



**Official Journal of B&B Hospital and HRDC**

**Vol. 4 | No. 1 | Jan-Dec, 2020**

# **B & B Medical Journal**

**Multidisciplinary**

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## **B & B Medical Journal**

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B & B Medical Journal (BBMed) is an official journal of B & B Hospital and Hospital and Rehabilitation Center for Disabled Children (HRDC). This journal has been brought out as a next step in the milestone of academic activities of B & B Hospital and HRDC. The main purpose of this journal is to provide a forum for scientific discussion and presentation in the field of medical science as a whole. BBMed will be a common forum for all the medical specialties dealing with the diseases of the whole human body.

Even though this journal is mainly for the medical professionals of B & B Hospital and HRDC, it accepts quality articles from anybody in medical field in Nepal and around the world.

The journal is devoted to the entire medical fraternity, patients and general public of Nepal.

It will be published once a year.

On average, a total of 10-12 articles will be published per issue. One review article from any medical field that is relevant to diagnosis and management of patients in Nepal.

There will be four to five original articles from any medical field. Priority will be given to the research articles and case series studies.

In addition, there will be four to five case reports. Priority will be given to the rare cases where a multispecialty and unique approach in the diagnosis and/or treatment has been utilized relevant to Nepalese context.

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## Published by

B & B Hospital, Gwarko, Lalitpur, Nepal

&

Hospital and Rehabilitation Center for Disabled Children (HRDC), Kavre, Nepal

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# **B&B Medical Journal (BBMed)**

*(An Official Medical Journal of B & B Hospital & HRDC)*

**Volume 4, Number 1, 2020**

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## Editorial

Editors-in-Chief

Prof. Jagdish L Baidya, FRCS, FCPS

Prof. Ashok K Banskota, MD, FACS

The differences between private and public becomes subtle when it comes to decision-making about the most appropriate venues for the training of medical personnel. A steady volume and variety of clinical material are of paramount importance. Then there is the need of committed teachers who are themselves keen students, coupling patient care with sincere efforts to update knowledge and skills. In this milieu, any interested student, with diligence, can achieve any goal he or she sets for him/herself.

Scrutiny and vigilance in the conduct of training programs in institutions are highly desirable, but these conditions should not apply selectively, as often happens in our scenario. Under the broad umbrella of “public” run institutions, many training programs continue to run with deficiencies that don’t seem to be accountable. The authorities that control various aspects of educational quality use heavy-handed and unreasonable approaches in dealing with so-called private training centers. Fair deals, easy procedural protocols, and an honest approach to genuinely promote the training process in public and private

institutions alike, would be a very positive step forward.

As the number of medical graduates increase to meet the overall requirements of the nation, the competition for the limited numbers of post-graduate teaching slots is going to be more acutely felt than ever before. The private institution and health care facilities spread throughout the country could play an important role in filling up the gaps for post-graduate training. Standardization of training facilities and ensuring a fair entrance and exit process for evaluation could play an important role in achieving a minimum uniform standard of education for all trainees at different institutions.

There are excellent examples where residency training programs in private institutions have been highly successful, initially in the USA and in the more recent past in neighboring India. Residents provide much-needed manpower for patient care and receive reasonable maintenance stipend through their period of training. The curse of capitation payments is eliminated in this system which however requires a commitment from all the parties at stake viz

the students, the teachers, and the institution wanting to participate in the training process. It is our hope that B & B hospital and HRDC can both continue to be

a part of this noble effort to provide quality services and also help address in some measure the specialized manpower requirement of the country.

# Early Outcomes of Trans-portal Anatomical Single Bundle ACL Reconstruction Using Autogenous Hamstring Graft: A Single Center Study

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Trans-portal anatomical single bundle ACL reconstruction using autogenous hamstring graft for ACL tear is a widely practiced standard procedure. From September 2015 to March 2016, 43 patients were treated for ACL tear using this technique at B&B Hospital. An analytical study of the early outcome (Post op 3 months) of the procedure is reported using Lysholm score. Different factors associated with the condition i.e. age, sex, height/weight of pt., side of the injury, BMI, mode of injury, duration of injury is analyzed.

Results were Mean Lysholm score 78.05 (13.35) with minimum of 37 and maximum of 100 at an average of three months. Age, gender, BMI and time of injury preceding surgery had no effect in the outcome after ACL reconstruction up to three months after surgery.

**Keywords:** acl tear, early outcomes, trans-portal anatomic acl reconstruction.

**A**nterior Cruciate Ligament (ACL) is the most commonly injured ligament of the Knee, and ACL tears represent more than 50% of all knee injuries.<sup>1</sup> This injury affects more than 200,000 people in the United States each year, with direct and indirect costs greater than \$7 billion annually.<sup>1</sup> Data regarding incidence and prevalence of ACL injury in Nepal is not available.

Surgical reconstruction of the torn ACL ligament is the standard treatment worldwide and has the highest success rate.<sup>2</sup> Arthroscopic trans-tibial ACL reconstruction was being done at B&B Hospital since 2001. With better understanding of anatomy and biomechanics of the Cruciate ligament, techniques of surgical reconstruction have improved. Cadaveric studies have shown that Anteromedial trans

portal drilling of the femoral socket allow better restoration of anatomy of ACL compared to conventional trans tibial drilling techniques.<sup>3</sup> Since 2015 in B&B hospital, ACL reconstruction has been carried by the anatomic transportal technique using single bundle autogenous hamstring graft.

Majority of arthroscopic surgeons worldwide prefer Hamstring as primary graft for ACL reconstruction mainly because of its ease of harvest, minimal morbidity at donor site and ease of fixation.<sup>4</sup> We also use Autogenous Hamstring graft for our primary ACL reconstruction except for some elite athlete where we prefer to use BPTB autograft for earlier incorporation of the graft and return to play.

A prospective observational study to evaluate the early outcomes of arthroscopic transportal anatomical single bundle ACL reconstruction using autogenous hamstring graft was conducted at the B&B hospital from September 2015 to March 2016.

**Materials and Methodology**

From September 2015 to March 2016, 43 patients were enrolled in the study after approval from the Institutional Review Committee (IRC) of the B&B hospital. All patients who met the inclusion criteria and Exclusion criteria were included (**Table 1**).

Data was collected preoperatively filling up a proforma and Lysholm knee scoring system at 3 months after surgery.

**Treatment Protocol**

All the patients underwent routine preoperative imaging, including plain radiographs anteroposterior (AP) and lateral view and MRI, of the knee.

Prophylactic antibiotic intravenously (cefazolin injection 1g 30-60 minutes before surgery) was administered. Surgery was performed under either Spinal anesthesia or General anesthesia under a tourniquet control. Patient was operated in Supine position using a standard anterolateral, anteromedial and far accessory anteromedial portal. Semitendinosus (ST) and Gracilis (GT), were used to make a quadrupled graft to make the graft diameter size of 8 mm or more and at least a length size of 8 to 9cm. The graft was fixed with an endobutton on the femoral side, and a bio absorbable interference screw on the tibial side. Torn meniscus was repaired using the outside-in technique with the suture-shuttle technique using orthocord (braided metal) sutures. Partial meniscectomy was performed for irreparable menisci.

**Postoperative rehabilitation**

Postoperative rehabilitation was started on the first postoperative day. Knee ROM was started doing heel slides aiming to achieve 90 degrees flexion within 2 weeks and 120 degrees

<b>Inclusion criteria</b>	<b>Exclusion criteria</b>
1. Patients undergoing transportal anatomic single bundle ACL reconstruction using hamstring autograft.	1. Patients who had previous knee surgeries
2. Age 17-58 years	
3. Patients providing written consent	

*Table 1: Inclusion and exclusion criteria*

gradually within 6 weeks. The patient was advised for ankle pump, static quadriceps exercises and heel hanging to achieve neutral extension. Pts were discharged on the 5<sup>th</sup> post op day after gaining 90 degrees flexion. Patients were advised for ambulation with crutches and use of hinge braces. Crutches were discarded in 4 to 6 weeks. However, weight-bearing were avoided for 4 to 6 weeks if the patient underwent meniscal repair. Patients were followed up every 6 weeks to assess the progress for initial 3 to 6 months.

**Data Analysis**

SPSS version 20 was used for data analysis. Frequency and percentage were calculated for (1) Gender, (2) Good to Excellent Outcome and (3) BMI. Mean and standard deviation was calculated for (1) age of patient, (2) duration of injury, (3) Lysholm score, (4) height, and (5) BMI. Effect modifiers like age, gender, duration of injury and BMI were dealt with using post-stratification. Chi square test was applied. P-value of <0.05 was taken as significant.

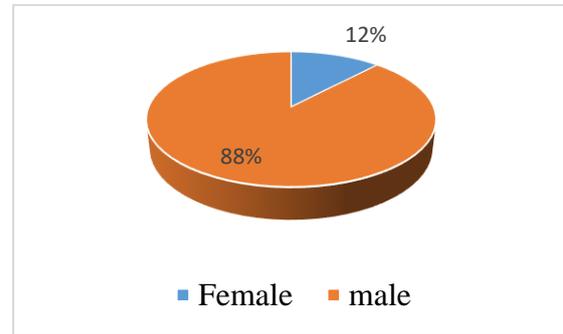
**Results**

A total of 43 patients were included in this study. One patient was lost to follow up. Stratification was done based on the age, gender, duration of injury and Body Mass Index (BMI) of the patients.

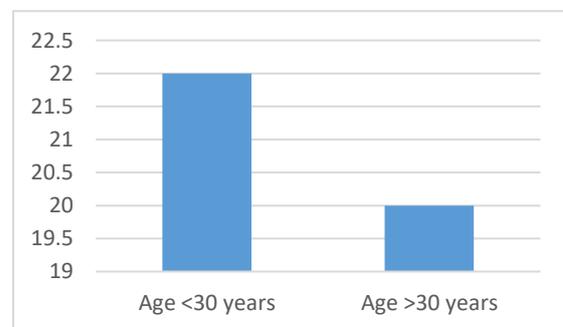
Male predominance was found in patients who undergo ACL reconstruction: 88% of the patients were male and 12% were female shown in **Figure 1**.

**Figure 2** shows stratification of the participants

according to their age group: 52% were aged below 30 years and 48% were aged above 30 years.



*Figure 1: Gender wise distribution of the participants (n=42)*



*Figure 2: Age wise distribution of the participants*

**Table 2** shows baseline characteristics of the patients who underwent anatomic ACL reconstruction. The mean age of treatment for ACL reconstruction in this study is 31.3 years (SD 8.66) with minimum age being 17 years and maximum 58 years. Mean height of the patients was 164.8 (SD 7.27) with minimum height of 150 cm and maximum height of 177 cm. Mean weight was 67.38 kg (SD 9.53) with minimum of 54 kg and maximum of 88 kg. It was found that the patients presented very late at a mean of 28.48 months (SD 54.44) with minimum time of 36 days and maximum 20 years. Mean BMI was 24.76 (SD 2.71) with minimum BMI 19.71 and maximum 29.94. The study had only one patient who belonged to

Variables	Minimum	Maximum	Mean	SD
Age (in years)	17	58	31.33	8.66
Height (in cm)	150	177	164.80	7.27
Weight (in kg)	54	88	67.38	9.53
BMI (kg/m <sup>2</sup> )	19.71	29.94	24.76	2.71
Time of presentation (months)	0.1	240	28.48	54.44

Table 2: Baseline characteristics of participants (N=42)

	Frequency	Percentage
Medial Meniscus	12	28.5
Lateral Meniscus	7	17
Isolated ACL	20	48
MCL, LCL	3	7
PCL	0	0

Table 3: Associated injuries confirmed in Arthroscopy

	Frequency	Percentage
Excellent (95-100)	4	10
Good (85-94)	11	26
Fair (65-84)	24	57
Poor (<65)	3	7

Table 4: Lysholm Scoring at average of 3 months post-op

obese (BMI >30 subgroup). Majority of the patients (57.14%) were within the normal weight range (BMI 18.5 – 24.9) and 42.86% were overweight (>25-30).

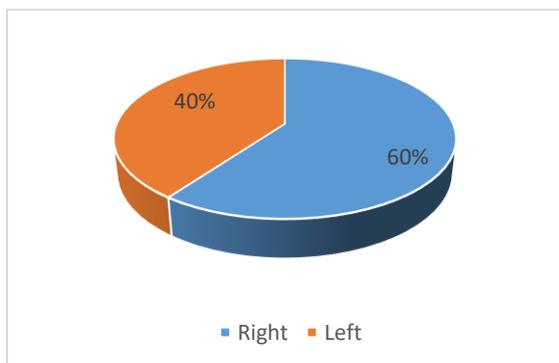


Figure 3: Side of injury

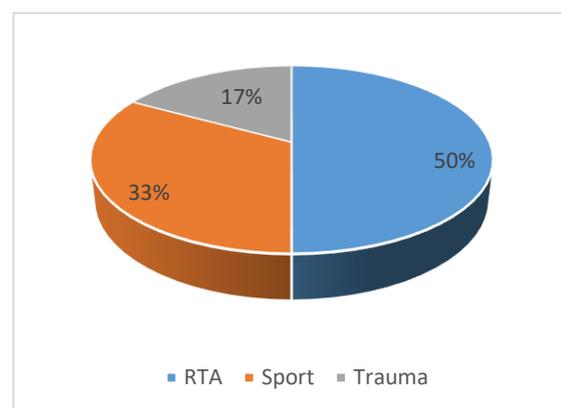


Figure 4: Mode of injury

Figure 3 shows that right side ACL injuries were common than left side ACL injuries. The causes show that 50% were due to road traffic

Age group	Lysholm score				Total	Chi square	P-value
	≥95	85-94	65-84	<65			
≤30	3	6	11	2	22		
	13.6%	27.3%	50%	9.1%	100%		
>30	1	4	14	1	20	2.003	0.596
	5%	20%	70%	5%	100%		
<b>Total</b>	4	10	25	3	42		

Table 5: Outcome analysis based on age of the patients

accidents (RTA), 33% were due to sports injuries, and the remaining 17% were due to trauma shown in **Figure 4**.

**Table 3** shows that ACL injuries occurred in isolation in 48% of the cases, while 45.5% had associated meniscal injuries. The remaining 7% cases had collateral injuries in this study.

Mean Lysholm score was 78.05 (13.35) with minimum of 37 and maximum of 100 at an average of three months.

It was found that majority of the patients (57%) had fair results followed by good results (26%) while 10% had excellent results and 7% of participants had poor results shown in **Table 4**.

**Table 5** shows that patients aged less than 30 and more than 30, both had fair to good results in the highest proportion of patients. In patients aged less than 30, the results were excellent in 13.6%, good to fair in 77.3%, and poor in 9.1%. In patients aged more than 30, the results were excellent in 5%, good to fair in 90% and poor in 5%. Age did not have a significant effect in the outcome after ACL reconstruction (p=0.596).

**Table 6** shows Lysholm score in female and male patients. All female patients (100%) had fair to good results, but no female patients had

excellent or poor results. In male patients, 11.1% had excellent outcome, 81% had fair to good results, and 8.3% had poor results. Gender did not affect the outcome.

Patients within the normal weight range (BMI 18.5-24.9) had fair to good results in 82% of the cases. It was excellent in 13% while only 4% had poor results. Similarly, patients who were overweight had excellent results in 5.9%, good to fair results in 81.5% and 11.8% had poor results. BMI did not affect the outcome (**Table 7**).

**Table 8** shows outcome after ACL reconstruction depending on the time the patients presented after injury. When patients presented less than six months after ACL injury, 9.5% had excellent results and 90.5% attained good to fair results. When the patients presented from six months to a year after ACL injury, 20% had excellent results and 80% had good to fair results. Presentation after one year eight weeks following the implantation is critical as the hamstring tendon graft increases strength and stiffness<sup>14-16</sup> and the muscle follows specific biochemical, mitochondrial and neurological adaptations.<sup>17,18</sup> However, the eight-week duration has also been considered

Sex	Lysholm score				Total	Chi square	P-value
	≥95	85-94	65-84	<65			
Female	0	1	5	0	6	1.983	0.532
	0%	16.7%	83.3%	0%	100%		
Male	4	9	20	3	36	1.983	0.532
	11.1%	25%	55.6%	8.3%	100%		
<b>Total</b>	4	10	25	3	42		

Table 6: Outcome analysis based on gender

BMI group	Lysholm score				Total	Chi square	P-value
	≥95	85-94	65-84	<65			
18.5-24.9	3	6	14	1	24	1.239	0.765
	13%	21.7%	60.9%	4.3%	100%		
≥25	1	5	10	2	18	1.239	0.765
	5.9%	23.5%	58.8%	11.8%	100%		
<b>Total</b>	4	11	24	3	42		

Table 7: Outcome analysis based on BMI

Duration	Lysholm score				Total	Chi square	P-value
	≥95	85-94	65-84	<65			
≤6 months	2	3	16	0	21	9.323	0.152
	9.5%	14.3%	76.2%	0%	100%		
6 months- 1 year	1	2	2	0	5	9.323	0.152
	20%	40%	40%	0%	100%		
>1 year	1	5	7	3	16	9.323	0.152
	6.2%	31.2%	43.8%	18.8%	100%		
<b>Total</b>	4	10	25	3	42		

Table 8: Lysholm score based on time of presentation

Complications	Frequency	Percentage
Laxity grade I (hard end point)	26	78.8%
Anterior skin pain and numbness	3	9.1%
Extension Lag (5 degrees)	2	6.1%
Graft donor site pain	1	3%
Clicks	1	3%

Table 9: Complications

as a critical time in establishing strength or more of ACL injury showed excellent results in only 6.2%, fair to good results in 74%, and 18.8% had poor results. However, it was not statistically significant.

Grade I Lachman test was found in 62% of cases with hard end point, anterior knee pain in 7% cases, extension lag in 4.6% cases, graft donor site pain in 2.3% cases, and clicks in knee in 2.3% cases in **Table 9**.

### Discussion

An ACL injury predisposes a patient to significant disability and untreated will lead to subsequent meniscal tear, chondral damage in the long run and thus is important to reconstruct the ligament. The choice of graft varies upon the surgeon's expertise and options available. ACL reconstruction success rate largely depends on technical expertise, choice of graft and its size, tunnel position and graft fixation and post-operative rehabilitation. Outcome goals should be consistent although the choice of graft is surgeon dependent. Mascarenhas et al with the systemic analysis showed that there is no difference between allograft and autograft in graft failure rate and clinical outcome, but the same study with a lower quality meta-analysis showed that allograft has a higher failure rate.<sup>5</sup> The most commonly chosen grafts are bone patellar tendon bone or hamstring tendon. Many studies showed similar outcomes between these two grafts. Princzewski showed OA changes after BTB (18%) autograft over Hamstring graft (4%).<sup>6</sup>

There are many other studies which show no change in long term result of BTB vs. hamstring autografts. In short term studies, there is less post-operative stiffness and faster recovery with hamstring autograft. However, taking hamstring graft is not free from complications like injury to infrapatellar branch of saphenous nerve, infection, flexion/extension deficit, recurrence of instability, intra-articular adhesions, hemarthrosis, and painful hardware. Similarly, there is risk of patellar fracture and anterior knee pain with BTB autograft.<sup>7-9</sup>

There are no significant differences between double bundle and single bundle ACL reconstruction in terms of function, translation, and complications rates.<sup>10</sup> An anatomic orientation of ACL reconstruction with femoral tunnel drilled from anteromedial port provides better tibial translation.<sup>11,12</sup>

Our study had 42 patients who underwent trans-portal anatomic ACL reconstruction using quadrupled hamstring autograft. The graft was anchored with interference screw on the tibial side and an Endobutton on the femoral side. The participants of the study included 88% male patients and 12% female patients aged between 17 and 58 years old. The mean age was 31 years. The injury involved 59.9% on the right side and 39.5% on the left. In contrast to other studies like Hewett et al, our study showed that 50% of ACL injury was due to RTA, 33% due to sports injury and 16.3% due to other kind of minimal trauma.<sup>13</sup>

This study had a short follow up and tended to measure functions using Lysholm score after

three months of average follow up. The first stimulus on weak quadriceps muscle following ACL reconstruction.

There are few studies which have been conducted to show Lysholm score after three months of surgery. Largely these studies have reported in context to show efficacy and adequacy of the type of muscle strengthening exercises of the quadriceps muscle. The rating system of Lysholm questionnaire is well established as an alternative mechanism to gather data on outcome when evaluating knee ligament injuries.<sup>19,20</sup>

A study by Maria et al<sup>21</sup> showed that at eight weeks after ACL reconstruction using hamstring graft with cross eccentric exercise, the Lysholm score was 92.95(+/- 4.35) at three days per week quadriceps strengthening exercise, and 90.57 (+/- 6.16) with five days per week quadriceps strengthening exercise with statistical significance.

The study by Bitun et al<sup>22</sup> measured Lysholm score and IKDC score in patients treated with patellar tendon and hamstring tendon grafts after ACL reconstruction in one month, three months and six months, with an interference screw on both tibia and femur. The Lysholm score was 71.6 after one month, 89.1 after three months and 96.6 after six months in patients who were treated with hamstring autograft.<sup>22</sup> The patients in our study had a mean score of 78.05 after three months. Therefore, these results are slightly inferior, this may be due to Endobutton anchorage on femoral side and may improve when the graft attaches to the femur in the future during the recovery process.

Those who had ACL reconstruction performed

with the anteromedial portal for femoral tunnel, which makes tunnel more anatomical, had returned to athletic activities a month and a half prior to those who had the reconstruction performed using the trans tibial technique. In contrast, no difference has been noted between the groups that underwent ACL reconstruction utilizing tunnels either through the trans tibial technique or through the anteromedial tunnel using BTB graft, in terms of VAS scale for pain, Lysholm, Tegner, and SF-12.<sup>23</sup> The femoral tunnel was made using anteromedial portal for our study as well. A longer follow up time is required to assess when the participating patients will return to their pre-injury status. Our study did not include ACL reconstruction in athletes who wanted to return to sports. Our study shows if age, sex, body mass index, and the time of presentation affect the Lysholm score and association between delayed ACL reconstruction and changes in other structures inside the knee joint with delayed presentation. A greater exposure to strenuous environments amongst males makes the ACL injury incidence higher in the male population as explained by Brown et al.<sup>24</sup> Likewise, in our study, 88% of the patients were male and only 12% were female. In a study by D. Ferrari et al found no difference in outcome in male and female patients with BTB graft.<sup>25</sup> However, they found slight increased difference for quadrupled hamstring grafts (0.87 v 2.46mm). This could directly influence functional outcome in female patients who undergo ACL reconstruction using autogenous hamstring graft. Hence, female patients could have inferior results. However, there was no statistically significant

Lysholm score difference. Our study had a smaller number of female participants (n=5) than the Male participants (n=37), and results could have been masked. The result of our study is consistent with the results of the study conducted by Aldhen et al who found that there was no significant difference between male and female in Lysholm score after ACL reconstruction using hamstring tendon in larger series of 141 male and 103 female participants in a two year follow up.<sup>26</sup>

Our study found no difference in outcome after ACL reconstruction using quadrupled hamstring autograft between patients with a normal BMI and a high BMI. This study was consistent with Ballal et al<sup>27</sup> which comprised of two groups of patients: the first 49 patients with normal BMI (18.5-24.9) and the second of 43 patients with a high BMI (>25). They matched mean age, male to female ratio, injury side, smoking status, duration before surgery, and same surgical technique with the same surgeons. They measured Lysholm score at 3,6 and 12 months. The score was 75.66, 87.17, and 88.12 respectively. They found no difference in the outcome.<sup>27</sup> In our study, 57% of the patients had normal BMI, remaining 43% had a high BMI. Only one patient had BMI >30. This study showed that there was no difference in outcome up to three months after surgery. More than 95% had fair to excellent results with normal BMI and almost 90% had fair to excellent results. But it was not statistically significant.

Some studies have shown an increased complication risk in patients with high BMIs, showing that patients with high BMIs have an

increased prevalence of wound complications and symptomatic venous thromboembolism.<sup>28,29</sup>

### Conclusion

Age, gender, BMI and time of injury preceding surgery had no effect in the outcome after ACL reconstruction up to three months after surgery. This study found higher meniscal injuries with late presentation in up to 45% patients in the study with mean time of presentation at 28.4 months +/- 54.4. In this study, the mean Lysholm score was 78.05 +/- 13.35 in three months after anatomic ACL reconstruction using quadrupled hamstring autograft. Thus, anatomic ACL reconstruction using quadrupled hamstring autograft is a safe operation with minimal complications, and it improves quality of life. However, longer follow up is required to see how these patients perform and to establish the result of ACL reconstruction.

### Limitation

It is a short follow up study of only three months. To see and establish the outcome of anatomic ACL reconstruction, a longer follow up is necessary. Besides, there can be many variables like patient occupation, history of smoking, patient taking some drugs, which can have different outcomes, which is not considered in this study. Using Lysholm score in the context of Nepal is also difficult as it was designed for people from western part of the world with different lifestyle. People are considerably less involved in athletic activities in Nepal which is a part of Lysholm questionnaire. So it sometimes created dilemma

while scoring. That difference might affect the results of this study. This study involves heterogenous group of people so study could have been improved by including a homogenous population and increasing the sample size.

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## Hydrocortisone, Vitamin C, and Thiamine for Treatment of Sepsis and Septic Shock: A Single Centre Retrospective Comparative Study

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**Background:** The global burden of sepsis is estimated to be 19 million cases annually, with mortality rate approaching 26 % in high-income countries and 50% in low-income countries

**Methods:** In this retrospective comparative study we looked at the outcome of consecutive septic patients treated with intravenous vitamin C, hydrocortisone, and thiamine over 6 months duration (treatment group) with a control group treated in our ICU (Intensive Care Unit) during preceding 6 months. The primary outcome was hospital mortality.

**Results:** There were 24 patients in treatment group and 30 patients in control group with no significant difference in baseline characteristics between the two groups. The hospital mortality of sepsis was 7.7% (1 of 13 patients) in the treatment group compared with 13.3 % (2 of 15 patients) in the control group (P value = 0.63) and hospital mortality of septic shock was 27.3% (3 of 11 patients) in the treatment group compared with 40% (6 of 15 patients) in the control group (P value = 0.52)

**Conclusion:** Our results suggest that there was no statistically significant reduction in mortality with early use of intravenous vitamin C, hydrocortisone and thiamine in sepsis and septic shock.

**Keywords:** hydrocortisone, sepsis, septic shock, thiamine, vitamin C.

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The global burden of sepsis is enormous with an estimated 19 million cases per year, majority of these cases occurring in low-income countries.<sup>1</sup> With timely diagnosis and improvement in supportive care, the 28-day mortality of sepsis in high income countries has declined to about 26%.<sup>1</sup> But it remains very high in low-income countries, approximately 50%.<sup>2,3</sup>

Over the last three decades, more than 100 phase 2 and phase 3 clinical trials have been performed testing various novel pharmacological agents and therapeutic interventions to improve the outcome of sepsis and septic shock; all these efforts ultimately failed to produce a novel pharmacological agent that improved the outcome of sepsis.<sup>4</sup>

Marik PE and colleagues have shown in their before and after study that early use of intravenous vitamin C, hydrocortisone and thiamine in sepsis reduced mortality from 40.4% to 8.5% ( $P < 0.001$ ).<sup>5</sup> This study was conducted to find out whether early use of intravenous hydrocortisone, vitamin C and thiamine reduced mortality in sepsis and septic shock in our setup.

## Methods

This study was done in B and B Hospital, Kathmandu, from June 2017 to June 2018. Patient consent was taken. All patients

more than 14 years of age admitted to ICU (Intensive Care Unit) from January 2018 to June 2018 with a primary diagnosis of sepsis and septic shock were treated with intravenous hydrocortisone, vitamin C and thiamine within 24 hours of ICU admission (treatment group). The control group consisted of consecutive patients above 14 years of age admitted in the ICU between June 2017 and December 2017 with primary diagnosis of sepsis and septic shock. During control period, patients with sepsis did not receive intravenous vitamin C or thiamine. The diagnosis of sepsis and septic shock were based on the 2016 The Third International Consensus Definition for Sepsis and Septic Shock.<sup>6</sup>

The overall treatment of sepsis and septic shock during control and treatment periods was similar except for the administration of combination of vitamin C, hydrocortisone, and thiamine during the treatment period. During control period, patients received hydrocortisone (50 mg I.V every 6 hours) at the discretion of attending physician. During the treatment period, all patients with primary diagnosis of sepsis and septic shock were treated with intravenous vitamin C (1.5gm every 6 hours for 5 days), hydrocortisone (50 mg every 6 hour for 7 days which was tapered over 3 days), and intravenous thiamine (200 mg every 12 hours for 5 days).

The demography, etiology and outcome of

the patients between the two groups were analyzed. The end point of the study was discharge from the hospital. All the categorical variables were expressed in number and percentage, while continuous variables were expressed in mean  $\pm$  SD or median (range). Statistical analysis was done using SPSS 14.0 for windows.

**Results**

There were 24 patients in the treatment group and 30 patients in the control group. The baseline characteristics of the two groups are given in **Table 1**. There were no

significant differences in baseline characteristics between the two groups. Most patients had multiple co-morbidities. The causes of sepsis were similar in both the groups, chest infection being the commonest. The hospital mortality of sepsis was 7.7% (1 of 13 patients) in the treatment group compared with 13.3 % (2 of 15 patients) in the control group (P value = 0.63). And the hospital mortality of septic shock was 27.3 % (3 of 11 patients) in the treatment group compared with 40% (6 of 15 patients) in the control group (p value = 0.52). (**Table 2**) (**Figures 1 and 2**).

Variables	Treatment group (n=24)	Control group (n=30)	p value
Age, mean $\pm$ SD years	58.9 $\pm$ 18.3	60.9 $\pm$ 18.7	0.69
Gender, No (%)			0.41
Male	10 (41.7%)	17 (56.7 %)	
Female	14 (58.3%)	13 (43.3%)	
Co morbidities, No (%)			
Hypertension	14 (58.3 %)	14 (46.7%)	0.40
COPD	16 (66.7 %)	16 (53.3 %)	0.32
Diabetes mellitus	10 (41.7 %)	10 (33.3 %)	0.53
CKD	3 (12.5 %)	5 (16.7 %)	0.67
Heart Failure	2 (8.33 %)	4 (13.3 %)	0.56
Primary Diagnosis, No (%)			
Chest Infection	14 (58.3 %)	17 (56.7 %)	0.91
Urosepsis	5 (20.8 %)	7 (23.3 %)	0.83
Intra-abdominal sepsis	0 (0 %)	3 (10 %)	0.11
Skin and soft tissue infection	3 (12.5 %)	0 (0 %)	0.05
Tropical Sepsis	2 (8.3%)	3 (10 %)	0.83

*Table 1: Baseline Characteristics of patients in Treatment and Control groups*

Outcome	Treatment Group (n=24)	Control Group (n=30)	p value
Sepsis, n (%)	13 (54.2%)	15 (50%)	NA*
Mortality in sepsis, n (%)	1 (7.7%)	2 (13.3%)	0.63
Septic shock, n (%)	11 (45.8 %)	15 (50%)	NA*
Mortality in septic shock, n (%)	3 (27.3 %)	6 (40%)	0.52

Table 2: Outcome of patients in treatment group and control group

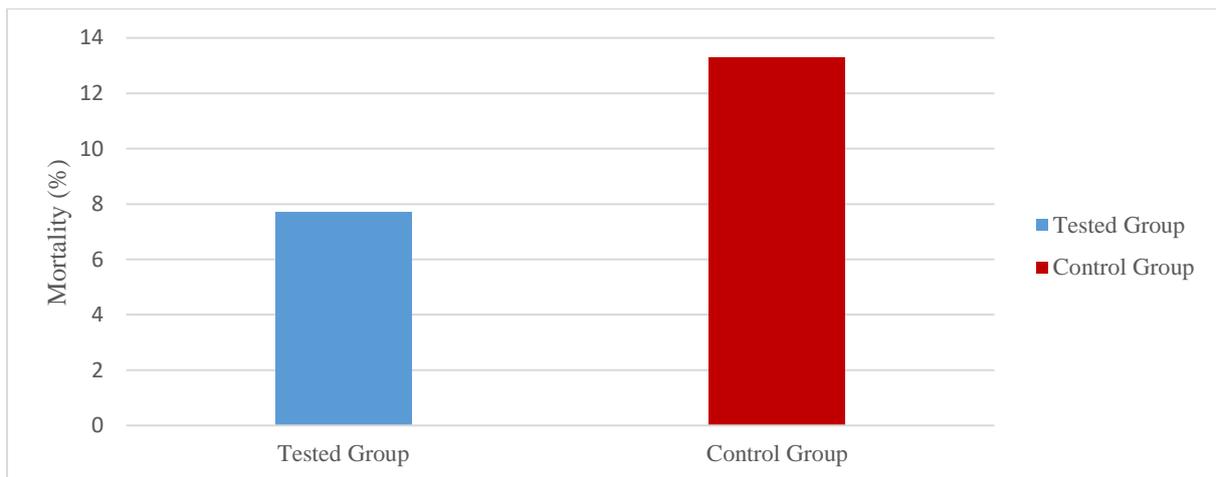


Figure 1: Mortality of sepsis in the treated and control group

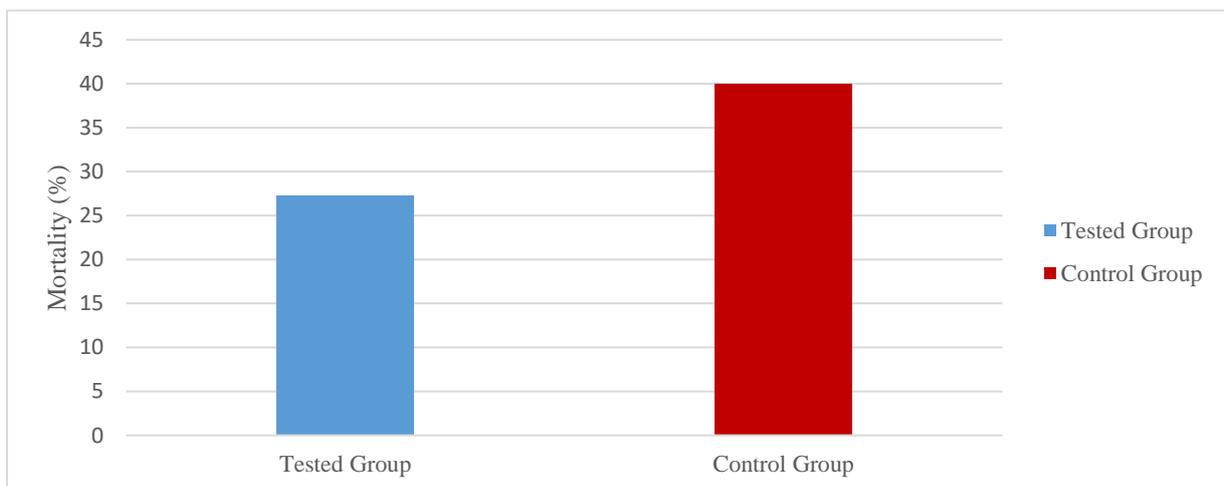


Figure 2: Mortality of septic shock in the treated and control group

**Discussion**

Critically ill patients, including septic patients, have critical illness related corticosteroid insufficiency (CIRCI).<sup>7</sup> Steroid has multiple beneficial effects in sepsis. It inhibits Nuclear Factor - kappa B,

the excessive stimulation of which causes cytokine storm in sepsis. It recruits microcirculation and re-sensitizes catecholamine receptors.<sup>8</sup> But, hydrocortisone did not improve survival in septic shock in CORTICUS<sup>9</sup> and

ADRENAL<sup>10</sup> trials. Steroid alone does not work well in sepsis, as steroid receptor gets oxidized in oxidative state like sepsis and the receptor loses its affinity for agonistic ligand and binds with the antagonistic ligand. This brings about a conformational change in the receptor, because of which steroid cannot bind with the receptor to exert its effect.<sup>11</sup> Vitamin C reduces the cysteine thiol groups of steroid receptors and displaces the antagonistic ligand and primes the receptor.<sup>12</sup>

Vitamin C also acts through multiple pathways in sepsis and has multiple overlapping effects with steroid.<sup>13</sup> It is required for synthesis of catecholamines and corticosteroids in the adrenal glands. It is required for synthesis of vasopressin and neurotransmitters. It is required for the functioning of T - cells and macrophages; it is an inhibitor of Nuclear Factor - kappa B and it maintains the integrity of tight junction.<sup>14,15,16</sup> Almost all septic patients have vitamin C deficiency,<sup>17,18</sup> but oral administration of vitamin C as high as 1.5 gm/day cannot restore vitamin C levels in patient with sepsis due to saturable gastrointestinal transporter, sodium - vitamin C co-transporter 1 (SVCT - 1).<sup>19</sup> To achieve normal vitamin C levels in critically ill patients, a daily dose of more than 3 gm is required.<sup>17, 20, 21</sup>

Thiamine deficiency is common in septic patients and is associated with an increased

risk of death.<sup>22</sup> Hyperoxaluria sometimes results in patients with renal impairment receiving megadose of vitamin C<sup>23</sup> and thiamine diverts metabolism away from oxalic acid synthesis.<sup>24</sup>

Although Paul E. Marik and colleagues have shown that early use of intravenous vitamin C, hydrocortisone and thiamine in sepsis reduced the mortality of sepsis from 40.4 % to 8.5 % ( $P < 0.001$ )<sup>5</sup>, our study has shown that there was a decline in mortality rate of sepsis from 13.3 % to 7.7% ( $p = 0.63$ ) and septic shock from 40% to 27.3% ( $p = 0.52$ ), but it did not achieve statistical significance.

There are three ongoing large multi centric trials, VITAMINS<sup>25</sup>, ACTS<sup>26</sup> and VICTAS<sup>27</sup>. VITAMINS is a multi-centric Australasian study comparing intravenous vitamin C, hydrocortisone, and thiamine with intravenous hydrocortisone alone in septic shock with vasopressor free days as the primary end point and 90-day mortality as the key secondary end point. ACTS is a multi-centric US study comparing intravenous ascorbic acid, hydrocortisone, and thiamine with placebo in septic shock with change in SOFA<sup>28</sup> (sequential organ failure assessment) score at 72 hours as the primary end point and incidence of kidney failure and 30-day mortality as the key secondary end points. VICTAS is a multi-centric US study comparing the combination of intravenous vitamin C,

hydrocortisone, and thiamine with placebo in sepsis with ventilator- and vasopressor-free days as the primary end point and 30-day mortality as the key secondary end point. We need to wait for the results of these RCTs for concrete evidence.

### **Conclusion**

The result of our study suggests that there was no statically significant reduction in mortality with early use of intravenous vitamin C, hydrocortisone and thiamine in sepsis and septic shock. However, larger multicentric studies are required to confirm our results.

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## Size of the Tibial Footprint of Anterior Cruciate Ligament: MRI Study

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**Introduction:** Anatomic ACL reconstruction (ACLR) is the surgical treatment for an ACL tear. Every individual is different and so is their ACL thus one size will not fit all. There is a concept of individualized ACLR. The occupancy of adequate footprint area by the graft has been a major determinant of individualized ACLR. For adequate representation, 50-70 % of the footprint must be covered by the graft. This is usually managed with single-bundle ACLR in footprint size 16 mm or less whereas a size more than 16 mm requires double-bundle ACLR. Many techniques have been described for the measurement of footprint size. Preoperative MRI measurements have shown comparable results with intraoperative direct measurement of footprint. A study of similar nature in our population is lacking. Thus, the aim of this study was to find out the footprint size in our patient population.

**Methods:** This was a MRI-based study. A total of 230 knee MRIs done for various indications were selected for the study. 15 of those MRIs were excluded as the footprint was not measurable or ACL had intrinsic pathology. Sagittal T2 weighted image showing the largest portion of the ACL footprint of the tibia was selected for the study. A line was drawn from most anterior and posterior points of the footprint parallel to the tibial plateau. Measurement was done using Radiant Dicom viewer and recorded. Analysis was done using SPSS 23.

**Results:** Out of a total of 215 cases, the mean footprint of our population was found to be 14.1 mm with a range of 10.8 - 17.6 mm. The footprint was found to be smaller in the female population compared to males (13.6 mm vs 14.7 mm) and this difference was found to be statistically significant.

**Conclusion:** The average footprint size of our study population is 14.1 mm. The size of the footprint is significantly smaller in females compared to males.

**Keywords:** anterior cruciate ligament, footprint, individualised ACLR.

**I**ndividualized anatomic ACL reconstruction has been established as a standard of care for individuals with an ACL tear. The concept behind individualized reconstruction is every individual is different and so is their ACL. So, one size won't fit all and it is necessary to change various parameters like the size of the graft, placement of the graft, number of bundles to reconstruct according to the preoperative and intraoperative evaluation of torn ACL.<sup>1,2</sup> One of the very important determinants in individualized ACLR is the size of the tibial footprint which will guide the size of the graft to be used during the reconstruction. Freddie Fu has recommended that graft should occupy 50-70% of the tibial footprint for better clinical results. Among various dimensions of footprint, the anteroposterior length of the footprint has been the most consistent measure.<sup>1,3</sup> Guidelines and tables for optimal graft sizes have been given in accordance with this variable. Length >16 mm will need double-bundle ACL reconstruction and single-bundle ACL reconstruction will be sufficient in footprint <16mm.<sup>4</sup> Multiple studies have been published which measured the length of tibial footprint with different results. Both preoperative and intraoperative measurements have been studied and compared.<sup>5</sup> Kim et al showed that preoperative measurement of the length of footprint in MRI correlates well with intraoperative management and recommended MRI measurement of footprint dimension for preoperative planning.<sup>6</sup>

Also, it has been shown that the size of ACL differs from the size of the knee which has been reflected in the size of ACL footprint too. Studies from the Asian population reported a smaller footprint compared to the western population.<sup>7,8</sup> We do not have data showing the footprint length of the Nepalese population. With this study, we aim to achieve the average footprint size of the Nepalese population.

### **Methods**

Random 230 MRI of knee done for various indications which were performed in single-center with same machine (1.5 tesla) with same settings were retrieved. While selecting age less than 10 years and more than 60 years were not included. All the images were evaluated at first to see the ACL footprint. Those with mucoid degeneration of ACL, osteophytes around footprint, showing signs of past trauma or surgery were excluded from the study. After exclusion, 215 cases were included for further evaluation of ACL footprint. We used RadiAnt Dicom viewer for measurement (**Figure 1**). To measure the exact length of the tibial footprint, a sagittal image showing the largest exposure of the ACL fibers at the tibial attachment site was chosen. The length of the ACL tibial footprint was measured as the distance from the most anterior to the most posterior fibers of the ACL tibial attachment. The anteroposterior length was done parallel to the tibial plateau (**Figure 1**). The sequence used was a proton density-weighted sagittal image. Results were recorded



Figure 1: RadiAnt dicom viewer. Anteroposterior length of ACL footprint measured with the line parallel to tibial plateau joining most anterior and posterior point of footprint

in Microsoft excel and further analysis was done with SPSS.

**Results**

Out of a total of 215 cases, the mean anteroposterior length of the tibial footprint of ACL was found to be 14.1 mm with range 10.8 - 18.8 mm (Figure 2).

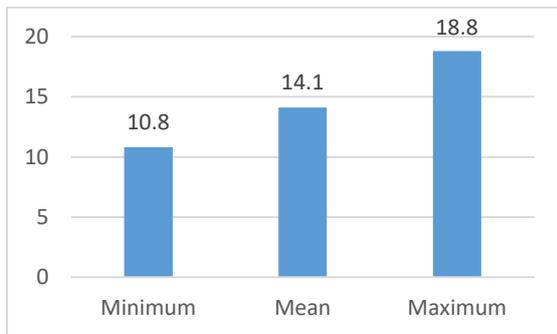


Figure 2: Size of tibial footprint

The footprint was found to be smaller in the female population compared to males (13.6 mm vs 14.7 mm) and this difference was found to be statistically significant (Figure 3).

The mean age of chosen patients was 24.3 years (range 15 – 58 years). The majority were male

comprising 52% of the total.

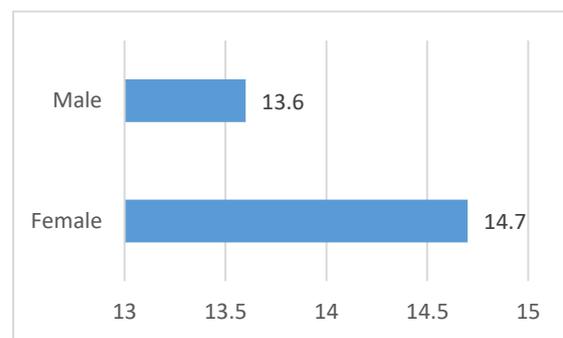


Figure 3: Male vs Female

**Discussion**

Freddie Fu with his multiple research popularized the concept of individualized anatomical ACL reconstruction. According to him, anatomical ACLR means functional restoration of ACL in its native dimension, collagen orientation, and insertion sites.<sup>1,2,9,10</sup> Insertion sites (footprint location and size) is a very important determinant for successful reconstruction according to Fu et al. Adequate representation of ACL footprint according to the studies is to cover at least 50-70% of the total footprint. Accordingly, the algorithms and

tables have been described for individualized anatomic ACL reconstruction with reference to the size of a tibial footprint. Anatomical studies of ACL have shown that ACL is the widest at the tibial insertion site. The femoral footprint comprises up to 70% of the area of tibial insertion and the substance of ACL constitutes only about 50% of tibial insertion. This shows that the tibial insertion footprint is the single most important measurement required for preoperative planning of individualized ACLR.<sup>4,10</sup> Anteroposterior length along with mediolateral width and area of the footprint has been studied in various studies both preoperatively with MRI and with intraoperative direct measurement. Anteroposterior length of the footprint is one of the most consistent variables that is measured preoperatively with MRI. Mediolateral width measurement requires oblique sequences which is not the usual sequence of MRI of the knee also like the shape of the tibial footprint is variable the measurement of the area also is not a consistent MRI variable to measure the dimension of the tibial footprint of ACL.<sup>5,7,8</sup>

In this research we also measured the anteroposterior length of the tibial footprint of ACL, we followed the technique used by Kim et al.<sup>7</sup> Average length of tibial footprint in our study is 14.1 mm which is similar to the study by Kim et al. They reported average length of 12.4 mm. Park et al,<sup>7</sup> similar size of the footprint was reported in Chinese study too by Feilong li et al.<sup>11</sup> in contrast studies from western population reports larger footprint.<sup>12</sup> This may be because of smaller knees in the Asian population which will have ACL with

smaller dimensions.

Our study also shows a shorter length of footprint in females compared to the male population which was found to be statistically significant. This finding is similar to many published Asian and western literature. If we follow the principles of individualized anatomical ACLR, considering the average size of footprint in our population being 14.1 mm, we can safely make a conclusion that most of the Nepalese population won't be needing double-bundle ACLR and the optimal size of graft comes around 8-9 mm diameter. Still, it's always preferable to have a preoperative MRI measurement of footprint and make individualized decisions on graft size.

### **Conclusion**

The average size of the tibial footprint of ACL in our study is 14.1 mm (range 10.8 – 17.6 mm). The footprint is shorter in females. Preoperative measurement of anteroposterior length of the tibial footprint is an easy and helpful tool for preoperative planning of individualized anatomical ACL reconstruction.

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## Disability Orientation Outcomes in Three Districts of Nepal: An Interventional Study

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**Introduction:** Disability orientation in the community can have positive effects in timely identification and treatment seeking behavior, as well as help mitigate the stigma that often burdens persons with disabilities (PWDs).

**Objective:** To assess the outcome of disability orientation in the community in three purposively selected districts of Nepal.

**Materials and Methods:** The study was conducted in three purposively selected project districts of Nepal (Rupendehi, Arghakhanchi and Gulmi). Fifty percent of the municipalities were randomly selected from each district. Twenty-five persons from each municipality were randomly enrolled in the study as study participants. Hence, the study was conducted among 425 participants in total. A community level course about disability awareness followed a structure focusing on awareness, interaction with PWD, and education, in order to make a positive impact on their knowledge. Pre-and post-test interviews were conducted before and after disability orientation at a 2-week interval.

**Results:** There were 52% female participants, and 44% were between 38 to 47 years. 44% had completed at least ten years of schooling, and 3% could not read or write. The average increase in the level of knowledge regarding causes of disability, rights and entitlements of the persons with disability and the preventive measures of disability were by 24%, 52% & 40% respectively. The overall average increase in the knowledge of the participants after disability

orientation was 39%, and this was maximum for those who could not read or write, or had no access to the formal education.

**Keywords:** causes of disabilities, disability orientation, outcome, preventive measures, rights and entitlements.

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It is estimated that around one billion (15%) of the global population experience some form of disability, and the prevalence is higher for developing countries like Nepal.<sup>1</sup> Persons with disabilities are more likely to experience adverse socioeconomic outcomes such as less education, poorer health outcomes, lower levels of employment, and higher poverty rates. It is estimated that 7 to 10 percent of Nepalese population live with some form of disability, out of which physical disability (32%) predominates.<sup>2</sup> Social stigma associated with disability, and lack of knowledge on rights of PWD's, leads to this group being marginalized in society. Attitudes towards PWD's are based on levels of knowledge, and experiences and interactions with PWD's.<sup>3</sup> Awareness plays a central role in creating positive attitudes towards PWD's, and in facilitating early identification and promoting treatment seeking behavior. Hence, the present study was designed with an objective to assess the outcome of disability orientation sessions conducted by the district supervisors of Hospital and Rehabilitation Centre for Disabled Children (HRDC).

### Materials and Methods

The study was conducted in three purposively selected project districts of Nepal (Rupendehi, Arghakhanchi and Gulmi). Fifty percent of the municipalities were randomly selected from each district. Twenty-five persons from each municipal were randomly enrolled in the study as a study participant (425 participants) (**Figure 1**). The survey was given twice at 2-weeks interval to 425 community people. A community level course about disability awareness followed a structure focusing on awareness, interaction with PWDs, and education, in order to make a positive impact on their knowledge. Pre-test interviews were conducted by HRDC's district supervisors and post-course interviews were conducted by independent enumerators to reduce the enumerator's bias. Awareness sessions were broken down into three key components: 1) Causes of disability, 2) Rights & entitlements of PWD's, and 3) Prevention & control measures of disability. The purpose of this research was to examine if such a disability orientation program can bring about a positive change in the level of knowledge of the participants. Only permanent citizens

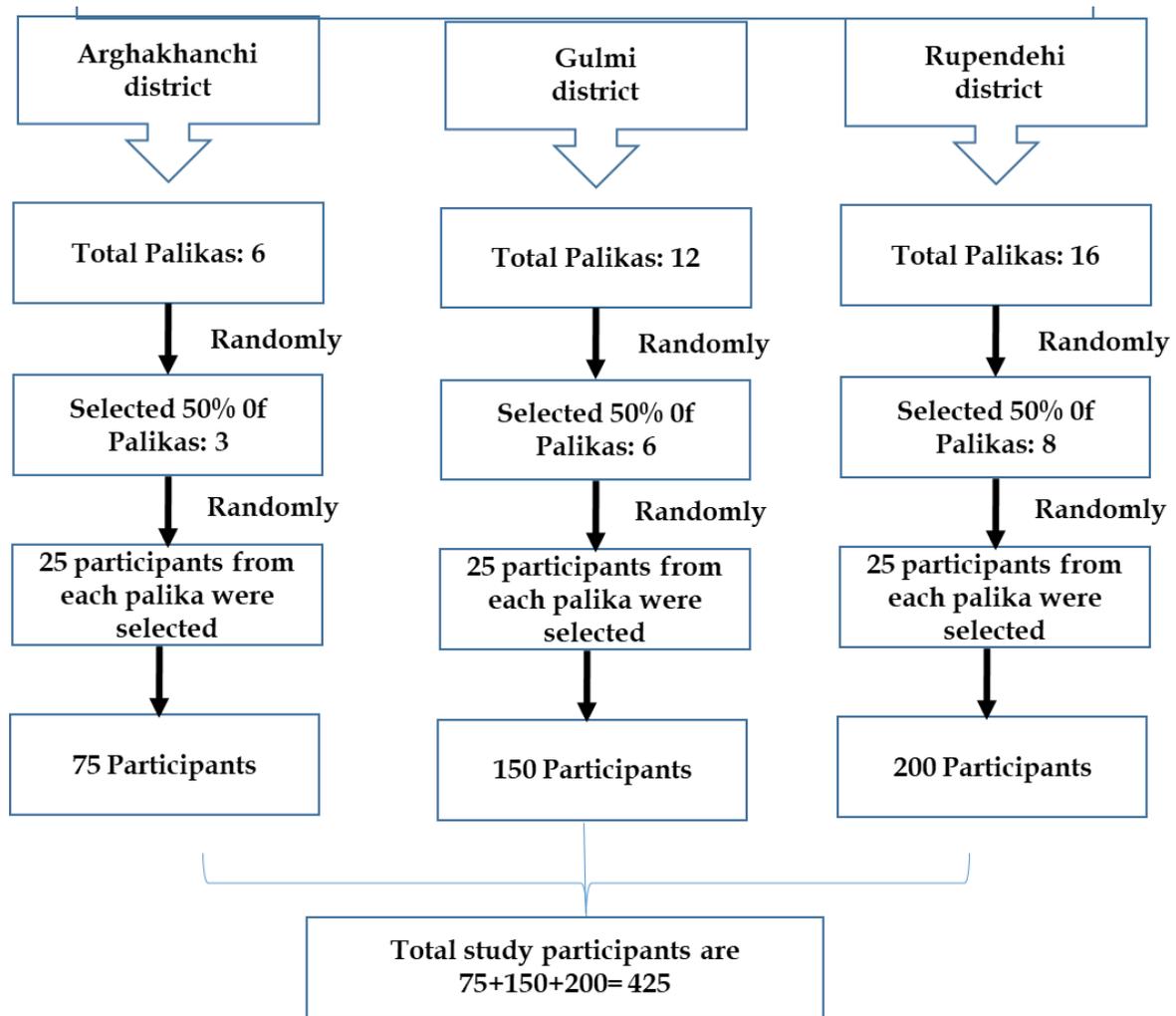


Figure 1: Participant selection

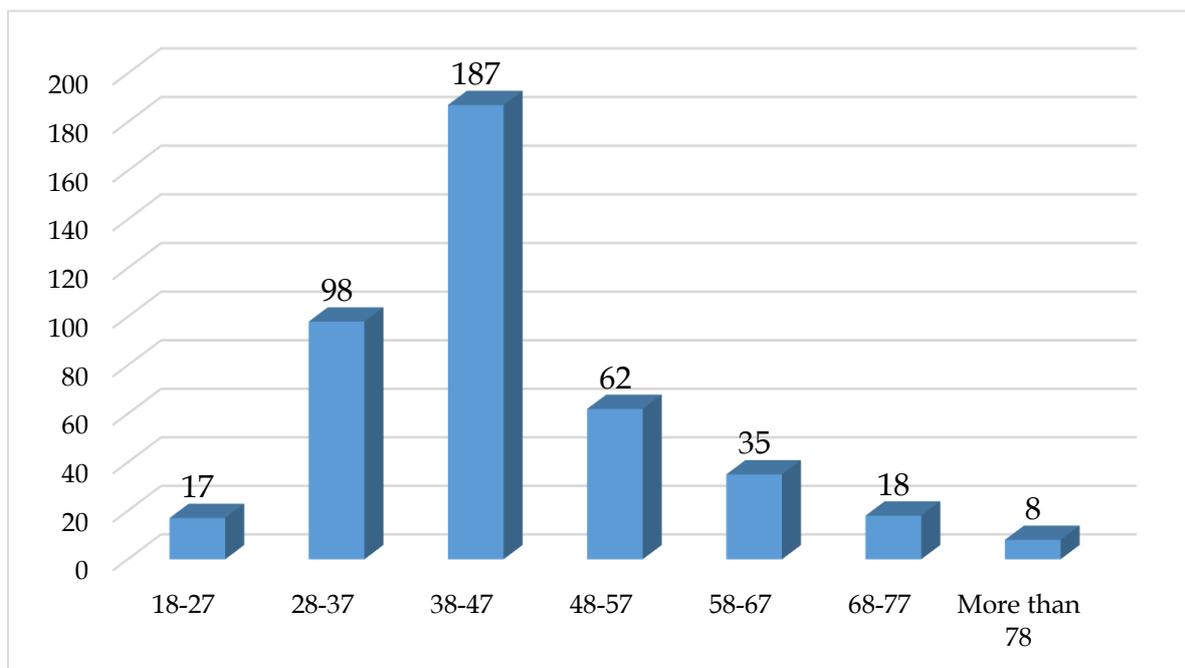


Figure 2: Age wise distribution of the participants

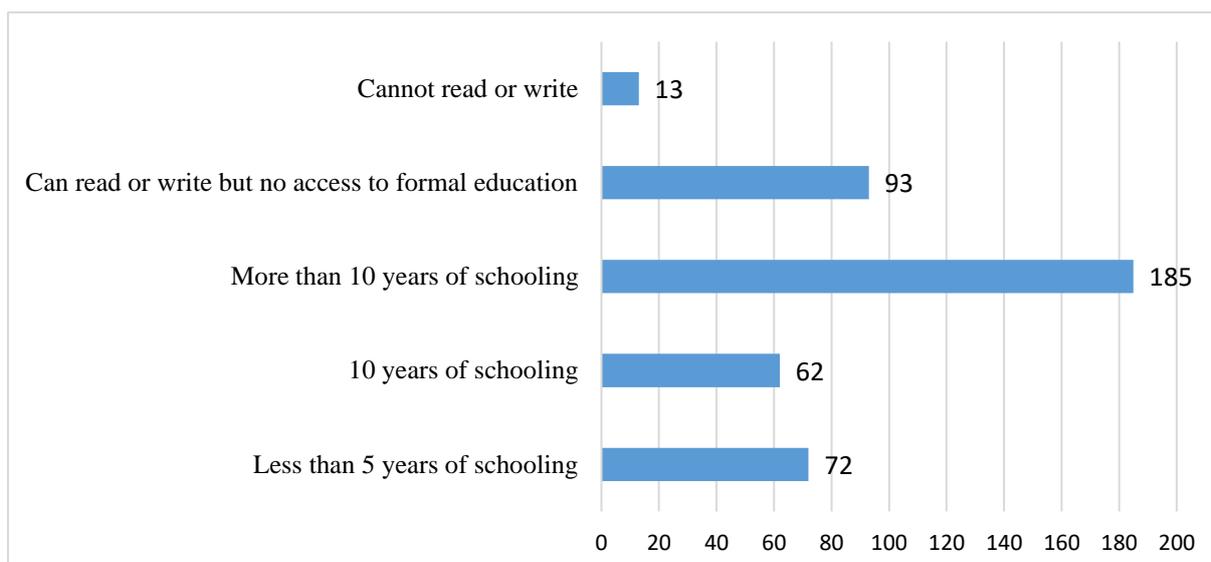


Figure 3: Educational state of the participants

of the respective municipalities who gave an informed consent and were above the age of 18 years were included. Any participant who did not complete the post-test survey was excluded. Ethical clearance was taken from the hospital IRC. The pre-test was administered immediately before the orientation session and the post-test was taken at 2 weeks' interval.

Data was analyzed using the SPSS 20. Descriptive data was presented in tables and graphs. This shows pre and post test scores and any changes thereof.

**Results**

There were 52% female participants, and 44% were between 38 to 47 years (Figure 2). 83% were married, 44% had completed at least ten years of schooling, and 3% could not read or write (Figure 3). Our results indicate a significant increase in the

level of knowledge of participants regarding causes of disability, rights & entitlements of persons with disability, and prevention of disability. The average increase in the level of knowledge regarding causes of disability, rights & entitlements of the persons with disability and the preventive measures of disability were by 24%, 52% & 40% respectively (Figure 4). The overall average increase in the knowledge of the participants after disability orientation was 39 percent. While improvement in knowledge after disability orientation was observed across all levels of education, this was maximum for participants who could read or write but had no access to the formal education, and those who could not read or write. The details of orientation subjects versus level of knowledge is shown in Tables 1, 2 & 3.

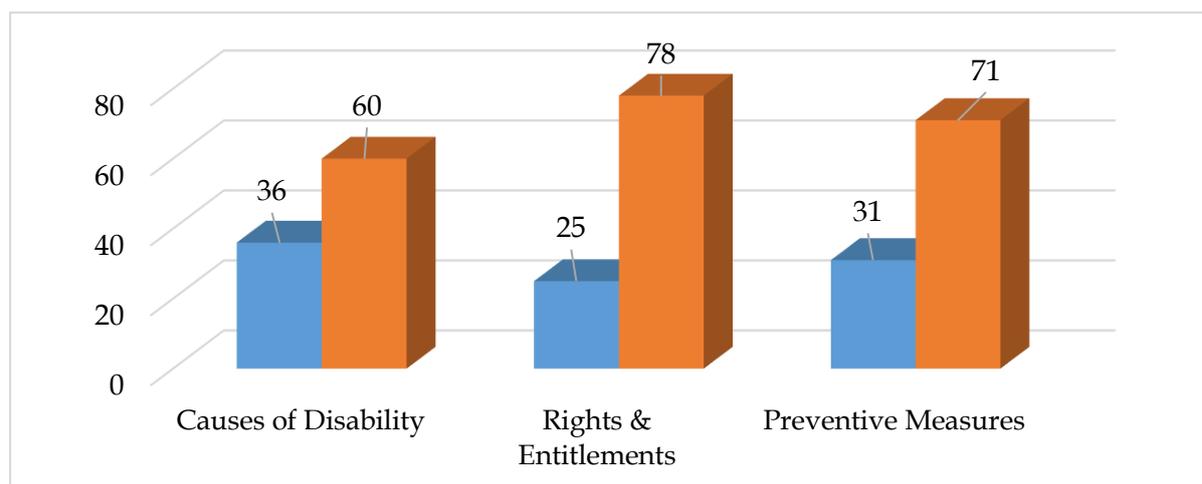


Figure 4: Correct response in %

Education Level	Causes of Disability			P- Value
	Pre-test score	Post-test score	% increase in participants with correct responses	
Less than 5 years of schooling	30	50	16	>0.5
10 years of schooling	56	70	11	>0.5
More than 10 years of schooling	62	73	9	>0.5
Can read & write but no access to formal education (literate)	20	72	43	<0.5
Cannot read and write (illiterate)	10	35	21	>0.5

Table 1: Percentage change in the level of knowledge regarding ‘causes of disability’

Education Level	Rights and entitlements			P- Value
	Pre-test score	Post-test score	% increase in participants with correct responses	
Less than 5 years of schooling	27	78	19	>0.5
10 years of schooling	37	72	13	>0.5
More than 10 years of schooling	47	78	12	>0.5
Can read & write but no access to formal education (literate)	10	98	34	<0.5
Cannot read and write (illiterate)	5	62	22	<0.5

Table 2: Percentage change in the level of knowledge regarding ‘right and entitlement of the persons with disabilities’

Education Level	Preventive Measures			P-Value
	Pre-test score	Post-test score	% increase in participants with correct responses	
Less than 5 years of schooling	25	61	18	>0.5
10 years of schooling	51	81	15	>0.5
More than 10 years of schooling	57	84	13	>0.5
Can read & write but no access to formal education (literate)	15	83	34	<0.5
Cannot read and write (illiterate)	5	46	20	<0.5

Table 3: Percentage change regarding 'preventive measure of disabilities

**Discussion**

Disability of any kind in the setting of poor socioeconomic environment poses a significant barrier to the right to education, employment, and livelihood, compounded by gender bias that is prevalent in such communities.<sup>4</sup> Addressing such barriers, thus improves the likelihood of the aforementioned rights. One such study from our center showed that disabled children who had received treatment were functioning well in physical skills and in their educational settings, and that parental attitudes were generally positive, including that for the female child.<sup>5</sup> Nepal’s ratification of the United Nations Convention on the Rights of Persons with Disability (UNCRPD) in 2010 and the passage of the Disability Rights Act in 2017 highlights Nepal’s commitment to addressing disability-related issues using the rights-based model rather than welfare or charity-based approaches.<sup>6</sup> The most basic tenet of a rights-based model is access

to information regarding available rights. Our study shows that even a simple orientation program has significant impact on the understanding of disability and rights related issues. This improvement was seen to be much more for persons who had no formal education, so programs catered to persons without formal education can be very impactful in raising awareness regarding the issues discussed in this paper.

**Limitation of the study**

The present study was limited to three purposively selected districts of Nepal where HRDC was already working, so results may not be generalized to other districts with different demographic characteristics. However, as the results of this study indicate that that orientation improves knowledge in the domains studied, and that level of education of the participants influences the degree of such improvement, these conclusions are likely to be valid to a greater or lesser degree in

different areas of Nepal. This can be a subject of future research. Another limitation is that the questionnaires used in this study were self-generated from our experience, and not validated.

### **Conclusion**

The results of the present study show that disability orientation brought a positive change in the knowledge of participants, especially those who had no access to formal education. Disability orientation is an important and cost effective (considering the number of preventive disabilities) tool/ medium for social inclusion of the persons with disability. Our study can be used as a pilot to design better studies suited to larger audiences, both with or without formal education, and administer such orientation to improve the overall understanding of disability related issues in the wider community.

This study was supported by CBM Global through HRDC's Inclusive Development of Children with Physical Disabilities (IDC) project.

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## **Surgical Outcomes of Intradural Extramedullary Tumor Excision- 5 Years of Experience at a Tertiary Care Center**

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**Introduction:** Intradural extramedullary tumors (IDEM) are one of the causes of back pain and weakness of the lower extremity. Diagnosis is usually made by myelography or magnetic resonance imaging (MRI). Complete tumor excision with decompression of neural elements is one of the preferred treatment methods. However, the outcome of surgical treatment is not well investigated. Hence, this study will evaluate the outcomes of surgical treatment in patients diagnosed with IDEM.

**Methods:** This is a retrospective study carried out between June 2013 and May 2018 at a tertiary care center. The medical records and MRIs were reviewed for demography, types of tumor, pain score, neurological recovery, and complications. Neurology was assessed using Frankel's grade, pain using Visual analog score (VAS), and functional status using Nurick's grade.

**Results:** The average age of the patient was 42.9 years (range 17-64 yrs). The average follow-up was 32.8 months. The average duration of illness was 10.5 months (range 2-36 months). The thoracic region was most commonly involved. Histo-pathological analysis showed

Schwannoma in nine cases, meningioma in two cases, and myxopapillary ependymoma in one case. Three had normal neurology before and after surgery. Seven patients had Frankel D neurology and one patient had Frankel B and one patient had Frankel C neurology at presentation. All improved to Frankel E at the last follow up. Ten patients who had Nurick's grade 3 preoperatively improved to grade 1 postoperatively. Three patients underwent instrumentation, one patient developed post-op cerebrospinal fluid (CSF) leak. No recurrence was reported within the final follow-up.

**Conclusion:** Surgical treatment in patients with IDEM results in improvements in pain-related outcomes and neurological recovery. However, further studies with larger case series and longer follow-up duration are recommended to evaluate risks of recurrence.

**Keywords:** extramedullary, intradural, intra-spinal, meningioma, schwannoma, IDEM, spinal cord tumor.

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**S**pinal tumors account for approximately 5-15% of the nervous system neoplasms. These tumors are classified as extradural (55-60%) and Intradural tumors (40-45%). Intradural tumors are further classified as intradural extramedullary and Intradural intramedullary tumors.<sup>1</sup> IDEM accounts for two-thirds of all primary intraspinal neoplasms including mostly schwannomas, meningiomas and ependymomas.<sup>2,3</sup> Most of these tumors are benign and usually occur in middle-aged people. Schwannomas and ependymomas usually affect younger age men whereas meningiomas affect elderly women.

Spinal meningiomas are mostly found in the thoracic region and are mostly seen in females most probable reason is the influence of female hormones.<sup>4</sup> Intradural tumors usually present with nonspecific

symptoms. These tumors can also cause back pain and weakness of the lower extremity. It usually presents with radiculopathy and mild pain which is more severe during nighttime. In one of the studies, Sphincter dysfunction, paraparesis, and erectile dysfunction occurred in 20%, 12%, and 2% of patients respectively.<sup>5</sup> Sometimes these tumors present like a prolapsed intervertebral disc or spinal stenosis, diagnosing them can be challenging. MRI/Myelography allows making early diagnosis and accurate identification of the location of a tumor in the dura mater and its dural attachment before surgery.<sup>6</sup>

Sir Victor Horsley in 1888 succeeded for the first time in surgically excising an IDEM tumor located in the thoracic region, and this was 44 years before the invention of myelography.<sup>6</sup> Even though surgical

management is widely preferred, the clinical and functional outcomes of surgery are not well investigated. Hence, we aim to investigate the surgical outcomes in patients with intradural extramedullary tumors.

### Materials and Methods

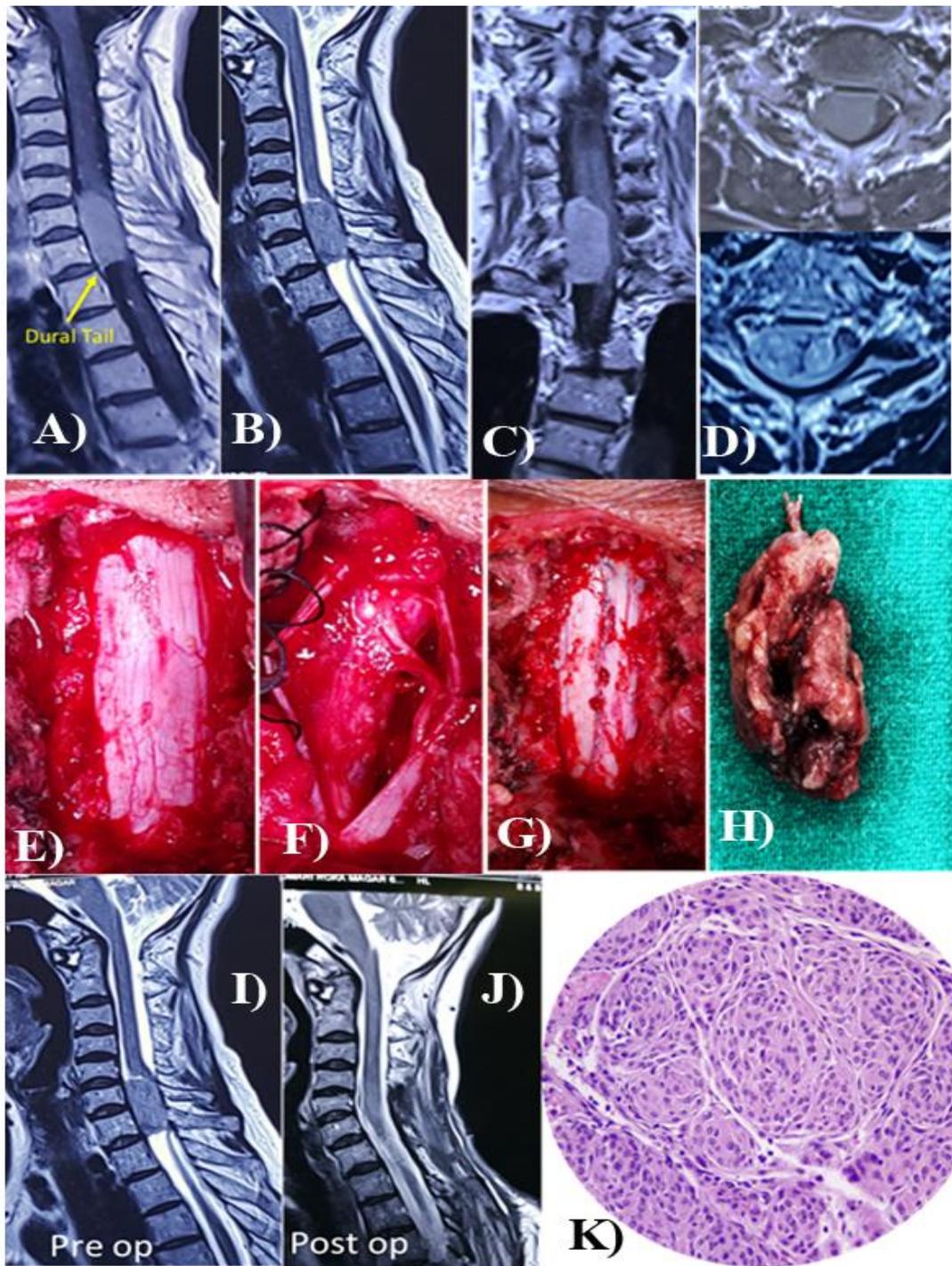
This study was conducted following the guidelines of the Institutional Review Committee (IRC). Medical records of patients who underwent surgery for Spinal tumors in a tertiary care hospital between June 2013 and May 2018 were screened. Patients operated for IDEM with histological confirmation of tumor were included. All medical records demographic details, clinical charts, operative notes, and outpatient's progress and MRI was traced. The patient's age, sex, vertebral level, duration of illness, neurology at presentation, and last follow-up, histological diagnosis, and follow-up period were recorded. All patients underwent Laminectomy and tumor excision en bloc via a posterior approach to the spine irrespective of location. Three patients underwent instrumentation as facet joints had to be sacrificed due to the location of the tumor leading to instability. En Bloc resection was possible in all patients. Nerve root was preserved in all cases. Closure of dura was done with 6-0 prolene. No microscope or

neuromonitoring was used for only of the cases. A negative pressure drain was used in all cases. Drain removal was done on 3<sup>rd</sup> postoperative day (POD) and then mobilization started. No braces were used postoperatively. Discharge was done on the discretion of the operating surgeon. Suture removal was done on 14<sup>th</sup> post operative day.

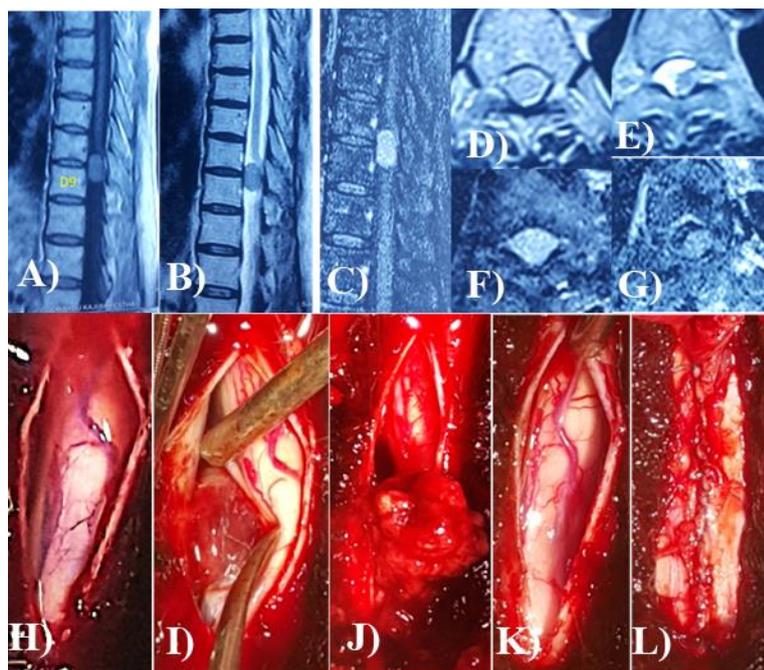
The patient's neurology was assessed; preoperatively and at last follow up using Frankel's grading system whereas neurological function was assessed using Nurick's grading system. Surgery was done by various members of the Spine team and not by a single surgeon. Preoperative and post-operative (at last follow-up) pain was assessed using Visual Analogue Score (VAS).<sup>7</sup> All patients had at least six months of follow-up (**Figure 1-3**).

For the assessment of the preoperative neurological function, the patients' ambulation ability was graded into 5 levels according to Nurick's grading system.<sup>8</sup> Neurological status was assessed using Frankel scale.<sup>9</sup>

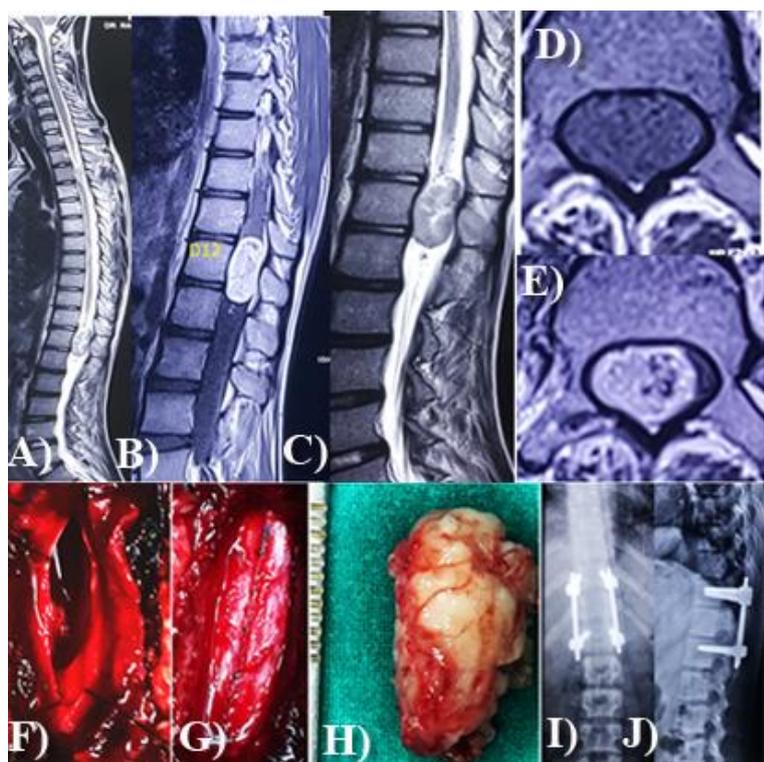
Continuous data were reported as mean (standard deviation) and categorical data were reported as frequency (%). Statistical analysis was done using paired t-test and the level of significance was set at 0.05. Data analysis was done using SPSS software version 20.



*Figure 1: Meningioma: (A, B) Preoperative sagittal T1, T-2 weighted, and T1 coronal MRI showing the hypo intense lesion; (C, D) Preoperative axial T1 Weighted Image and post-contrast T-2 weighted image showing the partially enhanced tumor and severely compressed spinal cord (E-H)Intraoperatively image showing tumor resection; (I, J) Pre and Postoperative sagittal T-2 weighted MRI showing radical tumor resection); (K) Biopsy of the same lesion showing typical lobulated whorl pattern of cellular growth typical of meningioma.*



*Figure 2: Meningioma: (A-C) Preoperative sagittal T1, T-2 weighted MRI showing the hypo intense tumor; (D-G) Preoperative sagittal and axial post contrast T-2 weighted image showing the partially enhanced tumor and severely compressed spinal cord (H-L) Intraoperative image showing tumor resection.*



*Figure 3: Schwannoma: (A-C) Preoperative sagittal T1, T-2 weighted MRI showing the hetero intense tumor; (D, E) Preoperative axial T1weighted image and post-contrast T-2 weighted image showing the partially enhanced tumor and severely compressed spinal cord; (F-H) Intraoperative image showing tumor resection; (I, J) post op x-ray after instrumentation.*

**Results**

31 spinal tumors were treated at the Department of orthopedics, spine unit, B & B Hospital during the period between June 2013 and May 2018. Out of which, 12 cases

were intradural extramedullary tumors (38%).

The average age of the patient was 42.9 years (range 17-64 years). Follow up period was 32.8 months (range 6-48 months) on

Case	Age(in years) /Sex	Vertebral Level	Histological Diagnosis	Duration of Illness (months)	Neurology at Presentation (Frankel)	Neurology at final follow up (Frankel)	Follow up Duration (Months)
1	17/Male	Dorsal 12	Schwannoma	24	D	E	8
2	32/Male	Dorsal 12- Lumbar 2	Maxillo Papillary Ependymoma	5	D	E	12
3	48/Male	Dorsal 9-10	Schwannoma	7	D	E	48
4	22/Male	Dorsal 8-10	Schwannoma	9	D	E	48
5	40/Female	Lumbar 3-4	Schwannoma	36	E	E	7
6	60/Female	Cervical 5-7	Meningioma	3	D	E	6
7	60/Female	Dorsal 11-12	Schwannoma	13	D	E	13
8	56/Female	Dorsal 10-11	Meningioma	2	B	E	24
9.	51/Female	Lumbar 4-5	Schwannoma	12	E	E	36
10.	40/Female	Sacral 1	Schwannoma	12	E	E	48
11.	64/Female	Lumbar 1	Schwannoma	7	D	E	48
12.	25/Female	Dorsal 7-8	Schwannoma	3	C	E	48

*Table 1: Demographic and Clinical Profile*

Case	Visual Analogue Score(VAS)		Nurick's Grade	
	Pre Op	Last Follow up	Pre Op	Last follow up
1	6	1	3	1
2	6	2	3	1
3	7	3	3	1
4	5	1	2	1
5	6	2	3	1
6	7	3	3	1
7	8	4	3	2
8	6	1	4	1
9	7	3	3	1
10	6	2	3	1
11	6	2	3	1
12	7	2	3	1

*Table 2: Clinical Symptoms*

	Pre Op	Last Follow up	p-value
VAS	6.41 (0.79)	2.16 (0.93)	<0.001*

\*One-tailed paired t-test,  $\alpha=0.05$

*Table 3: Clinical Improvement*

average. The mean duration of illness was 10.5 months (range 2months-36 months). Most common location of the tumor was thoracic (n= 6(50%)) followed by thoracolumbar junction (n=4 (33.3%)) and cervical (n=1(8.3%)) and sacral (n=1(8.3%)). Histo-pathological analysis showed Schwannoma (n=9(75%)), meningioma (n=2(16.5%)) and myxopapillary ependymoma in (n=1(8.5%)) cases as shown in **Table 1**. Three had normal neurology before and after surgery and seven patients had Frankel D neurology which improved to Frankel E. One patient developed a post-op cerebrospinal fluid (CSF) leak, which was managed conservatively and the patient improved.

As shown in **Table 2**, ten of twelve patients were Nurick grade 3 preoperatively and showed marked improvement postoperatively. However, one patient only improved from grade 3 to grade 2. The pain was improved in all patients who were clinically significant as shown in **Table 3**.

## Discussion

Our case series had a mean age of 42.9 years. We found that schwannoma affects

much younger patients than meningioma, which affects older women. The most common location was the thoracic region. This demographic finding was similar to other studies.<sup>4,10,11</sup> Joshi et al<sup>10</sup> reported meningioma as a common tumor (8 out of 19 cases) in their case series however our case series showed 9 out of 12 cases to be schwannoma.

In our study, the average duration of illness till diagnosis was 10.5 months. There is a delay in making an early diagnosis, as IDEM spinal tumors tend to be slow-growing, produce vague symptoms and produce symptoms similar to intervertebral disc prolapse and spinal stenosis.<sup>12</sup> Hence, taking a detailed history, conducting a thorough physical examination, and performing MRI scans on the proximal regions are helpful for patients with back pain and radiating pain in the lower limbs.

In our study, the mean VAS score was reduced from 6.41 preoperatively to 2 postoperatively. This suggests that surgical treatment results in a marked reduction in pain-related symptoms. The outcome of this study is comparable to other studies<sup>12-16</sup> (14-16). Ahn et al<sup>13</sup>, in 2009, showed that the mean VAS score was reduced from 8 to

1.2 after surgery. Similarly, Ahsan et al<sup>14</sup> reported a reduction of the mean VAS score from 7.67 to 1.14.

Most studies showed marked improvement in neurological function after surgery. Gu et al<sup>15</sup> in his study showed 93% of his patients had improved Frankel grade and 7% had no change. In our study, 9 patients had neuro deficit and all of them showed marked improvement at the last follow-up. Similarly, Ahn et al.<sup>13</sup> in their study showed a marked improvement in mean Nurick's grade after surgery. Similar findings were also seen in our study. This suggests that surgical treatment of these tumors results in an improvement in neurological functions. One of the main reasons behind that could be the benign nature of these tumors with the possibility of complete excision.<sup>4</sup> In our study, all patients had a tumor that was well surrounded by a capsule with a well-defined margin, and complete excision of the tumor was possible in all cases.

The prognosis of these tumors depends upon several factors, such as pre-operative duration of symptoms, the severity of neurological deficits, and proximal or ventral location.<sup>2,6</sup> According to Asazuma et al<sup>17</sup>, the recurrence rate of intraspinal neoplasm was 7.2% and 46% of recurred masses were IDEM spinal tumors which recur more commonly than other intraspinal tumors. They also reported that the ventral location of a tumor, extradural

invasion, neurogenic tumors, and ependymomas were the risk factors for recurrence. No recurrences were reported at last follow-up. However, a longer follow-up period is needed to substantiate the finding.

### **Limitations of the Study**

The limitations of the study were smaller sample size and study design, i.e. retrospective study, related limitations. This suggests that there are risks of biases. Surgery was performed without using a microscope or any neurophysiological monitoring and was not done by a single spine surgeon. Only a few patients had a long follow-up period.

### **Conclusion**

Surgical management of IDEM tumors results in a marked reduction of pain-related symptoms and improvement in neurological function as the complete excision of tumors is possible. Although the chance of recurrence is low, further study with long-term follow-up is warranted.

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**Annexure**

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**Nurick’s grading system**

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Grade 1	Normal walk, possible clinical spinal irritation	
Grade 2	Slight difficulty in walking with normal domestic and working life	
Grade 3	Functional disability limiting normal work and domestic activities	
Grade 4	Significant weakness making walking impossible without help	
Grade 5	Bedridden or wheelchair-bound	

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**Frankel Scale**

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Frankel A	Complete	No motor or sensory function below level of lesion
Frankel B	Sensory only	No motor function, but some sensation preserved below level of lesion
Frankel C	Motor useless	some motor function below level of lesion
Frankel D	Motor Useful	Useful motor function below level of lesion
Frankel E	Recovery	Normal motor and sensory function, may have reflex abnormalities

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## Primary Retroperitoneal Synovial Sarcoma: A Diagnostic Dilemma

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Synovial sarcoma is a rare soft tissue tumor comprising of 1% of all malignancies. It usually involves the extremities in adults in third to fifth decade of life. Retroperitoneal soft tissue sarcoma accounts for only 15% of all soft tissue sarcomas. Here, we report a rare case of primary retroperitoneal synovial sarcoma confirmed by ancillary tests performed on cell block sample.

**Keywords:** cell block, retroperitoneal, synovial sarcoma.

**S**ynovial sarcoma accounts for approximately 5-10% of all soft tissue sarcomas with extremities being the most common site.<sup>1,2</sup> Other less common sites are head & neck, mediastinum, abdominal wall and retroperitoneum.<sup>3</sup> Primary retroperitoneal synovial sarcoma is an extremely rare entity and has worse prognosis.

Cell block plays a significant role in

cytological diagnosis by maintaining architecture which closely resembles that seen in surgical specimens.<sup>4,5</sup> The cell block is prepared by processing the small tissue fragments or sediments retrieved from cytological specimens into paraffin blocks.<sup>6</sup> It is widely accepted that the diagnostic accuracy is increased by cell block techniques.<sup>4</sup>

### Case Report

A 77 years old female presented to our OPD with history of abdominal pain for a month.

Her general condition was fair. Systemic examination was normal. Blood investigation was within normal limits.

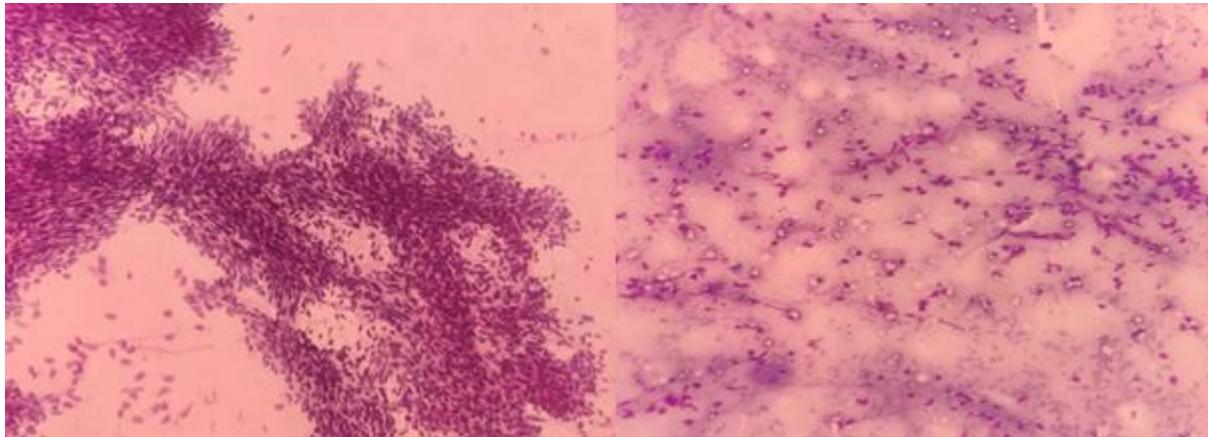


Figure 1: Cytology smears showing spindle shaped tumor cells (Pap stain & Giemsa stain)

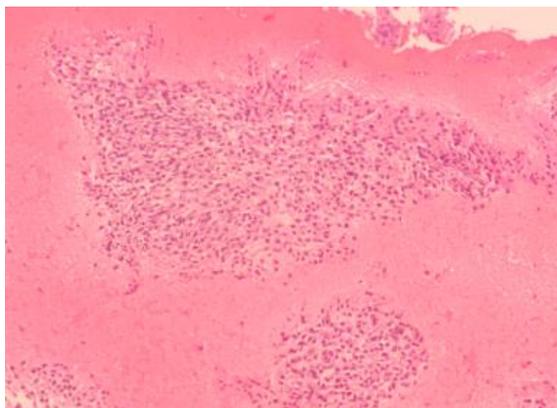


Figure 2: Cell block preparation showing tumor cells arranged in diffuse pattern (H&E stain)

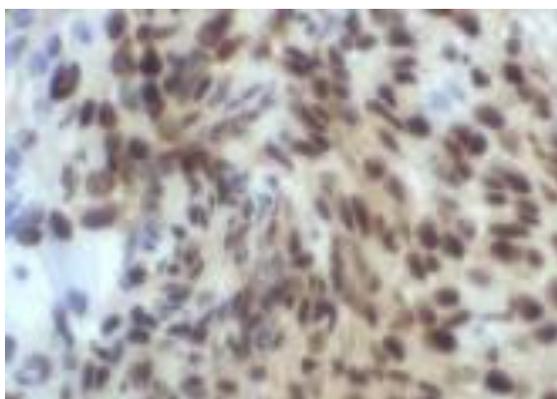
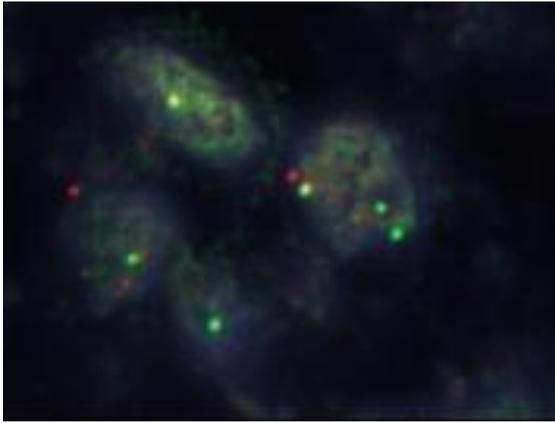


Figure 3: Immunopositivity for TLE1

Blood investigation was within normal limits. There was past history of pulmonary tuberculosis for which she had taken complete medication. CT scan of abdomen and pelvis was done and showed multiple conglomerated necrotic lymph nodes in the retroperitoneum in the para-aortic region measuring 8 x 5 cm suggestive of metastatic lymph nodes. CT guided FNAC was done and the smears were highly cellular comprising spindle shaped cells (**Figure 1**) with plump to elongated nuclei, coarse chromatin and moderate pleomorphism. Mitotic activity was seen with some atypical ones. The cytological features in conjunction with cell block preparation (**Figure 2**) findings were suggestive of malignant spindle cell lesion. The differential diagnoses included retroperitoneal sarcoma and gastrointestinal stromal tumor.



*Figure 4: FISH showing SS18 gene rearrangement*

Immunohistochemistry was performed on cell block sample which showed positivity for TLE1 (Figure 3), CK and negativity for desmin, SMA, S100, CD117, CD45, FLI1, MDM2, CDK4 and MyoD1; suggestive of synovial sarcoma. The diagnosis of synovial sarcoma was further confirmed by FISH studies showing SS18 gene rearrangement (Figure 4). Detail clinical and radiological evaluation was done but did not reveal any other significant findings. The final diagnosis was given as primary monophasic retroperitoneal synovial sarcoma. The patient was given three cycles of AIM regimen (Adriamycin, Ifosfamide and mesna). Then the patient was lost to follow-up.

### Discussion

Synovial sarcoma is a well-defined entity with uncertain histogenesis. Despite its name, there is no evidence that tumor arises from or differentiates towards synovium.<sup>7</sup>

Preoperative diagnosis of retroperitoneal synovial sarcoma is challenging due to its nonspecific imaging features mimicking benign lesions or metastatic nodes.<sup>1,3</sup> However, these tumors appear hypodense on CT scan with poor central enhancement due to necrosis, hemorrhage, cystic change and about 30% cases show intra tumoral calcification.<sup>8</sup> Pathological examination remains the mainstay for the diagnosis of such tumors.

Synovial sarcoma is divided into biphasic, monophasic spindle, monophasic epithelial and poorly differentiated types.<sup>2,3</sup> Monophasic spindle synovial sarcoma is the most predominant subtype which can be confused with other spindle shaped tumors like fibrosarcoma, leiomyosarcoma, MPNST, spindle cell melanoma, GIST and sarcomatoid carcinoma.<sup>9,10</sup>

Cell block increases the diagnostic accuracy owing to the paramount role they play in ancillary tests like immunohistochemistry and molecular studies like FISH.<sup>6</sup> Immunohistochemistry was done in cell block sample and showed positivity for CK, CD99 and TLE; suggestive of SS.<sup>2,10</sup> Negativity for desmin, S100, CD117, SMA and CD45 ruled out other tumors with spindled morphology.<sup>11</sup> However, the golden diagnostic tool in 90% cases is SS18 gene rearrangement which has been demonstrated in our case in cell

block sample highlighting the immense role of cell block in diagnosis.<sup>3,4,5,10</sup>

### **Conclusion**

Even though primary retroperitoneal synovial sarcoma is a rare entity, it should be kept in differential diagnoses of retroperitoneal mass. The diagnosis is based on histopathological, immunohistochemical and molecular studies. Our study has highlighted the significance of cell block in diagnosis of tumors.

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## An Interesting Incidence Following General Anesthesia: Anesthesia Mumps?

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We report a case of 50 years old man with rapid onset of unilateral buccal and periauricular swelling after extubation of the trachea. Described by some pre-existing literature, the most likely cause is acute postoperative parotitis occurring due to various causes like pneumoparotitis, venous congestion, Stenson's duct obstruction, the retrograde passage of air, and excess saliva secretion in the perioperative phase. The resolution was relatively rapid, uncomplicated, and spontaneous in around 48 hours in our case. Most cases are managed with rehydration and anti-inflammatory drugs like ours while some mandate airway management due to obstruction. General anesthesia with endotracheal intubation demands caution as complications ranges from trivial to major. Awareness of benign complications helps prevent unnecessary worries.

**Keywords:** anesthesia mumps, retrograde parotitis.

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**A**cute transient swelling of the parotid gland related to general anesthesia called postoperative parotitis or anesthesia mumps is relatively infrequent. It is an extremely rare complication. The occurrence is variable (0.2–17%) and the exact incidence is

unknown. It is a benign complication. (1-3). The reason for its recent decline is the frequent use of antibiotics after major operations, combined with better oral hygiene, adequate control of fluid and electrolyte balance, and blood replacement. Presently postoperative parotitis occurs

mostly in uremic patients as they are prone to dehydration.<sup>2</sup>

Postoperative salivary gland swelling is typically mild and doesn't have many subjective symptoms. Most cases resolve spontaneously without being noticed and thus the discrepancies in the incidence rate. The exact cause and pathophysiology of anesthesia mumps remain unknown, but salivary duct occlusion, sialorrhea, venous congestion and venostasis, involvement of the autonomic nerves, a stormy induction or emergence from anesthesia and side effects of medications like belladonna, depolarizing neuromuscular-blocking drugs, succinylcholine, straining, coughing, and the Valsalva maneuver are few of the presumed causes.<sup>2,3</sup>

Salivary gland obstruction may be caused by physical compression in the lateral position, positions that rotate and flex the neck, compression by endotracheal tubes leading to mucosal lesions and edema. Pneumoparotitis is caused by the penetration of air by retrograde flow into the salivary duct due to positive pressure in the oral cavity by mask ventilation, pharyngeal reflex, and cough reflex; and dehydration may also cause duct obstruction. Use of atropine, and sympathetic nervous system activation due to invasiveness of surgery causing increased salivary viscosity, may in turn itself cause an occlusion. Intratracheal

manipulation stimulates parasympathetic nerves and mediates the pharyngeal reflex leading to salivation, vasodilation and hyperemia in the salivary gland. Stimulation of the sympathetic nerves also causes salivation which is short-lasting, thick and mucinous, and vasoconstriction occurs. A noradrenaline infusion increases salivary alpha-amylase.

Most cases of postoperative parotitis resolve spontaneously with follow-up and observation alone. At the most rehydration therapy and anti-inflammatory drugs may be needed to treat it. In extremely rare conditions patients suffer from severe complications, including upper airway obstruction and thus may mandate airway management. Laryngoscopy and tracheostomy might both be difficult if there is massive edema and swelling of the neck.<sup>3</sup>

### **Case report**

A 50-yrs-old, 55 kgs male presented to the operation room for MIPPO of left tibial fracture. His Hepatitis B antigen was positive but there was no active hepatitis. Also, he had a history of spinal instrumentation and subsequent removal under General anesthesia which was uneventful. He denied any past H/O Anesthetic complications. His baseline peripheral oxygen saturation(Spo2) was 99% on ambient air, heart rate was 76 beats



*Figure 1: Periauricular swelling when viewed from the side*

per minute, blood pressure was 140/95 mmHg and respiratory rate was 16 breaths per minute before induction. Preoperatively, his chest examination results were normal and a chest radiograph indicated clear lung fields. His baseline lab report including hemoglobin, total leucocyte count, platelet count, blood sugar, renal function tests, electrolytes, and liver function tests were all within normal range. The patient was premeditated with 2mg intravenous (IV) midazolam, and 150 mcg IV fentanyl was given for analgesia, and anesthesia was induced with 180 mg iv propofol, and 7 mg iv vecuronium for facilitation of tracheal intubation. The trachea was intubated with a 7.5-mm internal diameter (ID) endotracheal tube using a no. 4 Macintosh laryngoscope with direct visualization of the vocal cords. The vocal cord visualization required two attempts at laryngoscopy with BURP (backward, upward, right, and posterior)



*Figure 2: Frontal view of periauricular swelling of the face*

maneuver, and Cormach Lehane grade was III. Bilateral breath sounds were confirmed. Intraoperatively, the lung mechanics, as well as the oxygenation, were normal. The patient was hemodynamically stable with minimal blood loss and was easily ventilated and oxygenated. Maintenance of anesthesia was done with isoflurane, vecuronium, oxygen, and positive pressure ventilation. A total of 1400 ml lactated Ringer's solution was administered during the 2hrs 30-mins surgical procedure. Reversal of neuromuscular (NM) blocker was done with 2.5 mg IV neostigmine and 0.6 mg iv glycopyrrolate. Suctioning of the oropharynx was done and the trachea was extubated. The patient coughed vigorously post-extubation.

The patient gradually developed swelling over the right periauricular and submandibular region which was prominent after 1.5 hours post-extubation in the evening (**Figure 1, 2**). There was no

pain over the region of swelling but he developed minimal pain the other day. On examination, there was fluctuant swelling over the right cheek with no tenderness or rise in temperature. There was no fever. The next morning there was slight tenderness present as the swelling had increased. The intraoral hygiene was poor and parotid swelling was observed. The maxillofacial team was consulted. The impression of retrograde parotitis secondary to general anesthesia was made. The patient was managed with intravenous fluids, NSAIDs, and prophylactic antibiotics were given considering the patient's poor oral hygiene and the chance of developing a secondary bacterial infection. At 36 hours post-extubation, the swelling had subsided and there was no pain. On examination too minimal parotid swelling was present, no tenderness was elicited. On manipulation, there was no pus or serous discharge from the right parotid duct opening and saliva flow was present. All lab parameters were within normal range. Serum lipase was normal but serum amylase was raised.

### **Discussion**

Unilateral or bilateral swelling of parotid glands can appear rapidly during the perioperative period, but has an uncomplicated course and resolves spontaneously. Few cases with massive

swelling leading to airway obstruction and subsequent need for airway management have also been described in the literature. In our patient, the onset of swelling was 1.5 hours after extubation of the trachea and was progressive. Fortunately, our patient did not develop airway obstruction or any major distress physically. The patient was psychologically affected due to sudden swelling around the facial region which as per him was quite unrelated to the course of his primary illness that he presented with to the hospital.

In a case report an extensive swelling in the left parotid region, extending to the buccal and cervical areas, developed in a 30-year-old man immediately after a partial nephrectomy which was performed under general anesthesia. Radiological examination immediately after the onset revealed no abnormality, but a large swelling was detected by computed tomography in the left parotid region. Serum amylase was significantly elevated. The clinical signs had almost disappeared approximately 2 weeks after the onset, following intravenous infusion of antibiotics and transfusion. This parotid swelling was considered to be acute postoperative parotitis induced during induction of anesthesia by luxation of the temporomandibular joint or by the positioning of the patient during operation.<sup>4,5</sup> The literature available

describes many causes of perioperative parotitis. Mechanical trauma and parasympathetic nerve stimulation during endotracheal intubation can be the cause. Obstruction of glandular ducts by calculi or thick secretion can cause bacterial infection and thus purulent sialadenitis. Increased airway pressure during mask ventilation combined with muscle relaxation causes air to enter the parotid orifice and obstruct the ducts. This could have been the potential cause in our case. Adverse drug reactions can be of type A that is an augmentation of a pharmacologically known effect with drugs like morphine infusion or of type B that is abnormal immune-mediated or non-immune-mediated reactions with Mechanical trauma due to endotracheal intubation is also likely in our case as there were multiple attempts at laryngoscopy and swelling developed quite early on after extubation. Excretory duct obstruction could not be ruled out due to a lack of imaging. An adverse drug reaction was also unlikely because none of the drugs already associated with drug-induced sialadenitis was administered. Although intraoperative venous stasis is less likely as the patient was in the supine position, it could have happened during laryngoscopy due to head extension. Also, vigorous coughing immediately post-extubation could be another potential cause in our case. Based on the findings and as suggested in

many kinds of literature, this condition resolves spontaneously without a complicated course. The management modality involves anti-inflammatory drugs and hydration. In our case too the swelling resolved within 48 hours, was not much of a problem for the patient and the management involved NSAIDs and intravenous and oral fluids along with good oral hygiene and close monitoring of the course of the condition.<sup>4, 5</sup>

### Conclusion

We report a case of postoperative parotid sialadenitis which resulted in unilateral facial oedema but without worrisome airway obstruction. Hypothetically, intra-operative mechanical trauma and head position or increased airway pressures during mechanical ventilation can best describe the genesis of swelling in our case. Our report suggests that physician anesthesiologists and surgeons must know this condition. The unawareness about this complication could bring about unnecessary doubts and worries and also there could be a lack of preparedness due to the absence of anticipation of potential progression to airway obstruction.

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## Myopericytoma: A Rare Soft Tissue Neoplasm

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Myopericytomas are rare, slow growing benign soft tissue neoplasm that arise from perivascular smooth muscles within the superficial subcutaneous soft tissue, and shares features of both glomus and smooth muscles. Myopericytomas depict one of the many related perivascular tumors of myoid lineage, with similar morphology and combined immunohistochemical profile, including positive staining for smooth muscle actin. We report a case of a forty-seven-year-old man with a slow growing nodule on the antero-medial aspect of the left knee. An excision biopsy was performed, which confirmed a myopericytoma with no evidence of malignancy

**Keywords:** glomus, myopericytoma, neoplasm, perivascular, smooth muscles.

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**P**erivascular tumors are soft tissue tumor that begin in the cells that wrap around blood vessels, and can be benign or malignant. This group traditionally has included glomus tumor and hemangiopericytoma. “Myopericytoma” is a recently delineated entity showing a hemangiopericytoma like

vascular pattern i.e. the presence of numerous thin-walled branching blood vessels and pericellular reticulin fibers.<sup>1</sup>

### Case Report

A forty-seven-year-old man presented with a mass over the anterior medial aspect of left knee which had been growing slowly

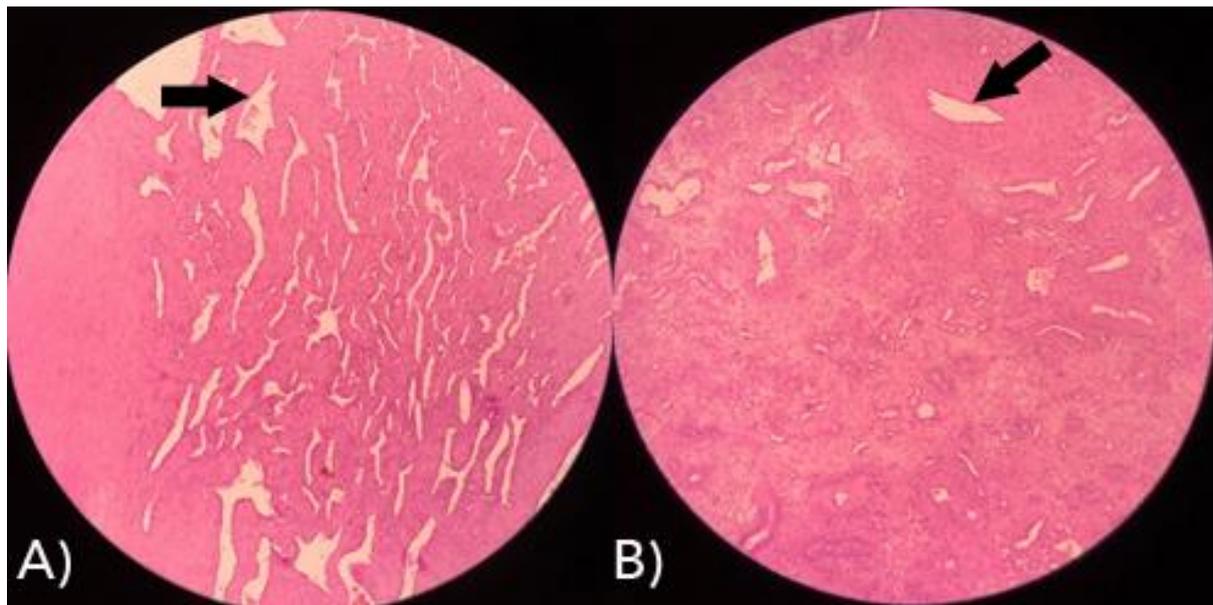


Figure 1: Photomicrograph of myopericytoma showing in blue arrow hemangiopericytoma like vasculature (A) and black arrow showing perivascular growth of myoid tumor cells, a characteristic myopericytoma morphology (B). [ H&E stain; 100X]

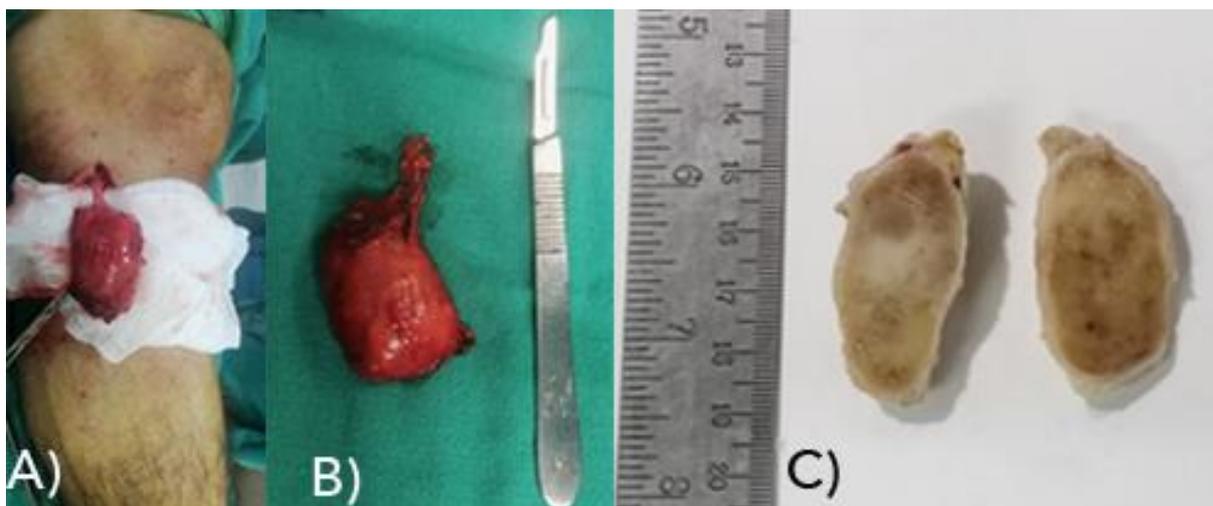


Figure 2: A) & B) Intraoperative picture showing an encapsulated mobile solid soft tissue mass with a vascular pedicle. C) Gross picture showing a circumscribed tumor with a solid, gray-white cut surface with interspersed brownish areas.

for few years, and was causing pain on movement of the knee. Physical examination revealed a 5X4 cm nodular and mobile mass, with minimal tenderness to palpation and normal overlying skin. Ultrasonography reported a well-defined

hyper-vascular solid soft tissue mass with a differential diagnosis of a lipoma or a hemangioma.

A complete surgical excision was done with clear margins, and the specimen sent for histopathologic examination. Macroscopic

examination showed a 5X3 cm solid, gray-white colored capsulated nodular tissue, with a solid, gray-white cut surface with interspersed brownish areas. Histology showed a circumscribed lesion comprising of numerous thin walled variably sized, branching hemangiopericytoma-like vasculature surrounded by cytologically bland oval to spindle shaped cells with multilayered concentric growth, and a loose and edematous stroma. The lesion was surrounded by a thick pseudo-capsule with areas of hemorrhage, brownish pigment deposits and myxoid change. No evidence of malignancy was seen.

### Discussion

Stout and Murray's description of hemangiopericytoma suggested that many vascular benign and malignant tumor have a similar pattern.<sup>2</sup> Requena et al proposed the term 'myopericytoma' for a solitary myofibroma<sup>3</sup> in 1996, which Granter et al adopted in 1998, and grouped myopericytoma, myofibromatosis, solitary myofibroma, and infantile hemangiopericytoma, to form a single morphological spectrum of tumors showing perivascular myoid cell/pericytes.<sup>4</sup> This spectrum was further expanded by McMenamin and Fletcher in 2002 with a report of 5 malignant myopericytomas that exhibited aggressive biological behavior.<sup>5</sup> Myopericytoma describes a benign, usually

subcutaneous tumor with, myoid-appearing oval to spindle-shaped cells with a striking tendency for concentric perivascular growth.<sup>6</sup> The World Health Organization officially named this tumor type as "myopericytoma" in 2002, and classified it into the group of peripheral blood cell/vascular cell tumors.<sup>7</sup> Myopericytomas can be multifocal involving single or multiple anatomic region,<sup>8</sup> and tends to occur predominantly in the skin and superficial soft tissue of the distal extremities (hand, foot, ankle, and leg), followed by the head and neck region, and the trunk. Clinically myopericytomas are often solitary and well demarcated slow growing soft tissue mass for several years. Multiple small clustered nodules termed 'myopericytomatosis' are less common. Prognosis is usually excellent with complete surgical resection. Indications for surgery include cosmesis, pain (with or without movement near a joint, as in our case), or suspected malignant transformation.<sup>7</sup>

Both myopericytoma and myofibroma are frequently misdiagnosed as other tumors, frequently sarcomas.<sup>9</sup> Most myopericytomas are benign lesions, though a few malignant cases have also been described.<sup>4</sup> Although malignant transformation of these usually benign lesions is rare, recurrence has been reported in 10%–20% of patients.<sup>2</sup> There is no role

for radiotherapy or chemotherapy. Malignant myopericytomas are rare, and typically deep seated, infiltrative tumors with nuclear pleomorphism, brisk mitotic activity, and an aggressive clinical course with or without distant metastasis disease.<sup>5,10</sup> Ultrasonography shows a well-defined hyper-vascular solid soft tissue mass, whereas MRI shows T1 hyperintense signal representing internal hemorrhage or fat within the lesion, and T2 hyperintense signal representing soft tissue components, thick peripheral tissue, and a varying amount of peritumoral edema that enhances avidly with gadolinium.<sup>11</sup>

### **Conclusion**

Myopericytomas are uncommon benign vascular tumors that can sometimes be mistaken for a sarcoma. Malignant transformation is rare. Ultrasonography shows a well-defined vascular lesion, and surgical excision confirms the diagnosis, as well as cures the symptoms, as in our case.

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## Congenital Elevation of the Scapula Associated with a Dislocated Glenohumeral Joint

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Congenital elevation of the scapula (Sprengle's deformity) is a rare entity resulting from an aberration in the caudal descent of the scapula. The glenohumeral articulation is intact but the range of motion may be restricted. Surgical interventions are aimed at improving cosmesis or improving range of motion. We present the case of a six years old male child with congenital elevation of his left scapula in association with a dislocated left glenohumeral joint and absent range of motion that significantly improved following a scapular resection.

**Keywords:** congenital elevation, dislocated glenohumeral joint, glenohumeral joint, scapula.

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**A** six-year-old male child presented to us with a huge lump on the left side of his neck with loss of the left shoulder contour since birth (**Figure 1A & 1B**). The patient did not have any active movements of his affected non-dominant left shoulder, but passive motion was possible. History revealed that the lump had been present since birth and

gradually enlarged causing cosmetic concerns. There had been no active range of motion in the affected extremity, but neurovascular status was intact. Plain radiographs and CT studies showed a high riding left scapula with complete dislocation of the left glenohumeral articulation with the humeral head lying below the inferior angle of

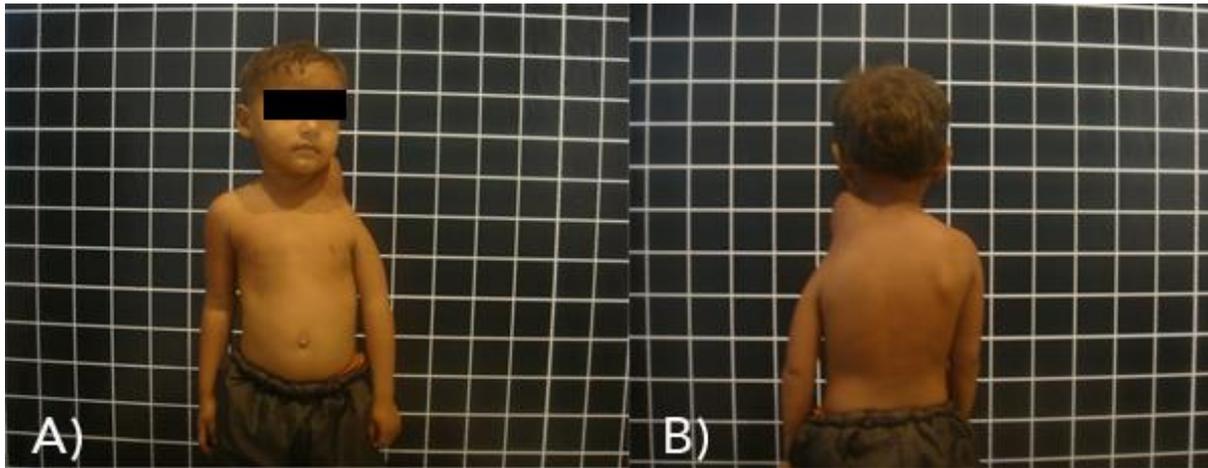


Figure 1: The large lump on the left neck and shoulder region as seen from A) the front and B) behind. Glenohumeral motion was absent on this side.



Figure 2: A) Anteroposterior radiograph and B) 3-D CT reconstruction showing an elevated left scapula with complete dislocation of the left glenohumeral joint.

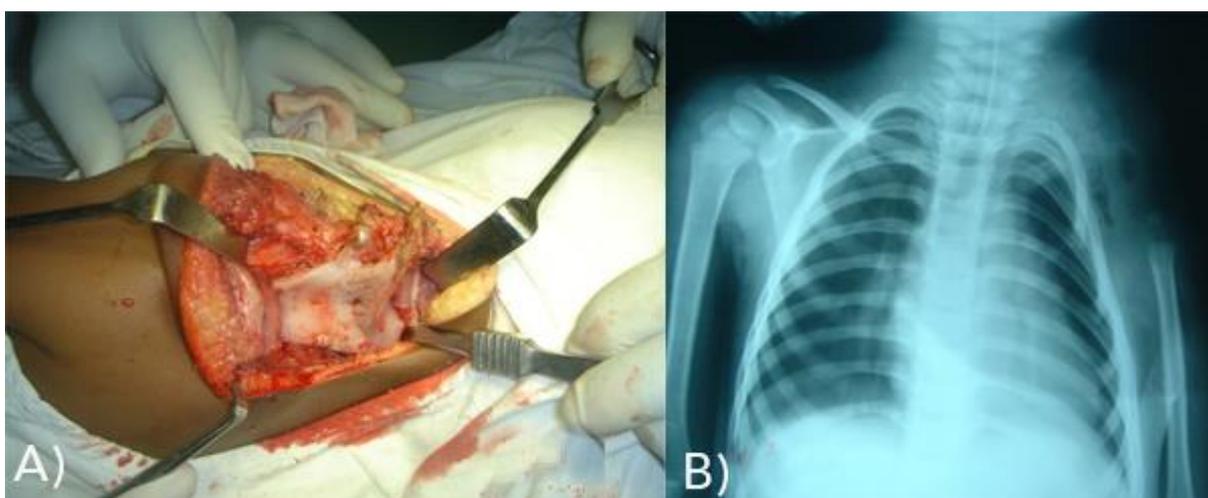
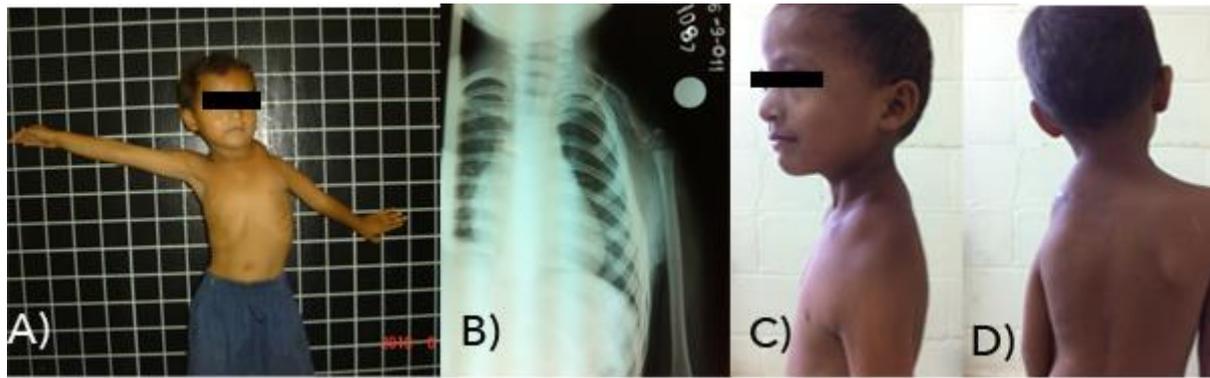


Figure 3: A) Left scapulectomy in progress; removed specimen showed a hypoplastic scapula with dysplastic glenoid and aplastic coracoid B) Post-scapulectomy radiograph.



*Figure 4: A) Six months post surgery, the child had regained functional range of motion with 80° of abduction B) Looked cosmetically better but C) Left shoulder contour remained flattened D) Radiograph at six months showing regeneration of the left scapula in the periosteal sleeve.*

scapula (**Figure 2A & 2B**). There was no associated omovertebral connection. The patient didn't have any other associated anomalies. In view of the cosmetic deformity, lack of range of motion of the affected shoulder, restricted neck range of motion on the affected side and concerns of neurovascular impingement if the mass was allowed to progress, a decision to undertake surgical resection of the mass was reached. With the patient in a lateral decubitus position, an incision was made over the mass and a left claviculo-scapular resection was performed subperiosteally (**Figure 3A & 3B**). The removed specimen revealed a hypoplastic scapula with dysplastic glenoid and poorly defined coracoid. Post-operative recovery was uneventful and range of motion exercises was instituted. To our great surprise, the patient regained functional range of motion of the affected shoulder and neck, including active

abduction of 80° (**Figure 4A**), over a period of six weeks and the cosmetic appearance was much better (**Figure 4B**) although shoulder contour remained flat (**Figure 4C**). At six months follow-up the child was fully functional with the affected extremity and could carry out all activities of daily living consistent with his age. Radiographs at six months showed evidence of regeneration of the left scapula and clavicle in the periosteal sleeve (**Figure 4D**).

### Discussion

High riding scapula (Sprengle deformity) is the commonest congenital anomaly affecting caudal descent of the scapula during development, often associated with other congenital anomalies like Klippel-Feil syndrome and congenital chest wall anomalies.<sup>1-3</sup> A bony, cartilaginous or fibrous omovertebral connection linking

the superomedial scapula to the lower cervical spine, the omovertebral body,<sup>4</sup> is present in up to 50% of cases and rarer variants like omo-clavicular and omo-occipital connections of the undescended scapula have also been described.<sup>5</sup> The etiology of failure of descent of the growing scapula is thought to be a result of omovertebral connections resulting from abnormal epiphysis originating from the superior angle of scapula or by factors such as increased intrauterine pressure or increased CSF permeability of the fourth ventricle leading to aberrations in mesodermal development.<sup>6,7</sup> Cavendish<sup>1</sup> classified these lesions depending on their clinical appearance whereas Rigault<sup>8</sup> classified them based on the degree of proximal arrest. The main problems with this deformity are cosmetic unsightliness and limitation of shoulder motion, mainly abduction. To the best of our knowledge, there has been no report of a high riding scapula in association with a completely dislocated glenohumeral joint.

The indications to intervene surgically, as was in our case, are cosmetically unacceptable deformity and lack of range of motion of the affected shoulder girdle and neck. We also felt that leaving the lesion as it were could risk traction on the neurovascular structures as the mass enlarged. Surgical treatments range from excision of the prominent medial part of the

scapula to more extensive procedures where the scapula is brought down to the desired level and muscles realigned to stabilize it there.<sup>9-11</sup> We deemed it necessary to perform a total scapulectomy and partial claviclectomy to ensure the neck range of motion would be unobstructed by the mass, to eliminate any evolving traction on the neurovascular structures and in the hope of achieving a better cosmetic appearance. The restoration of the shoulder joint motion post-operatively was unanticipated and most gratifying to both the patient and his family and our team. The implications of the re-growth of the removed scapula which became evident by six months is unclear and the question of whether an extraperiosteal excision should have been carried out will only be answered on longer term follow up.

### Conclusion

In summary, congenital elevation of scapula is a rare disorder and its association with a dislocated glenohumeral joint has not been described in the literature to the best of our knowledge. Total resection of the scapula has led to excellent range of motion gain from zero to functional active motion of the affected shoulder and a better cosmetic result at short term follow up.

Longer term follow up until skeletal maturity will define the role and

effectiveness of such radical surgery in patients presenting with this rare disorder.

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