**Conflict of interest.**

All authors declare that they have no conflicts of interest.



### CASE REPORT 3

## **Sacral dome resection and single-stage posterior reduction in the treatment of high-grade spondylolisthesis in a young girl.**

### ABSTRACT

#### Introduction:

The treatment of high-grade spondylolisthesis remains controversial in terms of in situ fusion versus reduction. The aim of operation in this patient is to restore the spino-pelvic alignment, sagittal profile of the spine and prevention of progression of slip with a minimal neurological risk.

#### Case presentation:

18 years old schooling girl referred with a history of backache and right lower limb numbness over lateral aspect of foot for four years duration. She also had intermittent lower back pain, produced by long distant walking and relived by rest. It was aggravated by standing for long duration. There was no radiation of pain, no red flag sign or no cauda equina symptoms. Apart from her right lower limb neurology which was her power MRC grade IV in L5 and S1 myotome, other examinations were normal. X-Ray revealed grade V spondyloptosis. MRI showed evidence of right exiting L5 nerve root entrapment with grade V spondyloptosis. She underwent single stage posterior instrumentation of L4 - S1 vertebrae, sacral dome resection, reduction of L5 over S1, interbody fusion of L5 - S1 and posterolateral intertransverse fusion of L4 to S1 vertebrae. She was ambulated on post-operative day one without brace. There was no worsening of neurology.

#### Conclusion:

Lumbosacral spondylolisthesis grade V can be treated using posterior approach alone to obtain reduction, decompression, and solid fusion. The sacral dome resection is a shortening osteotomy of the lumbosacral spine which allows a single-stage reduction of L5 without lengthening of lumbosacral region in high-grade spondylolisthesis, which helps to avoid neurological complications.

**Key words:** High grade spondylolisthesis, Sacral dome resection, Posterior reduction, Spino-pelvic alignment

#### Introduction:

The treatment of high-grade spondylolisthesis remains controversial in terms of in situ fusion versus reduction. While satisfactory

clinical outcome has been reported after in situ fusion, this procedure is associated with higher rates of pseudarthrosis and slip progression. Without reduction the lumbosacral alignment does not improve. The aim

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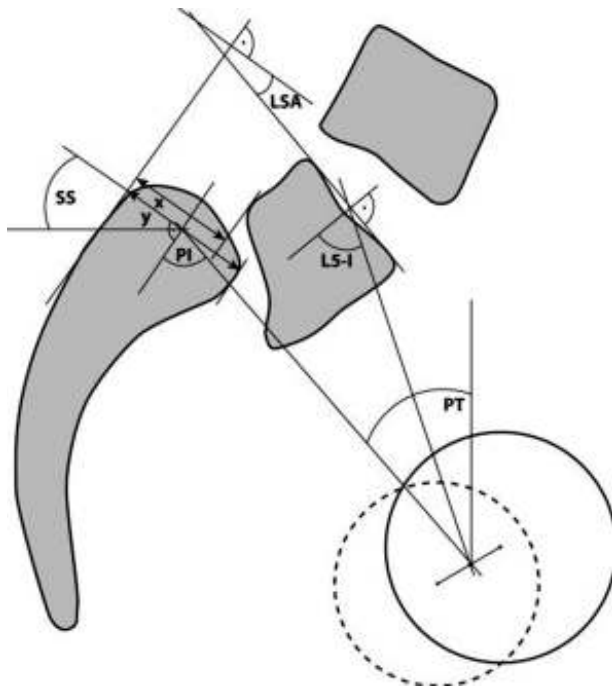
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of operation in this patient is to restore the spino-pelvic alignment, sagittal profile of the spine and prevention of progression of slip with a minimal neurological risk.

Reduction of the slipped L5 over S1 in high-grade spondylolisthesis places the L5 nerve root under tension which can lead to neurological complications. The reduction restores the segmental lordosis, improves

lumbosacral alignment and therefore the overall sagittal profile of the spine. Pelvic morphology and spino-pelvic alignment are abnormal in high-grade spondylolisthesis. While pelvic incidence (PI) remains constant as a morphologic descriptor, surgical reduction of L5 over S1 can improve lumbosacral and spino-pelvic alignment as reflected by changes in pelvic tilt (PT) and sacral slope (SS).



**Fig. 1** L5 Slip is  $x/y$  in percent, L5 incidence (L5-I), lumbosacral angle (LSA), pelvic incidence (PI), pelvic tilt (PT), sacral slope (SS)

The severity of spondylolisthesis is measured as percentage of forward slip of L5 over S1. Lumbar lordosis (LL) is the Cobb angle from the superior endplates of L1–L5. L5 incidence (L5-I) is the angle between a perpendicular line to the L5 superior endplate and a line joining the center of the bicoxo-femoral axis and the center of the superior endplate of L5. The LSA or slip angle is the angle between the lines on the superior endplates of L5 and S1. Pelvic incidence is the angle between a line connecting the centre of the upper endplate of S1 to the bicoxo-femoral axis and a line perpendicular to the end plate of S1. Pelvic tilt is the angle between a vertical line and a line connecting the centre of the upper endplate of S1 to the bicoxo-femoral axis, and SS is the angle between a horizontal line and the endplate of S1.

### Case description:

18 years old schooling girl referred to clinic with a history of backache and right lower limb numbness over lateral aspect of foot for last four years duration. It was a lower back intermittent pain, produced by long distant walking and relieved by rest. Standing for long duration induced pain in her lower back. There was no radiation of pain to groin or legs. Gradually claudication distant reduced. She was able to walk as usual. She was able to climb stairs up and down without pain. There was no alteration in bladder bowel function. There was no neck or upper limb pain. She denied any history of trauma, recent weight loss, nocturnal fever, contact history of tuberculosis, history of cancer or steroid use.

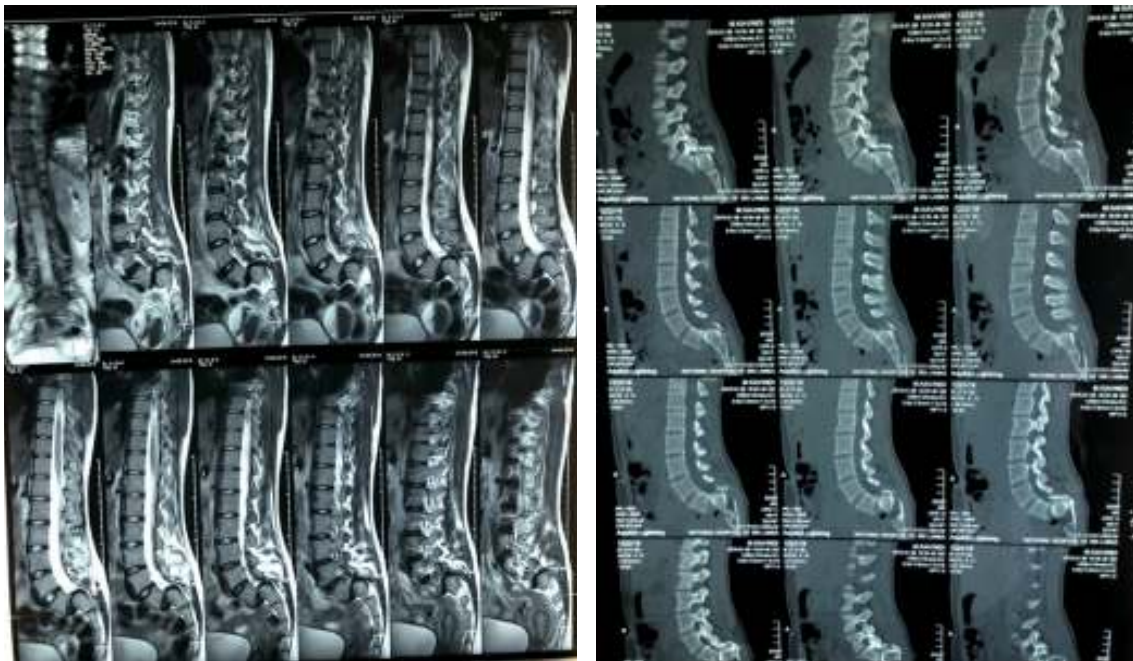
She walked without pain. Her shoulder and pelvis stayed in symmetrical position. There were no

leg length discrepancies. Her bilateral straight leg raise test was negative. She had right lower limb power MRC grade IV in L5 and S1 myotome. But her left lower limb power was MRC grade V. There was no sensory alteration in both lower legs. Her perianal sensation and peripheral pulse were intact.

X-Radiograph showed grade V spondyloptosis (Fig 2.). Followed by she underwent an MRI scan to assess her cord and nerve root status. It shows there was right L5 exiting nerve root entrapment with grade V spondyloptosis (Fig 3.).



**Fig 2. :** X-Ray lumbosacral lateral & AP view in standing position



**Fig. 3. :** MRI scan

### **Surgical technique:**

L4–S2 region was exposed from the posterior midline. Pedicle screws were inserted in L4, L5 and S1.(Fig4.) S1 pedicle screws were placed in a more caudal position to leave room for the sacral dome osteotomy and resection. Both S1 pedicle screws were placed to the anterior cortex for bicortical purchase after visualization of S1 existing nerve root. A complete removal of lamina L5, flavectomy of L5/S1 were performed. L5 roots were thoroughly decompressed in the isthmus region by removing bony callus and granulation tissues of the spondylolysis. The L5 roots were exposed laterally until exiting from the foramen. The cranial part of the ala of sacrum was excised to release the L5 roots from tension completely. The annulus fibrosus in high-grade spondylolisthesis had a bulging part in the foramen below the existing L5 roots and the roots found to be under tension. Care was taken to remove this bulging part far laterally under the L5 roots. The L5/S1 disc was exposed bilaterally between the S1 and L5 roots and excised. Temporary connecting rods were used with minimal distraction. The osteotomy of the sacral dome was performed from both sides in an antero-medial direction using ordinary straight osteotomes, after which the upper part of the sacrum together with attached disc fragments were removed in piece meal. Anterior lip of the lower plate of the L5 vertebra body osteotomised and excised through the disc space to remodel the trapezoid shape of L5 body. Position of osteotome and osteotomy sites were confirmed with image intensifier. A lateral fluoroscopy or a lateral radiograph was helpful to make sure that the extent of the osteotomy was adequate. During this procedure, the segment L5/S1 gradually became mobile. The rods were contoured in lordosis and firmly fixed to the S1 screws first. The L4 and L5 screws were sequentially reduced to the fixed rods, reducing the slipped L5 on to the osteotomised surface of S1. L5 roots were continuously visualized to make sure that they were not stretched. When adequate amount of sacral dome was resected, the reduction was possible without lengthening of L5–S1 and without tension on the L5 roots. The amount of slip reduction was determined by the development of tension in the L5 roots. It was not necessary to aim

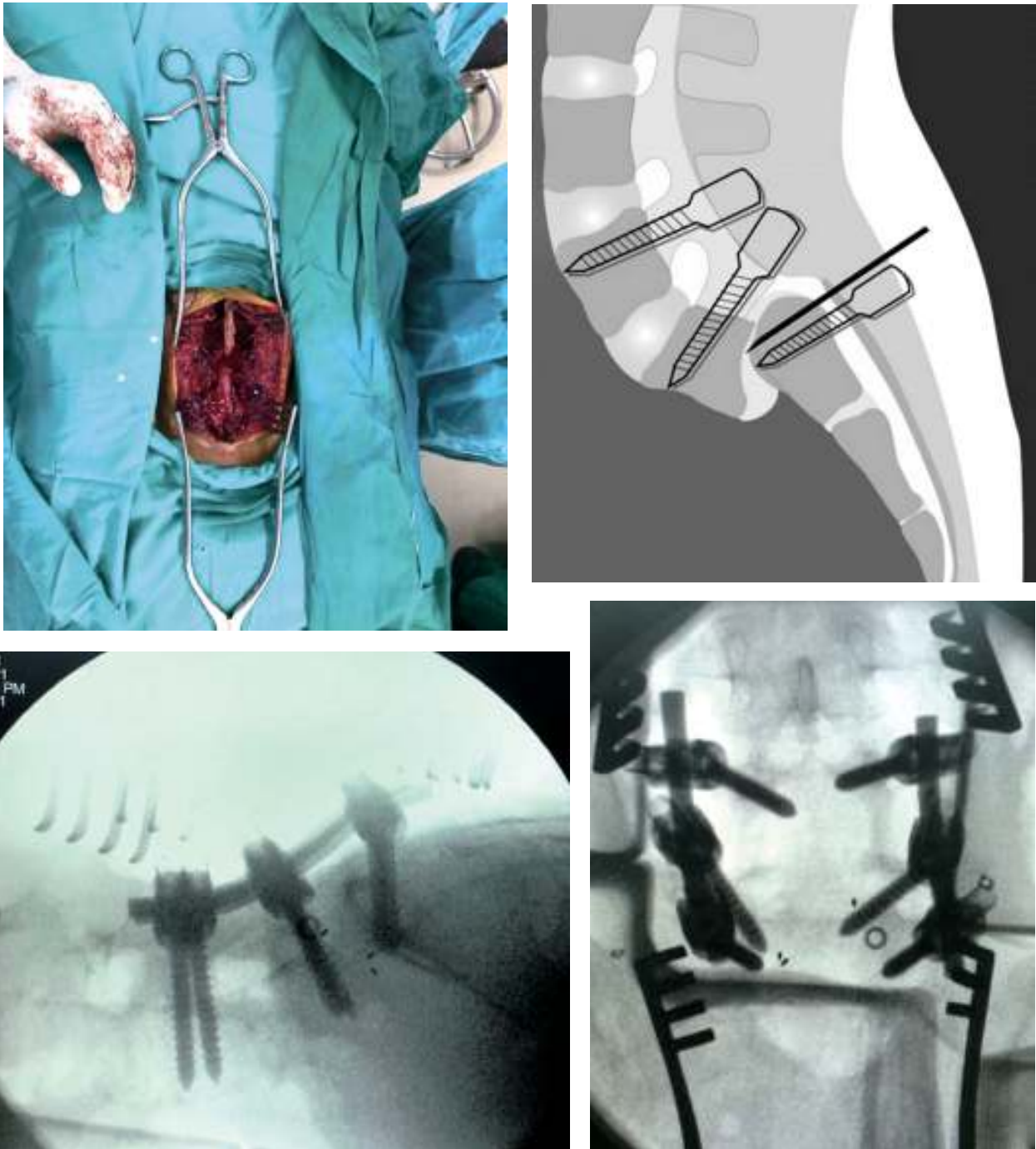
for full reduction. Correction of the lumbosacral kyphosis and a good L5 nerve root decompression were more important than a full slip reduction. The sacral dome resection was a shortening osteotomy of the lumbosacral junction and any maneuver causing lengthening of the lumbosacral junction was avoided during the whole procedure. The disc space between the end plates of L5 and S1 was cleared fully, removing all cartilages. The space was supported by PEEK cage filled with autologous graft in order to achieve 3600 fusions. Prior to insertion of cage, autologous bone graft was placed to partly fill the anterior disc space. Subsequently the position of the cage was checked by direct vision to prevent impingement of nerve root and dura. The cage position was further confirmed using image intensifier, by conforming the position of radio-opaque markers. Posterolateral intertransverse fusion of L4–S1 was done using bone graft to achieve bony fusion.

It took around two and half hours to complete whole procedure. There was around 500ml to 600ml of blood loss during the procedure. Within this 600 ml blood loss, approximately only 100ml to 200ml loss due to osteotomy. The procedure can also be performed with neuromonitoring if facilities permit.

Ambulation of the patients began on the second postoperative day. No braces were worn. Postoperative neurology was remained as preoperative without any worsening or much improvement. Up to now, there was no pseudarthrosis or implant failure.

### **Radiographic parameters:**

The preoperative standing radiograph of lumbosacral spine showed slip percentage of 90%, L5 incidence 820, lumbosacral angle -370 indicating a kyphotic deformity, pelvic incidence 760, pelvic tilt 380 and sacral slop of 380. Post-operative standing radiograph of lumbosacral spine showed slip percentage 15%, L5 incidence 480, lumbosacral angle 60, indicating a reversal of kyphotic deformity to lordosis. The pelvic incidence was not changed and remained as 760. But pelvic tilt improved to 400 and sacral slop improved to 360.



**Fig 4 :** Intraoperative image after screw insertion before rod application (Upper left), performing osteotomy of sacral dome (Upper right), after reduction and rod application II view lateral (Lower left) and antero-posterior (Lower right).

**Discussion:**

The best way to treat a high-grade spondylolisthesis is to correct the multidirectional deformity of lumbosacral junction with minimal neurological risks. Even though there are conflicting reports about the in situ fusion for high-grade spondylolisthesis, the instrumented fusion with reduction has a clear advantage

like facilitation of full nerve decompression, promotion of bony union, restoration of body posture and mechanics, as well as improvement of appearance.

The reduction procedure is known to be associated with neurological complications. There are various descriptions of reduction from posterior alone or anterior posterior

combined procedures. The aim of the surgery is to decompress the spinal canal and nerve roots, as well as to improve the lumbosacral deformity. The reduction of a severely slipped L5 is usually associated with elongation of the lumbosacral junction. Bohlman and Cook first described the removal of the upper corner of the S1 vertebral body to decompress the nerve roots in a surgical procedure where the reduction was not undertaken. Gaines and Nichols described an extensive anteroposterior procedure for L5 vertebrectomy and reduction from L4 on to S1 in the treatment of spondyloptosis, which was a procedure of shortening of the lumbosacral junction.

The sacral dome is excised from posterior approach to produce shortening in this region. In addition to this, the sacral dome resection results in complete mobilization of the L5/S1 segment, facilitates complete L5 nerve root release laterally.

Instrumented fusion from L4 to S1 has advantages over monosegmental L5/S1 fusion. Firstly, screw purchase in severely dysplastic L5 pedicles may be weak and unreliable and secondly, the L4/5 facet joints are usually abnormal in severely dysplastic high-grade spondylolisthesis. We recommend an instrumented fusion from L4 to S1 to avoid loss of correction and sacral bending, as well as development of spondylolisthesis of L4.

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Sacral dome excision and reduction produce ample bony surfaces between the bodies of L5 and S1 for anterior column fusion. In L5/S1 segment, the interbody fusion is important and able to achieve good anterior column fusion without an additional anterior procedure. Posterolateral fusion is done between the transverse processes of L4–S1 vertebrae.

The posterior alone approach with shortening sacral dome resection, single-stage reduction and pedicle-screw fixation from L4 to S1 allowed the restoration of spino-pelvic alignment towards more physiological values, with minimal risks for neurological injury.

### Conclusion:

We conclude that sacral dome resection from posterior approach in high-grade spondylolisthesis is a shortening osteotomy of the lumbosacral junction. It is very useful for single-stage posterior reduction of L5–S1 with the use of pedicle screws avoiding lengthening of lumbosacral junction and avoiding additional anterior surgery. This procedure followed by the instrumented fusion of L4–S1 produces a good multidimensional deformity correction with a minimal risk of neurological injury and a satisfactory clinical outcome. This is a safe surgical procedure to restore spino-pelvic alignment and the sagittal profile of the spine in the treatment of high-grade spondylolisthesis.

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## CASE REPORT 4

# Osteoblastoma of the first thoracic spine treated with corpectomy and fusion: A rare case report

## ABSTRACT

**Keywords** - Osteoblastoma, Osteoid osteoma, First thoracic spine, Corpectomy, Upper back pain.

## Abstract

**Introduction and importance:** Thoracic spine involvement of the osteoblastoma is a rare occurrence. A multidisciplinary approach will provide a good outcome.

**Case presentation:** We present a 16-year-old patient with a first thoracic spine managed surgically.

**Clinical discussion:** A 16-year-old schoolboy presented with a history of dull aching type neck pain which progressively worsens over the last four months without any neurological weakness. Radiographic studies of the spine show an expansile well-corticated lesion with central radiolucent nidus with bony sclerosis in the first thoracic spine which is suggestive of osteoblastoma. A corpectomy with a tricortical iliac graft fusion and anterior instrument stabilization was achieved by a multidisciplinary team. The patient is pain-free and a full range of neck movement is achieved.

**Conclusion:** osteoblastoma of the spine can be treated surgically with a low risk of recurrence successfully. Torrential bleeding during the surgery is a devastating complication. Preoperative radiological embolization and Intraoperative navigations are the available options to overcome it. A combined multidisciplinary team approach will provide a good outcome.

## Introduction

Osteoblastoma is a rare osteoid-producing primary bone tumor with a predilection for the long bone(1). Jaffe apud Samdani et al first describe osteoblastoma and osteoid osteoma in 1935(2). Incidence of spinal osteoblastoma was noted in the literature as 36% with a high prevalence in the cervical and lumbar region(1)(3). Clinical presentation varies according to the location of the tumor; however, dull aching type night pain is common. Surgical excision and stabilization

of the spine will give an excellent outcome and return of normal life to the patient. We shared our Sri Lankan experience of managing a patient with osteoblastoma of first thoracic spine osteoblastoma with a corpectomy and fusion with an iliac bone graft.

## Case

A 16year old previously healthy schoolboy presented with a history of a dull aching type of pain in the lower cervical and upper thoracic region which was difficult

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to localize by himself for six months duration. The pain gradually got worsen over the last four months which it was not responding to the analgesics. He had on and off right-side hand numbness but no weakness. He had strongly denied any history of trauma and family history of malignancy. He had midline tenderness at the lower cervical spine region and a global restriction of neck movement. His detailed neurological examination and systemic examination were completely normal.

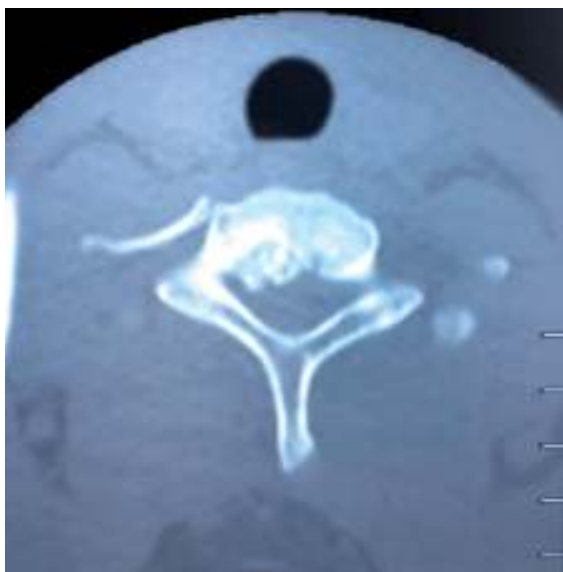
His initial cervical spine X-ray showed a lucent area with surrounding sclerotic lesion at the first thoracic spine mostly on the right side. Computed tomographic studies revealed an expansile well-corticated lesion with central radiolucent nidus with bony sclerosis. The transitional zone is preserved. It was causing a spinal canal narrowing. T1 images of MRI showed iso/intermediate signal intensity with an area of low/flow void due to calcification. T2 imaged of MRI shows a hypodense with surrounding vertebral body high intense favor of bone marrow edema. This edema also extends into the right pedicle, lamina, and spinous process. In the surrounding paravertebral soft tissue illuminated a high signal. There was no spinal cord compression, and a mild thecal sac impingement was seen. The initial blood investigation is within the normal range.



**Figure 1**

**Figure 1** shows a radiolucent area in the first thoracic spine in the lateral of cervical spine Xrays

**Figure 2** shows an expansile well corticated lesion with central radiolucent nidus and bony sclerosis in an axial view of first thoracic spine computed tomographic image



**Figure 2**



**Figure 3**





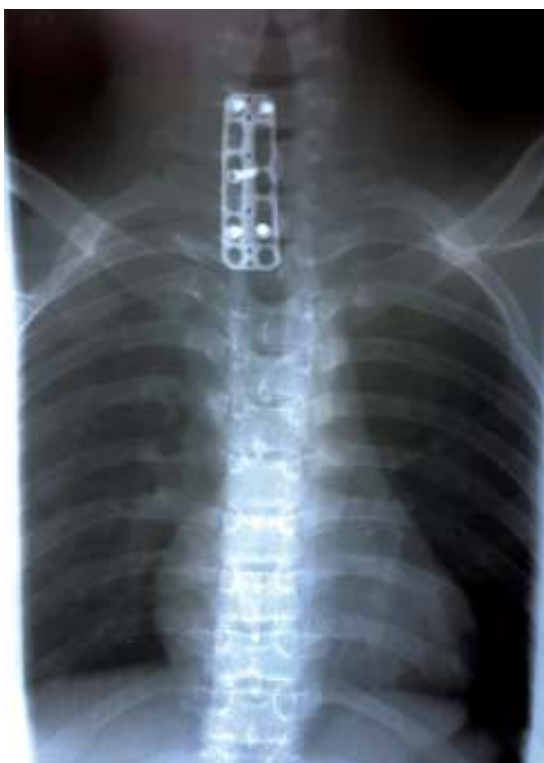
**Figure 4**

A corpectomy and fusion with instrument stabilization using an anterior approach were planned in the multidisciplinary team meeting. The surgery was performed along with a neurosurgeon. His first thoracic vertebral body was completely excised using a standard Smith and Robinson approach. the tumor was peeled off from the dura and hemostasis was achieved. A tricortical iliac graft was harvested and used as a strut graft for fusion. An anterior plating was applied to provide instrumental stability. Initial postoperative care was uneventful. His histological report confirmed it as osteoblastoma. He is now pain-free, his normal range of movement is achieved and followed up in the clinic.

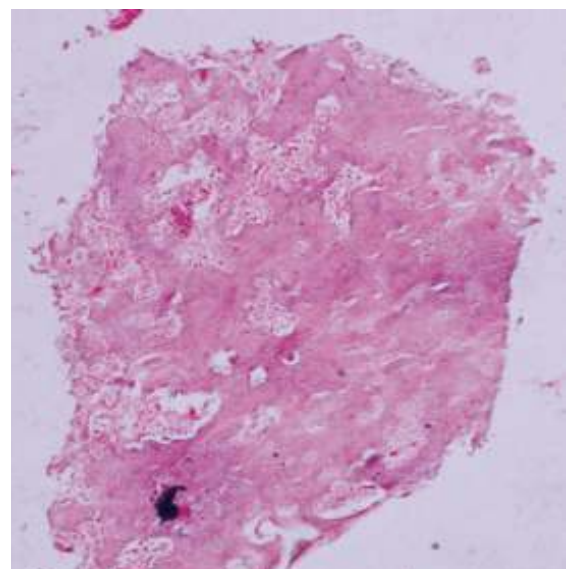


**Figure 6**

**Figure 5** (an anterior posterior view of lower cervical and upper thoracic spine) & **figure 6** (a lateral view of lower cervical and upper thoracic spine) shows a immediate postoperative Xrays of corpectomy of first thoracic vertebra and instrumental stabilization.



**Figure 5**



**Figure 7**

**Figure 7** shows a bony lesion composed of woven bone trabeculae. These trabeculae are haphazardly arranged and lined by a single layer

of osteoblasts. A rich vascularity is identified with extravasated red blood cells.

## Discussion

Osteoblastoma is a benign primary bone tumor that mimics osteoid osteoma histologically(4). The size of the lesion is the main differentiating feature. The diameter of the osteoblastoma is usually more than 2cm in diameter whereas osteoid osteoma is less than 1.5cm(5). The incidence of osteoblastoma in all bone tumors is only 1% and among the spinal tumors, it is 10%(1). It predominately affects the posterior elements of the spine mainly the pedicle and lamina(6). It affects the children with the age of 10 to 15years mostly, but the age of presentation varies from 6 months to 75 years(3). However, nearly 80% of the people will present before the age of 30years. Males are having 2.5 times more risk than females to develop osteoblastoma in their lifetime(3).

Clinically presentation of the lesion varies according to the location of the tumor. But typically, a dull aching type of night pain that is difficult to locate and respond to the simple analgesics is the common presentation(7). Thoracic osteoblastoma may present with upper back pain, intercostal neuralgic pain, or even with myelopathic features(7). Cervical osteoblastoma patients may present with neck discomfort or oropharyngeal pain(8). Lower cranial nerve palsy with the craniovertebral junction involvement of osteoblastoma and abdominal symptoms with the sacral bone involvement was also described in the literature(8). New onset of scoliosis and torticollis in a child warrant exclusion of spinal osteoblastoma(9). One-third of the spinal involvement of the osteoblastoma presented with neurological involvement. Thoracic lumbar osteoblastoma or osteoblastoma involving the ribs may present with painful rapid progressing scoliosis(10).

The aggressive and conventional type of osteoblastoma is the two types of osteoblastoma elaborated in the literature(11). Aggressive type

osteoblastoma can extend to the paravertebral and epidural spaces. It is larger than conventional type osteoblastoma(11). Histologically osteoblastoma is an expansile lesion with high vascularity that exhibits both osteolytic and osteoblastic activities. Evidence of aneurysmal bone cyst formation and areas of hemorrhage were also noted histologically(12). Large epithelioid osteoblasts and invasion of cortical bone are the two differentiating histological features to differentiate aggressive type from the conventional type(11). The transition of osteoid osteoma into osteoblastoma was documented in the literature but less commonly(13).

Osteoblastoma usually appears as an osteolytic lesion in the plain X-ray. It can also appear as a central osteolytic nidus which is surrounded by osteosclerotic margins like osteoid osteoma(14). Matrix calcification, mineralization of the nidus, cortical bony destruction, and extension into the surrounding structures are the common radiological features of aggressive type tumor which can be demonstrated with the help of computer tomographic studies(14). MRI imaging technique is needed to demonstrate the involvement of the surrounding structures. Technetium-99 bone scan shows uptake at the site of the lesion. But bone scintigraphy studies are more sensitive in diagnosing the lesion of the osteoblastoma(5).

Enneking classification system is commonly used to classify musculoskeletal tumors which classify osteoblastoma into three types. It is based on the histological, radiological, and clinical presentation of the tumors(15). The Latent (S1) stage is an inactive tumor that grows slowly and is surrounded by a well-circumscribed capsule. The Active (S2) stage is a slowly growing active tumor with a thin capsule. The Aggressive (S3) stage is an aggressively growing tumor with an incomplete or no capsule(15). It tends to acquire a pseudo capsule by invading into the surrounding structures. It is associate with a high level of recurrence rate(15).

The goal of surgical treatment is resection of the tumor and prevention of recurrences.

Radical excision, curettage of the lesion combined with or without radiotherapy are available surgical treatments. Radical excision of the tumor rather than a curettage is proven to be beneficial in the outcome(11). Stabilization with instruments and fusion is necessary to prevent progressive deformity in the future. Because of the high vascularity, bleeding is a devastating complication during the surgery(15). Cervical spine osteoblastoma is closed to the foramen to the vertebral artery. Iatrogenic damage to the vertebral artery is also one of the commonest complications during the excision of cervical spine osteoblastoma. Intraoperative navigation and pre-operative embolization of the feeding vessel to the tumor are options available to reduce the bleeding during surgery(15).

Radiotherapy is not commonly used as a primary modality of treatment because of the risk of late transition of sarcomatous changes(16). It has been used as an adjuvant to surgical treatment for stage 3 osteoblastoma which is not suitable for complete excision. Methotrexate, bisphosphonate, and bevacizumab are used as chemotherapeutic agents' adjuvant to surgical excision. A combination of doxorubicin, methotrexate, and cisplatin was also used as

chemotherapeutic agents which successfully stopped the tumor progression up to three years(5). Radiofrequency ablation of spinal osteoblastoma is also a treatment option described which requires experts and the facilities(17).

The recurrence rate of tumors is less than ten percentage following complete excision of tumors(5). survival of stage 3 osteoblastoma following a surgical excision adjuvant with radiotherapy reported up to 25years(5). Profuse bleeding which leads to death during surgery, local recurrence, progressive development of kyphosis, and hardware failure are the documented surgical complication following excision of spinal osteoblastoma(18).

## Conclusion

Thoracic spine involvement of osteoblastoma is a rare entity that can be successfully treated surgically. Young males are the most affected population. Early diagnosis and a multidisciplinary team approach need to provide a better outcome to the patient. A preoperative radiological guided embolization will reduce the devastating bleeding during the surgery. A multidisciplinary approach is important in diagnosis and treatment.

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## Case Report 5

# Hemangiopericytoma of lumbar spine: A mystery behind low back pain

### Case history

A 45 year old previously healthy lady presented with mechanical type of low back pain of 4 months duration. She was treated by a general practitioner with analgesics and a course of physiotherapy but did not help with the pain. The onset was gradual and slowly progressing pain dull in nature. There was no radiation to the lower limbs but recently she noticed numbness and weakness of both legs two weeks prior to the admission. She denied recent falls or trauma to the back, she had no issues passing urine and no changes in the bowel habit. With time her symptoms affected badly and she had to depend on walking aids for mobility. There were no features suggestive of an infective process as well as no symptoms related to any primary malignancy such as lung, thyroid, breast, bowel and genitourinary.

On physical examination, her gait pattern was affected due to the back pain. There was midline and tenderness over the lumbar spine with limited spinal movements. Decreased sensation over bilateral lower limbs from mid thigh downwards was noted. Further neurological assessment revealed bilateral paraparesis of medical research council (MRC) power 3/5. There was no saddle anaesthesia and anal sphincter tone was preserved. Bilateral knee and ankle reflexes were weak and planter reflex was equivocal. Laboratory investigations

didn't reveal any significant alteration. Erythrocyte sediment rate (ESR), C-reactive protein (CRP), and tumour markers were within normal range. Chest radiograph was unremarkable.

Plain X ray of lumbo sacral spine anteroposterior and lateral views were obtained and it showed significant height loss of L2 vertebra with lytic lesion within the body (Figure 1). Disc space was maintained and grade I spondylolisthesis was also reported at L5-S1 level. Magnetic resonance imaging (MRI) was then requested and it showed an extra medullary enhancing lesion extending anteriorly and posteriorly causing significant cauda equina compression with severely collapsed L 2 vertebral body (Figure 2). Computerized tomography guided percutaneous biopsy was performed and the histological analysis showed a very vascular spindle cell tumour with slit like and staghorn vascular spaces, suggestive of haemangiopericytoma.

A decision was obtained to operate the patient in view of severe cord compression, progressive neurological deficit and to obtain a tissue sample for confirmation of the diagnosis. Considering vascular control during surgery a preoperative angiogram was performed and it showed the tumour receives blood supply from both L2 lumbar segmental arteries and angio embolisation was performed two days prior to the surgery.

Surgery was performed through anterolateral retro peritoneal approach with the patient in right lateral position. L2 vertebral body was identified. Segmental arteries were ligated. L2 corpectomy was performed (Figure 3,4) and the reddish fleshy mass of tumour was removed.

Spinal stability was achieved with expandable lumbar cage and lateral plating. Post operatively significant pain improvement neurological recovery was observed. Histology report confirmed the diagnosis of haemangiopericytoma without evidence of malignant potential.



Figure 1



Figure 2



Figure 3



Figure 4

## Discussion

Haemangiopericytoma is a rare vascular tumour originates from Zimmerman's pericytes which are contractile spindle cells lining the capillaries and post capillary venules. This was first described by Stout and Murray in 1942. It accounts for less than 1% of all vascular tumours. It exhibits both benign (80%) and malignant forms (20%). It may arise from any part of the body where capillaries are present, but most common sites are lower extremities, retro peritoneal region followed by head and neck region. Spinal haemangiopericytoma is extremely rare and only 80 cases have been reported worldwide. There is no sex preference but a slight predominance in males is reported. The soft tissue form commonly occurs in fifth and sixth decades whereas the osseous form commonly arises in fourth and fifth decade.

The tumour can be locally aggressive and it has a very high potential for local recurrence (80%). The malignant form is highly aggressive and preferably metastasizes to lung and bone (23%). Histologically it is characterized by dense blunt spindle cell proliferation with a richly vascular stroma. The capillary vessel proliferation

often acquires a staghorn configuration. Each pericyte is characteristically surrounded by rich reticulin network. The histological feature that favours malignant potential includes mitotic count more than 4 per high power field, nuclear atypia and necrosis.

Clinical manifestations may vary depending on the size of the tumour and the location. Pain may not be reported in soft tissue form whereas pain is the first symptom commonly experienced in osseous type of tumour. Neurological deficit may result from vertebral tumours causing cord compression or as a result of pathological fracture.

There are no specific radiographic features of these tumours. However, characteristic signs that may help in the diagnosis. A "spider shaped" appearance in the arterial phase and dense well demarcated round or oval tumour staining in the venous phase are the characteristic features.

Surgical resection remains the first choice of treatment for all type of tumours when feasible. Spinal tumours should be resected as en bloc so as to relief neural compression. However, radical resection is often impossible without causing significant neurological deficit. Pre operative

Angio embolisation is an efficient strategy to minimize intra operative bleeding. Angiographic studies help in identifying the feeder vessels. Complete resection of benign tumours is sufficient whereas in malignant cases additional radiotherapy or chemotherapy may be considered especially in high grade tumours, large tumours and resection with positive margins. Radiotherapy alone is indicated in unresectable tumours, similarly chemotherapy alone is indicated in unresectable tumours and metastasis. However, the effectiveness of adjuvant chemotherapy and radiotherapy is still uncertain. The prognosis of the tumour largely depends on resectability and the histologic grading. Usually the benign tumours have a good outcome but in contrast malignant counterparts have a high rate of local recurrence and propensity to metastasize.

## Conclusion

Haemangiopericytoma is an extremely rare vascular tumour which can mimic a destructive metastatic lesion in the spinal column. Therefore the primary goal in the management of a lytic lesion in patients over 40 years, is to exclude secondary metastatic deposits from common primary sites. Haemangiopericytoma of spinal column should be considered potentially malignant and therefore be treated promptly and aggressively. Surgical resection is always the mainstay of treatment and should be attempted whenever feasible. The role of radio therapy and chemotherapy is still uncertain and therefore may be considered in high risk patients. Both benign and malignant tumours should be followed up for long term for the early detection of recurrence.

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## Case Report 6

# Fibrodysplasia Ossificans Progressiva : A Case Report

### Introduction

Fibrodysplasia ossificans progressiva, also known as myositis ossificans progressiva, stone man disease or Munchmeyer's disease is a rare connective tissue disorder characterized by heterotrophic ossification of extra skeletal sites such as muscles, ligaments and tendons resulting in severe disability.

### Case Report

A fifteen-year-old girl presented with a history of episodic spontaneous swelling of the inter scapular region for two months duration. These lumps were associated with throbbing pain at onset which lasted a few days followed by resolution of the swelling leaving firm immobile mildly tender lumps of the size of about 3 cm. Ultrasound scan (USS) at this point revealed subcutaneous benign hypo echoic lesions without an increase in vascularity. Excision biopsy was carried out following which she developed painful neck and chest wall swelling. At this point USS scan and CT scans revealed inflammatory changes in trapezius, scalene anterior and sternocleidomastoid muscles with subcutaneous oedema. The histology revealed fibrotic muscle, vascular stroma, scattered lymphocytes and osteoclast like giant cells, immature bone formation surrounded by osteoblasts and injured myosites suggestive of myositis ossificans.

She further developed restricted movements of the left

shoulder at which point she was referred for tertiary hospital for second opinion where she underwent a contrast enhanced CT which revealed coarse calcification at the origins of right latissimus dorsi at the inferior angle of the scapula, spinous process of T10 vertebra and ileac crest. There was also calcifications at the origin of serratus anterior at the 4-6 ribs and in the right scalene anterior at its insertion to the 1st rib. These findings were in keeping with dystrophic calcification seen following trauma or strain. With this she was offered a tenotomy of latissimus dorsi muscle to improve shoulder mobility. Histology at this procedure revealed skeletal muscle tissue with central area of fibrosis and foci of metaplastic cartilage with enchondral ossification which was again concluded as myositis ossificans.

She was referred to the rheumatology unit for further evaluation where she underwent screening for possible immune mediated myopathy (ESR 7mm/1st hr, CRP 1.9mg/dl, CPK 49u/l, ANA negative) and disorders of calcium metabolism (ALP was 132.2 U/l, AST 23.6 U/l, ALT 11.7 U/l, GGT 17.7 IU/l, Serum creatinine 50.1 µmol/l, Serum Calcium 2.38 mmol/l, serum PTH 17.4 pg/ml, serum PO4<sup>3-</sup> 0.99 mmol/l, Vitamin D level 21.8 nmol/l ) all of which were normal except for low vitamin D levels. However as vitamin D deficiency does not give rise to this clinical picture it was excluded as the cause



for her presentation. Endocrine opinion was also taken at this point and it was concluded that this is unlikely to be a disorder of calcium metabolism.

With the given history of spontaneous myositis ossificans at multiple sites without a history of previous significant trauma with characteristic flare ups and with the presence of characteristic bilateral hallux valgus deformity (Figure 1) a diagnosis of Fibrodysplasia Ossificans Progressiva was made. She had a repeat CT scan at 5 months from the onset which showed extensive longitudinally extending dense calcifications in bilateral latissimus dorsi, serratus anterior, serratus posterior inferior, erector spinae, capitis and quadratus lumborum muscles which was in keeping with the diagnosis (Figure 2). Second opinion was taken from tertiary care rheumatologist who also agreed with the diagnosis and the management plan.



**Figure 1**



**Figure 2**

She was started on Celecoxib 200mg bd, Vitamin C 400 mg daily, Montelukast 10 mg daily and desloratadine 5m daily. Flare ups (acute attacks with new onset lumps) were treated with Prednisolone 1mg/kg/day for 4 days. Her vitamin D was corrected with oral D3

200000IU 2 doses two weeks apart followed by 2000 IU daily. She was given IV pamidronate 1mg/kg/day for 3 days for one flare up which did not respond to steroids. She was advised to avoid falls, injuries, exposure to infections, surgical procedures and IM injections. She was advised

to take prednisolone 1mg/kg/day for 3 days following trauma, surgical or dental procedures as prophylaxis. Her immunization was up to date and she was given Covid 19 pfizer vaccine subcutaneously. She was offered occupational therapy and gentle physiotherapy to aid her with the activities of daily living. After 8 months of onset she had limited mobility of left shoulder and neck movements and continued to get flares once every three to four months.

## Discussion

Fibrodysplasia ossificans progressiva (FOP) is a heritable disease inherited in an autosomal dominant pattern. However, most of the cases are sporadic. It is caused by a mutation of ACVR1/ALK2 gene encoding a bone morphogenetic protein type 1 receptor1. The prevalence of this condition is 1 in 2 Million<sup>2</sup>.

Usually, patients develop clinical signs and symptoms during the first decade of life. They present with episodic painful soft tissue swelling (flare ups) which may resolve or result in ribbon like ossification of underlying muscles, ligaments, tendons and aponeuroses. Neck, back and shoulders are the first sites to be involved. The swelling typically shows three stages: first painful stage (first few weeks) followed by painless induration phase and the late phase (after 12 weeks) which shows radiographic ossification<sup>3</sup>. Flare ups are triggered by injury, viral infection, surgical procedures, muscle stretching and intramuscular injections. The heterotrophic ossification progresses and spans across joints leading to limited mobility. This is a progressive disease which leads to severe disability including feeding difficulty caused by ankylosis of the jaw and respiratory insufficiency due to reduced mobility of the thoracic cage. The average life expectancy is 40 years.

The patient with FOP has characteristic bilateral hallux valgus deformity with malformed first metatarsals and fused interphalangeal joints. They may also have short malformed thumbs, clinodactyly and neck stiffness. Hearing loss may be associated in about 50% of the cases<sup>3,4</sup>.

Typically, biochemical studies are normal in these patients however alkaline phosphate maybe elevated during heterotrophic ossification phase of flare ups. Characteristic radiographic findings include soft tissue ossification, malformed first metatarsals and fused interphalangeal joints. There may be medial tibial osteochondromas, fusion of posterior elements of cervical spine and malformation of thumbs.

Diagnosis can be confirmed by genetic testing. Genetic testing was not carried out in our patient due to unavailability, however her clinical features were in keeping with the diagnosis with characteristic great toe malformations and sites of heterotrophic ossification and the presence of characteristic flare ups.

Other condition that needs to be excluded in the presence of progressive soft tissue ossification is progressive osseous heteroplasia (POH). In POH ossification starts from cutaneous tissue and extends to involve deep connective tissue, lacks flare ups and the great toe abnormalities.

Aggressive juvenile fibromatosis can present with rapidly growing soft tissue swelling but can be differentiated from FOP in the absence of great toe malformations and heterotrophic ossification.

Extensive calcinosis cutis (Calcinosis universalis) can rarely be seen in autoimmune connective tissue diseases like juvenile dermatomyositis and systemic sclerosis. Typically calcinosis start in limbs and is associated with other clinical features of connective tissue disease and auto antibodies. Histological findings are that of dystrophic calcification and differ from that seen in myositis ossificans.

There is no definitive management for FOP. The current treatment considerations by The international clinical council for FOP (ICC) and consultants (2022) include management of flare ups with steroids (prednisolone 1-2mg/kg/day) for 3- 4 days, steroid prophylaxis (prednisolone 1mg/kg/day for 3 days) following blunt muscle trauma, surgical and dental procedures and Cox 2 inhibitors to ameliorate pain<sup>5</sup>.

IV Bisphosphonates<sup>6</sup>, ascorbic acid<sup>7</sup> and mast cell stabilizers have been used to achieve some improvement in disease course. Patient education on avoidance of precipitants, supportive care with occupational therapy, gentle physiotherapy (avoiding passive muscle stretching), addressing mental health issues, orthotics, addressing respiratory health and prevention of viral infections play an important role in the management.

## Conclusion

It is important to consider FOP in the differential diagnosis of children presenting with progressive soft tissue swelling and look for characteristic clinical features to minimize surgical procedures which would lead to rapidly worsening of the condition.

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## Case Report 7

# Management Of Acute Pulmonary Embolism Following Chest Trauma In A Resource Limited Setting – A Case Report

**KEY WORDS** - Pulmonary embolism, Trauma, Frontal contusion, Cardiac arrest, Resource limited setting

### ABSTRACT

**Introduction:** Pulmonary embolism is a known complication among trauma patients and contribute to significant mortality and morbidity. In the setting of trauma, the signs and symptoms of VTE may be masked. Therefore, a high index of suspicion and timely intervention can improve outcome.

**Presentation of case:** A 57-year-old healthy female presented with a history of fall from a train, in to a gap between the train and the platform. She had sustained cerebral contusions in the frontal lobes and right sided lung contusion. She was resuscitated and managed in the trauma ICU and send to a ward.

She desaturated in the ward and followed by a cardiac arrest. Bed side 2D echocardiogram and CT pulmonary angiogram were suggestive of PE. Thrombolysis was followed by clot retrieval. Patient was discharged home on day 13 after cardiac arrest on warfarin therapy.

**Discussion:** Causes for PE are multi-factorial. Failure to initiate prophylaxis therapy for deep vein thrombosis in trauma victims is a major cause for PE. Management of PE in a haemodynamically unstable patient with a background history of trauma is challenging. Neuro-imaging revealed bi frontal cerebral contusions hence, the treatment of choice was clot retrieval. However, due to limited resources and logistics, medical thrombolysis was initiated with the involvement of a multidisciplinary team. Clot retrieval was initiated within 14 hours of diagnosis.

**Conclusion:** Clinical suspicion and early diagnosis of PE can improve outcomes. Management is challenging in a low & middle income countries, where resources are limited. In the presence of contraindications and failed thrombolysis, clot retrieval remains an important aspect in the management of PE.

### INTRODUCTION

Acute PE is a major cause of mortality and morbidity. The exact prevalence of PE is not known but the incidence is reported at 50-75 per 100

000 in Australia and New Zealand, with a 30-day mortality rate ranging from 0.5% to more than 20%<sup>1</sup>. In recent studies overall mortality due to PE is reported to be as high as 18% to 65 %. Mortality is around

20% in patients who have received thrombolysis as treatment. Associated cardiogenic shock increases mortality to 25% -30%. Patients who had cardiopulmonary resuscitation mortality was as high as 65%<sup>2,3,4</sup>.

PE and VTE are widely reported among medical, surgical and trauma patients. Trauma is responsible for 12% of VTE occurring in the community<sup>5</sup>. According to a study conducted by Bahloul et al PE complicates 18% of ICU admissions due to trauma<sup>6</sup>. It was reported to be the third most common cause of death in patients who survived the first 24 hours<sup>5</sup>.

Local prevalence data on PE are not available, despite a number of reported cases of PE with varied presentations in Sri Lanka.

This case report highlights the timely diagnosis and intervention of a trauma victim who suffered a cardiac arrest following PE but recovered successfully in a setting with limited resources.

## CASE REPORT

A previously healthy, 57-year-old woman was admitted to National Hospital accident service following a rail way tract accident. While trying to get on to a train she had fallen in between train and the platform. She was conscious and rational on admission to hospital with a GCS of 15/15. Pulse rate was 104 beats per minute and blood pressure was 110/69 mmHg. She complained of headache and difficulty in breathing. Non-contrast CT scan of the brain showed bi frontal cerebral contusions. She had a right sided lung contusion, clavicular fracture and a haemothorax.. Following insertion of a right sided intercostal drain She remained comfortable and haemodynamically stable. She was transferred to the surgical ward for further care.

Patient developed desaturation with poor respiratory efforts and was transferred

to the trauma intensive care unit, where she received intermittent non-invasive ventilation and multi modal analgesia. Intermittent pneumatic compression devices were applied. Pharmacological DVT prophylaxis was not initiated due to cerebral contusions and haemothorax. After three days of supportive management in ICU she was re transferred to the ward. Intercostal tube was removed after one week of insertion and patient was mobilized.

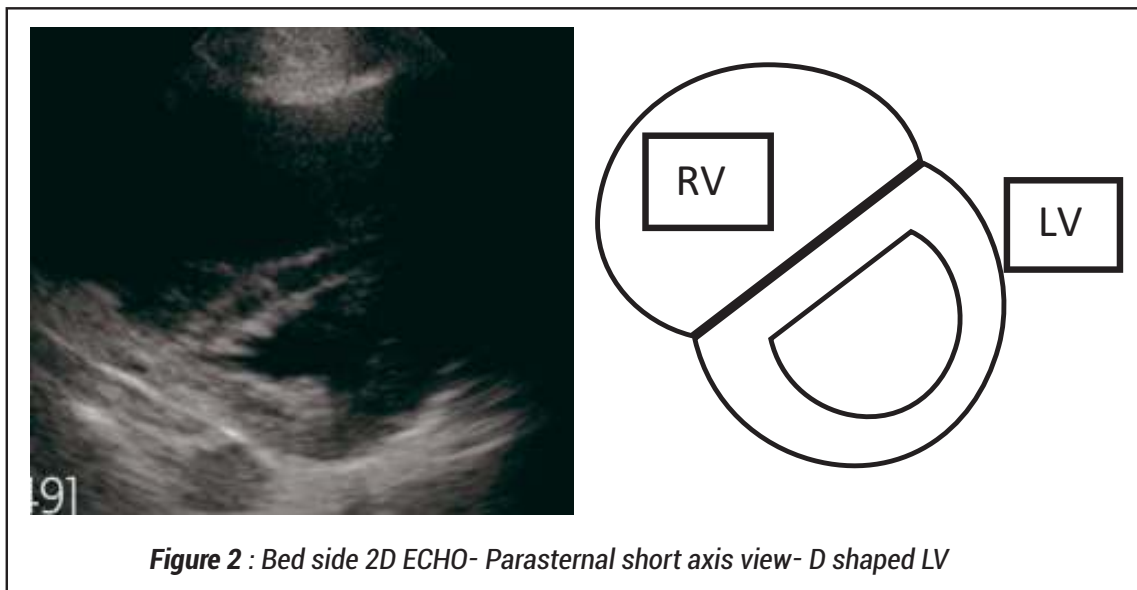
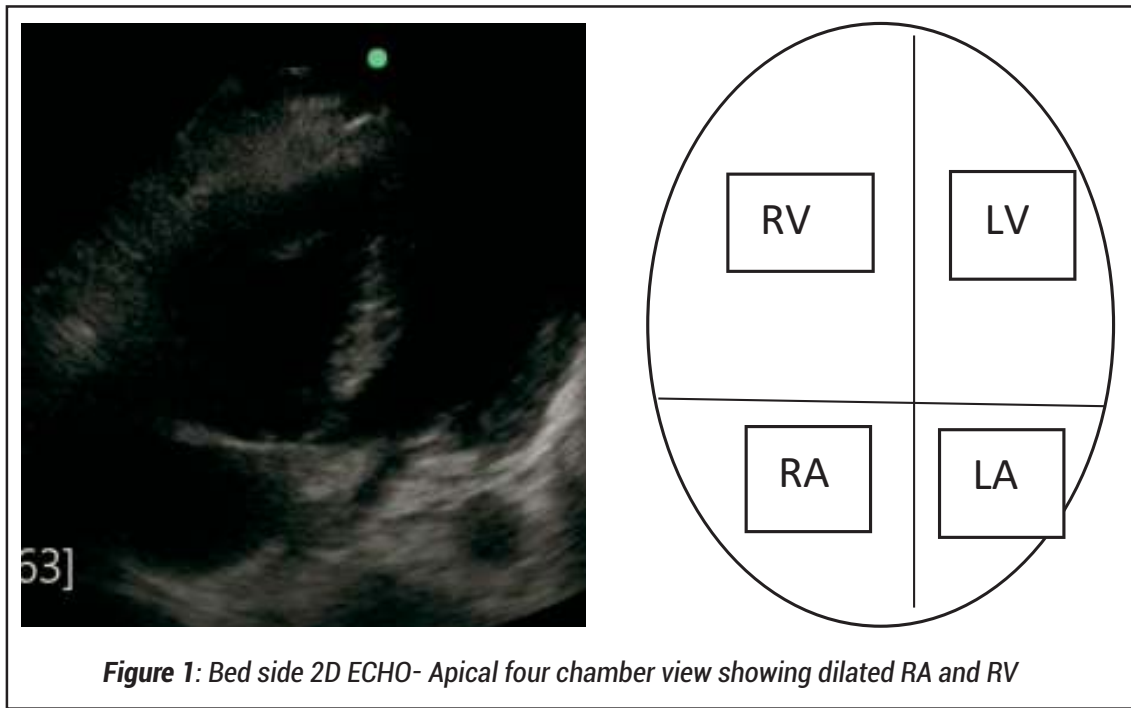
On the eighth day following the initial injury, she complained of sudden onset breathlessness on walking. Her respiratory rate was 40 breaths/min with oxygen saturation 75% on room air. She was given 15L of O<sub>2</sub> via a non-rebreathing mask. Patient developed an asystolic cardiac arrest in the ward. Cardio pulmonary resuscitation was initiated as per the standard ALS guidelines. Return of spontaneous circulation was noted within six minutes of the cardiac arrest. She was intubated and an intercostal tube was re-inserted on the suspicion of a pneumothorax as the patient had reduced air entry over the right lung base.

Patient was transferred back to trauma intensive care unit for post resuscitation care. On arrival she had GCS of 6/15 (Eye-1. Motor 4, Verbal-1). Bi lateral pupils were 3mm with equal reaction to light. Pulse rate was 174 beats/min, blood pressure was 100/55 mmHg on an infusion of noradrenalin. Bi lateral lung fields were clear. SpO<sub>2</sub> was 90 % on 100% oxygen.

Initial arterial blood gas showed a PH of 7.188, PCO<sub>2</sub> -26.2 mmHg, pO<sub>2</sub>-65.3 mmHg, Lactate 8.6 mmol/L, HCO<sub>3</sub><sup>-</sup> 10 mmol/L and a base excess of (BE)- -18.4 mmol/L. Electrolytes were normal.

Electrocardiogram showed sinus tachycardia

A focused 2D ECHO was done in the ICU, which showed good LV contraction with significantly dilated RV and RA, flattening of the interventricular septum, and a dilated inferior vena cave with minimal respiratory variations.



Clinical presentation of sudden onset of shortness of breath followed by a cardiac arrest in a poorly mobilized patient with given 2D ECHO findings lead to a diagnosis of PE.

Patient underwent an urgent CTPA which showed a large pulmonary embolus with in the right upper segmental pulmonary artery and multiple large thrombi in the left lower segmental pulmonary arteries. Main pulmonary artery was dilated up to 3 cm in diameter. The RV was grossly dilated with the interventricular septum bulging in to the left ventricle.

Clot retrieval remained the treatment of choice. Medical thrombolysis could worsen the bi frontal contusions and the haemothorax. We opted for thrombolysis after considering the risk versus benefit with the multidisciplinary team. In a resource constraint setting organizing for clot retrieval was difficult. Patient was haemodynamically unstable for transfer to the radiology suite. The decision between the two treatment options was a challenge to the clinicians and posed a dilemma.

Intravenous alteplase 100 mg was given over two hours, followed by intravenous heparin

1000 IU per hour as an infusion, whilst monitoring APTT according to the standard protocol.

Despite improvement in oxygenation the vasopressor and inotrope requirement increased over the next few hours of initiation of thrombolysis. Her blood pressure was 94/65 mmHg and heart rate 130 beats per minute. She was on noradrenaline 0.5 mcg/kg/min, intravenous vasopressin 4U/hour and intravenous dobutamine 10 mcg/kg/min.

2D ECHO showed poor right heart functions.

Decision was made for clot retrieval via interventional radiological technique under general anaesthesia. Following retrieval haemodynamic parameters improved and cardiovascular support was tailed off gradually.

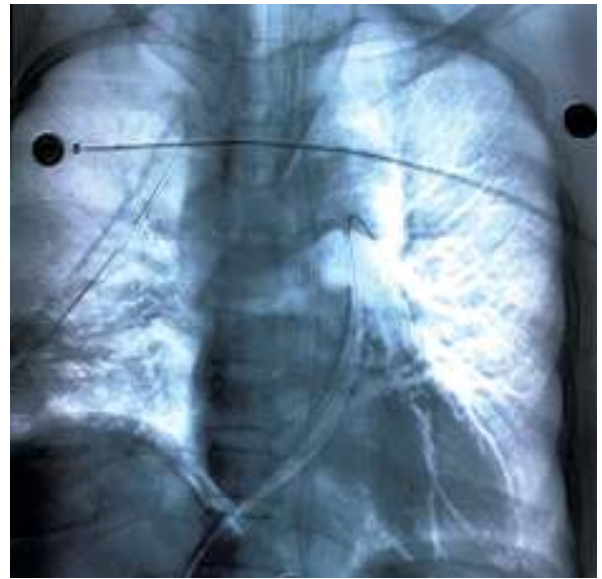
Unfractionated heparin infusion was converted to therapeutic doses of low molecular weight heparin.

Venous duplex scan of both lower limbs was negative for deep vein thrombosis.

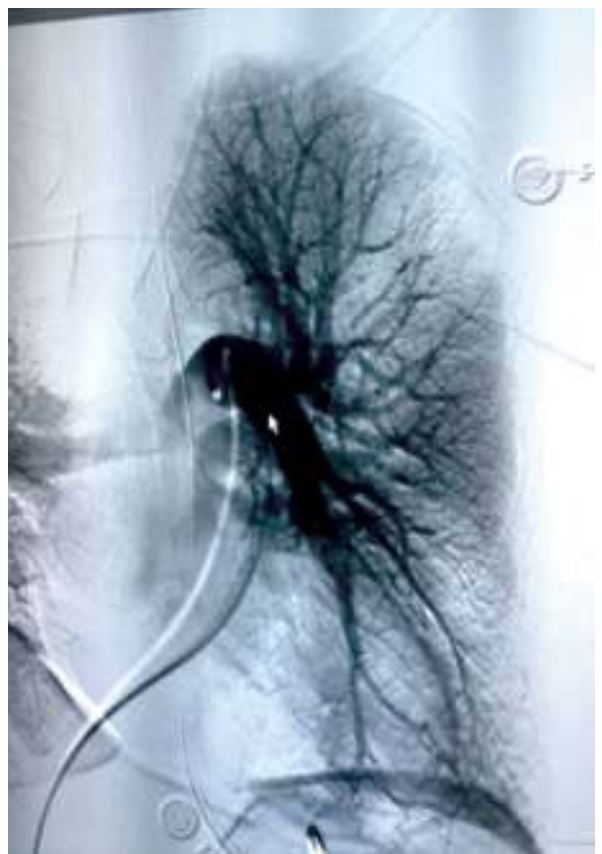
Seven days after the cardiac arrest patient was transferred back to the ward on warfarin and enoxaparin as bridging therapy. She was discharged from hospital on day 13 following the cardiac arrest with no neurological sequelae.



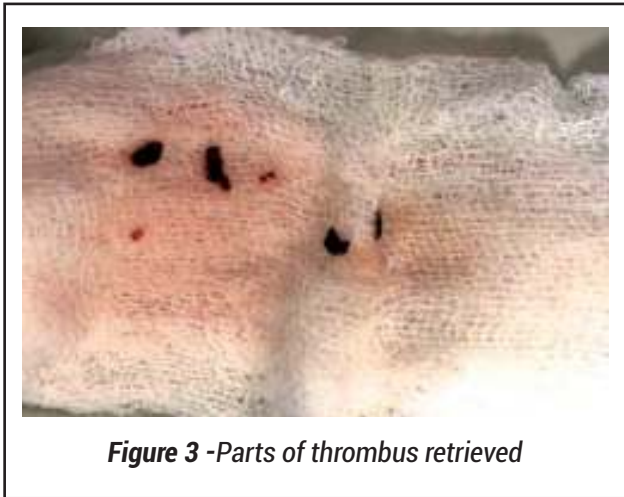
**Figure 3 - Filling defects in bi lateral segmental pulmonary arteries- CTPA image**



**Figure 4 - Filling defects in bi lateral segmental pulmonary arteries- image acquired during clot retrieval**



**Figure 3 - Filling defects in left sided segmental pulmonary arteries- image acquired during clot retrieval**



**Figure 3 -Parts of thrombus retrieved**

## DISCUSSION

DVT and PE are known complications in trauma patients. Incidence of PE among trauma patients vary considerably, ranging from 0.35% to 24%<sup>5</sup>. Contrary to popular belief PE occur more commonly during days 5 to 7 after trauma many studies have shown increasing evidence of early onset PE. A descriptive study conducted by Menakar J et al reported 37% of all cases of PE were diagnosed within day 1 to 4, 18 % during day 5 to 7, 23% in day 8 to 14 and rest over day 14 to 20 after trauma<sup>7</sup>. Transient hypercoagulability that occurs in the first few days following injury leads to early on set PE<sup>5</sup>. Some studies have demonstrated that tissue factors and thrombin generation increase after trauma and levels of natural anti coagulants are diminished. (e.g.: anti thrombin III, protein C and S )<sup>8</sup>

Fractures of the lower limbs, obesity and age are known risk factors to develop PE<sup>5</sup>. In trauma victims' spinal injuries, traumatic brain injuries, severe chest trauma and high injury severity score were identified as predictive risk factors<sup>5,9</sup>.

These injuries lead to prolong periods of immobilization and also hinder the use of anti-coagulants prophylactically within the first 48 hours after injury<sup>5,9</sup>. Traditional risk factors such as active cancer, prior history of DVT or PE, major venous repair, mechanical ventilation for more than four days, central line placements

are also relevant to this cohort of patients<sup>10</sup>. Our patient had chest trauma, immobilization and delayed initiation of anti-coagulants as risk factors for developing PE.

Treatment of PE in a trauma patient pose challenges to the clinician. Management decisions are guided according to relative or absolute contraindications for thrombolysis or anti coagulation and weighing benefit vs harm. If there are contraindications for thrombolysis in a trauma patient, pulmonary interventional thrombolysis and thrombus aspirations techniques should be considered. There is a place for ECMO selected cases<sup>11</sup>.

When compared to an ideal setting we faced many challenges during the management of this patient. This patient suffered major trauma just eight days prior to the cardiac arrest secondary to PE. Major trauma, or head injury in the previous three weeks is considered an absolute contra indication to thrombolysis according to the European society of cardiology guidelines on management of PE <sup>12</sup>. Due to non-availability of appropriate size catheters for clot retrieval in our institution , we resorted to thrombolysis.

Other options discussed in the literature for management of PE include catheter directed thrombolysis, catheter embolectomy, surgical embolectomy and or mechanical circulatory support devices<sup>13</sup>. catheter embolectomy was the next available option available in our setup.

Within 14 hours after ICU admission, we offered clot retrieval for the patient with smaller size catheter. By using smaller catheters they can disturb the clot facilitating clot lysis as well as perfusion through a disturbed clot.

Lower limb duplex scan was negative in our patient. Becher M et al in a recent study studied 168 patients with PE and 46.4% of patients had negative duplex scan for lower limb DVT. Most of these patients had peripherally located PE, and mostly diagnosed with V/P-SPECT rather than CTPA <sup>14</sup>.



A recent article introduces a new concept i.e., PE and in situ PT to be two different clinical variants. This debate has arisen as some patients with PE were persistently negative for DVT as in the case of our patient. In a prospective cohort study done in the United States of America, it was found that independent risk factors for PT include shock on admission, major chest injury with an Abbreviated Injury Score of three or more and major venous injury<sup>15,16</sup>. In this trial pulmonary thrombosis was defined as pulmonary clots on chest CTPA without concomitant lower extremity DVT. Despite the new terminology, the management of these patients remain the same<sup>15</sup>.

## CONCLUSION

Acute PE is a major cause of morbidity and mortality following cardiac arrest.

A bed side echocardiogram helped to identify RA and RV dilatation, so that management was initiated without undue delay. Despite the presence of contraindications to initiate thrombolysis in a trauma victim this patient was successfully treated with thrombolysis followed by clot retrieval with the best possible available devices in our institution.

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Early detection, immediate management and involvement of the multidisciplinary team lead to the successful outcome.

**ABBREVIATIONS**–Pulmonary embolism -PE, Deep vein thrombosis- DVT, Venous thrombo embolism – VTE, Intensive care unit-ICU, Advanced life support ALS, Pulmonary thrombosis -PT, Computerized tomography pulmonary angiogram- CTPA, Right ventricle -RV, Right atria- RA, left ventricle-LV, Left atria- LA. Two-dimensional echo cardiogram- 2D ECHO, Activated partial thromboplastin time, APTT, Ventilation/perfusion single photon emission computed tomography V/P SPECT, Extra corporal membrane oxygenation- ECMO.

## CONSENT:

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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## Case report 8

# Supracondylar fracture humerus with pink pulseless hand and angiogram confirmed vascular injury – Case report

**Key Words:** supracondylar fracture humerus, paediatric, pink pulseless hand, vascular injury

## Abstract

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- Supracondylar fracture of the humerus is a common injury in paediatric population. Neurovascular damage is a well-known serious complication of this injury. This should be managed urgently by the treating clinician in order to prevent severe disability to the child. In this case report, we illustrate the presentation, management and follow up of a 7-year-old boy presented with a supracondylar humerus fracture, highlighting the diagnostic and therapeutic measures taken in sequential manner.
- The child presented following a domestic fall and having pain, swelling and acute deformity of the left elbow. Clinically, no distal radial pulse or oxygen saturation using pulse oximetry detected on admission, even though the left hand was warm and pink with normal capillary refill time. X-ray confirmed the clinical diagnosis of supracondylar fracture humerus, and it was classified as Gartland type III.
- Manipulation and casting done without internal fixation on the next available casualty list and even after 72 hours of the procedure no return of distal pulse despite warm, pink hand. After vascular opinion, CT Angiogram revealed no flow segment of 2.2cm of brachial artery at the trauma site and minimum demonstrable flow distally. However, watchful waiting was selected as the management option by both vascular and orthopaedic teams as this child continues to have warm, pink hand with normal capillary refilling and minimal pain.
- In 6 months follow up, the child has gained both radial and ulnar pulses, full range of elbow motion, complete neurological recovery and good hand function.

## Introduction

Supracondylar fracture humerus is one of the most common fracture seen in paediatric orthopaedic practice and it represents up to 1/5th of all paediatric fractures. [1-3]

During the initial assessment with advanced trauma life support

protocol, once life threatening conditions has been ruled out, the distal neurovascular status is been assessed with the x-ray imaging.[4] Although neurovascular damage is relatively uncommon, it is a serious complication. Brachial artery injury following this fracture is reported between 0.4 and 11% in literature. [5, 6]

Vascular injury is assessed commonly by feeling distal pulse status and pulselessness can be of two main entities [7]. Firstly, frankly cold, pale, and pulseless hand and secondly a warm, pink hand without palpable pulses distally. Former has clear consensus for immediate fracture fixation and vascular repair [8]. However, in the case of latter- pink pulseless hand (PPH) the optimal management strategy has been widely debated. [7] [9]

In this case report, we would like to illustrate the management and follow up results of a child presented with PPH with angiogram confirmed vascular injury highlighting the diagnostic and therapeutic decisions made stepwise.

### Presentation

A 7-year-old average-built boy for his age presented to paediatric accident and emergency

unit with his parents at 11.00pm complaining of severe pain and swelling of left elbow (non-dominant) following a fall on staircase.

Immediate life-threatening conditions were ruled out and limb examination revealed a haematoma (not enlarging nor pulsating) in the cubital fossa and deformity in the left elbow. There were no radial or ulnar pulses felt and no distal saturation detected. However, the hand was warm and pink and capillary refill time was less than 2 seconds. Neurologically the child has difficulty in performing “OK sign”, but all other motor and sensory examination was normal. No signs of other soft tissue swellings or contusions noted. Clinical diagnosis of supracondylar fracture humerus with pink pulseless hand and anterior interosseous nerve damage was made.

Initial radiographs confirmed a Gartland type III supracondylar fracture humerus (fig 1).



**Fig 1 : Initial X rays**

The child’s left upper limb was put on a temporary splint to ease the pain and oral analgesics given. The limb was kept elevated with continuous monitoring for anxiety, agitation and increased analgesic requirements which are the key predictors of paediatric compartment syndrome. The child was prepared for surgical intervention on next morning casualty surgical theatre after proper counselling and obtaining informed written consent from both parents.

During surgery, about 8 hours after the presentation to A&E, the surgeon re-evaluated

the neurovascular status and it was the same as presentation. Furthermore, preoperative 8 hour stay in A&E was unremarkable.

The fracture was manipulated under image intensifier by applying axial traction followed by elbow flexion and forearm pronation. After satisfactory manipulation, plaster of Paris (POP) back slab applied holding elbow in 90 degrees of flexion and keeping forearm pronated. No K wiring or any other internal fixation method attempted. (Fig 2)



**Fig 2 : immediate X-rays after manipulation**

After the procedure, the pulse status remained the same with absent pulses and no detection of saturation, but the hand was warm, pink and capillary refilling time was less than 2s.

The child was sent to ward for further monitoring. Even after 72 hours of the procedure, the vascular status remained the same and vascular surgical opinion was sought. According

to their advice, an angiogram was done to assess the vascular status.

Angiogram revealed non visualization of flow in a short segment of 2.2cm in the left distal brachial artery at trauma site and minimal demonstrable flow distally. (Fig 3) Furthermore since, the angiogram done after 72 hours of initial injury and manipulation, the possibility of arterial spasm was considered unlikely.



**Fig 3: Angiogram**

However, despite angiogram findings, both orthopaedic and vascular teams agreed to continue vascular status monitoring without embarking on open exploration as the child was clinically

well. No heparin was administered. Child was monitored for 5 days inward and discharged.

On the 7th day postop the child was seen at vascular outpatient clinic and revealed vascular

status to be the same with no palpable pulses. However, triphasic vascular flow revealed in distal radial artery with saturation of 96%. The

child had pointing index and unable to perform OK sign. Follow up x-rays on the same day shows maintenance of satisfactory reduction.



**Fig 4 : 1 week post MUA x-ray**

Follow up x-rays in 3 weeks shows excellent fracture healing with satisfactory bony alignment. However, distal pulses were not felt. Child had a little improvement with his AIN injury. POP cast was removed in 3 weeks and encouraged elbow movements as tolerated by pain.

At 6-month follow up, the x rays show perfect bony union. Child was able to do full range of elbow movements with flexion more than 110 degrees and no extension lag. No pointing index and he was able to do OK sign with completely intact distal neurology.



**Fig 5: 6 months follow up x-ray**



**Fig 6: Child after 6 months**

## Discussion

Child with a suspected supracondylar fracture humerus following a possible high energy trauma must be assessed thoroughly to exclude immediate life and limb threatening conditions. This includes detailed neurovascular examination of the injured limb. Clinical examination coupled with initial x rays will help in determining the possible soft tissue structures at risk. For instance, a lateral displacement of fracture fragment will damage the radial nerve along with AIN and brachial artery while a medial displacement will damage the median nerve. This case demonstrated lateral displacement of fracture fragments along with radial artery and AIN damage. [10]

During the initial hours pink pulseless nature of the hand can be assumed due to local arterial spasm. Hence, watchful waiting while careful attention for major and minor vascular injury signs and impending signs for paediatric compartment syndrome is warranted. However, if its for spasm, the effect should settle with

time and during general anaesthesia since it is an adaptive response mediated by sympathetic nervous system. [11]

On the other hand, mostly the literature reveals the majority of the patients presents with PPH had brachial artery entrapment at manipulated or pinned fracture site or local brachial artery thrombosis due to intimal tear. [12]

A case reported by Choi et al, reported a similar case of PPH where fracture reduction is offered in all cases. They also noticed that the patients never had compartment syndrome. However, with signs of severe soft tissue damage such as ecchymosis and severe swelling, the risk is high. [13]

On the contrary in a review by White et al states that in their study population, 70% of PPH patients had brachial artery injury whereas only 9% had brachial artery spasm. They conclude that absence pulse should be considered as an indicator for arterial injury. [14]

Nonappearance of pulses and doppler undetectable perfusion and no detectable distal saturation after 72 hours was alarming and CT angiogram is warranted.

However, in this case the treating surgeons relied more on clinical signs than on clear vascular damage depicted on CT angiogram.

In contrast, it must be stressed that if a child is presented with pale, pulseless hand with cold extremity, then the emergency exploration with vascular team must be performed immediately.

In this patient, during reduction the axial traction is given to restore the extremity length, bowman angle, olecranon fossa shadow and

medial and lateral condylar contours. Then the elbow is flexed and assessed under image intensification whether anterior humeral line is crossing the humeral condyle in its anterior 1/3 and whether mid radial line bisects the capitellum.

Then the stability of the reduction is assessed and if stable POP back slab applied above elbow to MCP joints.

If the fracture is not stable or close reduction is not possible an internal fixation and/or open reduction with internal fixation is performed, usually by K wires. As this technique and procedure is beyond the scope of this article, it is not discussed.

### Take away lessons

- ❖ Pink Pulseless hand following supracondylar fracture humerus is a relatively common clinical scenario. However, PPH presenting with frank vascular injury is a rare entity.
- ❖ After successful and satisfactory closed manipulation, if the patient is clinically showing no features of compartment syndrome and continue to have pink, warm hand, watchful waiting is advised.

### Consent

- ❖ To write this article informed verbal consent taken from both parents of the child.

### Conflict of interest

- ❖ The authors have no conflict of interest.

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