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**Announcement**

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Pamidronate, alendronate and zoledronate are nitrogen based bisphosphonates with an affinity for bone. Pamidronate is extensively used in pediatric osteoporosis and has many orthopaedic applications. Nitrogen based bisphosphonates act by preventing osteoclastic resorption. The affected osteoclasts undergo apoptosis and their proliferation is prevented by the down regulation of Rab, Rho, Rac and Ras, the guanosine triphosphate binding proteins responsible for membrane trafficking, cytoskeletal maintenance and reorganization, protein kinase activation, cell growth and cell proliferation. In addition, in vitro, pamidronate has been shown to decrease cell viability and the proliferation of human alveolar osteoblasts, causing apoptosis in 20% of the cells. Its effect on RAS is important for its antitumor activity and is actively used in many malignancies.

Recent studies have used alendronate for homing Mesenchymal Stem Cells (MSCs) to bone in in-vivo models of osteoporosis in mice by tagging it to Leucine-Leucine-Proline-2-Alanine (LLP2A), which has an affinity for MSCs, subsequently exhibiting bone anabolism. Pamidronate has been demonstrated to be a suitable tagging agent for the gamma emitting isotopes for bone imaging. In a rabbit model of distraction osteogenesis, it has been shown to improve short term healing of the regenerate as assessed by quantitative computerized tomography and 4 point bending test.

There are numerous clinical uses of pamidronate in children. It has been used for osteogenesis imperfecta and other paediatric osteoporotic disorders for the last two decades. The other common uses of pamidronate in paediatric practice are osteoporosis associated with steroid use, juvenile rheumatoid arthritis and cerebral palsy. It is widely used in oncology for cancers associated with hypercalcemic conditions and tumors which secrete Parathyroid Hormone Related Peptide (PTHrP) such as leukemia, lymphoma and neuroblastoma.

Bone diseases associated with hematological conditions like beta thalassemia, rare conditions such as osteoporosis - pseudoglioma syndrome (congenital blindness, osteoporosis and fractures), idiopathic hyperphosphatasia (Juvenile Paget's disease) and bone crisis in storage disorders such as Gaucher's disease are also indications for its use.

There are many other less well known indications for the use of bisphosphonates. In congenital pseudarthrosis of tibia, zoledronate/pamidronate has been shown to improve efficacy of BMP2 in treatment and is now used as an adjunct to surgery in an ongoing multicenter clinical trial. Fibrous dysplasia and its variants are other clinical conditions where bisphosphonates have been shown to reduce deformity and possibly reconstitute cystic lesions with normal bone. Kiely found that when zoledronate/pamidronate was used in children with poor regenerate in distraction osteogenesis, six out of seven children healed without further intervention.

Pamidronate was initially used for treatment of flare ups of myositis ossificans progressiva where clinical improvement was seen. Cyclical use is
being carried out on a trial basis to assess if it can impair endochondral osteogenesis in a heterotopic site\textsuperscript{19}. In Perthes’ disease (and in avascular necrosis due to other causes), it has been shown to prevent femoral head deformation. Zoledronate also forms part of a treatment trial for this condition. (Trial ID: ACTRN 12610000407099)

A rare hereditary osteodysplasia (Hadju Cheney syndrome) has acroosteolysis as one of its prominent features. There are anecdotal reports of pamidronate use in this condition leading to improvement in the bone mass density and reduction in the risk of fractures and acroosteolysis, thus delaying morbidity\textsuperscript{20,21}.

There is evidence to suggest that bisphosphonates are effective in controlling the disease progression possibly by their direct action on osteoclasts in primary or recurrent giant cell tumour of bone\textsuperscript{22}. In the cystic lesions of bone with secondary Aneurysmal Bone Cyst (ABC), there is a clinical response to systemic bisphosphonate therapy\textsuperscript{23}. Pamidronate has been found to be a useful, safe and adjunctive therapy effective in decreasing pain in chronic recurrent multifocal osteomyelitis involving the spine when anti-inflammatory therapy fails\textsuperscript{24,25}.

While pamidronate is the most commonly used drug for children, there are now studies with zoledronate for osteogenesis imperfecta in children at a dose of 0.025 - 0.05 mg/kg/year in two half-yearly doses as a 30 minute infusion, which was found to be as safe and effective as pamidronate\textsuperscript{26,27}.

The transient complications of bisphosphonates in children include fever, hypocalcaemia and respiratory distress\textsuperscript{5}. The major complications are renal, ophthalmic and cardiac (zoledronate) and many are dose related\textsuperscript{28}. While the short term use with standard dosage protocols is safe, the safety of long term usage is not established. The main problem with the prolonged use of bisphosphonates is the accumulation of micro damage (microscopic cracks which do not heal) due to suppression of remodeling, leading to an increased theoretical fracture risk. In clinical dosages, however, there is no evidence that micro damage occurs, but there are reports of stress fractures occurring in the femur while on therapy and improving spontaneously on stoppage of therapy\textsuperscript{15}. The therapeutic effects of pamidronate are marked within the first two to four years, and there is no decrease in bone density for one year after discontinuing the treatment\textsuperscript{29}. There is no exact therapy break recommendation for children and this issue needs to be looked at using pooled data from multiple centers. There is also a need to watch for the cumulative effects of all bisphosphonates administered in the young because of teratological effects seen in rats, long half-lives and their ability to cross the placental barrier.

References:


Age and gender distribution of common childhood fractures in an urban population in Malaysia.


ABSTRACT

Accurate information on various aspects of childhood fractures is important to allow planning of effective preventive measures. This type of information is not readily available in many developing countries. We conducted this study to provide information on demographic pattern and anatomical distribution of common childhood fractures in an urban population of our country. Between 1st July 2009 and 31st July 2010, we interviewed family members and caretakers of 309 children below the age of 12 diagnosed with fractures. Questions were asked based on a research proforma. The most common fracture site was distal radius (23.63%), followed by distal humerus (21.36%), radius/ulna shaft (17.47%), tibia/fibula (8.74%) and clavicle (7.12%). Overall male to female ratio was 2.47:1 and male dominance was obvious after the age of 2 years. Most fractures occurred at home (35.60%) and majority were due to fall from level higher than bed height (48.22%). We concluded that higher rate of fracture with increasing age is mainly seen in boys. Since home is the most common place of injury, and fall from height above level of bed the most common mechanism of injury, fracture prevention should start from home.

INTRODUCTION

Trauma is the most common cause of hospital admission in children\(^1\). It has been estimated that about one third of children would have suffered a fracture before the age of 17\(^,\)\(^2\)\(^,\)\(^3\). Pattern of these fractures is influenced not only by gender and age, but also by seasonal variation and many other socio environmental factors\(^4\)\(^,\)\(^5\). The load of trauma related problems in developing countries is expected to be higher since larger proportion of the population belongs to this age group\(^6\). It has also become an important source of overall morbidity and mortality since increasing standard of living has resulted in a general decline in the incidence of malnutrition, diarrhoea and infectious disease\(^7\).

However, available information on patterns of fractures and demographic characteristics of fractures in children in developing countries is scanty\(^5\)\(^,\)\(^8\)\(^,\)\(^9\). This study was designed to look into the age and gender distribution of fractures in children at various anatomical sites in an urban population in Malaysia. The demographic characteristics and socio-environmental factors were also analysed with the aim to provide recommendations on injury prevention.

MATERIALS AND METHODS

Our institution is a tertiary referral hospital catering to trauma patients of Western part of Kuala Lumpur and the adjacent satellite city...
Petaling Jaya. Children who sustained fractures within the 12 month period between 1st July 2009 to 31st June 2010 were included in this study. All fracture cases seen in this institution were given a follow up appointment in the paediatric orthopaedic out-patient clinic irrespective of whether they were admitted or not. The exception to this would be children with skull and rib fractures as they would be managed by the neurosurgical or general surgical teams. The study was approved by the ethical committee of the institution.

Parents or caretakers of children who presented or came for follow up to the clinic were approached and invited to participate. Patients with chronic medical conditions and those with pathological fractures due to underlying bone pathologies were excluded. When there was more than one bone involved, only the fracture of the larger bone was considered. A questionnaire was designed and used to collect relevant information through an interview conducted by one of the authors. Information on demographic characteristics, timing and environment of injury were gathered for analysis.

We also reviewed the radiographs of all the fractures to ensure accuracy of diagnosis and fracture classification based on AO system\textsuperscript{10}. Physeal plate injuries and incomplete fractures were classified into proximal or distal fractures of the bone. For the forearm, we also classified the fracture sites as proximal, middle and distal thirds of the whole forearm length from the tip of ulna olecranon to tip of radial styloid. Age and gender distribution of five of the most common anatomical fracture sites were further analysed and compared with available information.

Data was analysed with Excel spread sheets. T-test was used to find the significant difference between the two proportions. Statistical significance was accepted at p<0.05. Statistical analysis was performed using SPSS version 15 (Chicago, IL, USA).

**RESULTS**

All the parents whom we approached agreed to participate in the study. A total of 309 children were recruited in this study. There were 220 boys and 89 girls giving a male to female ratio of 2.47:1. There was no difference between the genders below the age of 1. Male predominance became obvious with increasing age and doubled the number of females after the age of 7 years. There were 230 (74.43%) upper limb fractures, 40 (12.94%) lower limb fractures, and 39 (12.62%) fractures involving the head, neck and trunk. Fracture of the left side was slightly more than that of the right especially in the upper limbs, but the difference was not statistically significant (p=0.299)(Table I)

<table>
<thead>
<tr>
<th>Anatomy</th>
<th>No. (%)</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Upper limbs</td>
<td>119 (38.51%)</td>
<td>0.2992</td>
</tr>
<tr>
<td>Lower limbs</td>
<td>20 (6.47%)</td>
<td>-</td>
</tr>
<tr>
<td>Total</td>
<td>139 (44.98%)</td>
<td>0.3136</td>
</tr>
</tbody>
</table>

The most common anatomical site of fracture was the radius and ulna (142/45.95%), followed by humerus (86/27.83%), tibia and fibula (27/8.74%), clavicle (22/7.12%) and femur (13/4.20%). Fracture is more common in boys especially for radius and ulna (p=0.004) (Table II). Based on the classification recommendation by the AO Paediatric Classification Group\textsuperscript{10}, there
were 73 distal radius / ulna fractures (AO type 23) compared to 54 shaft (AO type 22) and 15 proximal (AO type 21) fractures (Table III). The distal radius / ulna contributed 23.63% of all fractures making it the most common fracture site in this study, marginally ahead of distal humerus fractures that contributed 21.36%. When we re-classify the fractures based on distal one third of the whole forearm length, there were 92 fractures and it contributed 29.77% of all fractures (Table IV). The difference between these two methods of classification was not obvious in proximal forearm fractures: 15 (10.49%) AO type 21 fractures as compared to 21 (14.69%) proximal third radius / ulna fractures. Incidence of forearm fractures increases gradually with age, especially in boys. Males predominantly sustained distal radius fracture especially after the age of eight. Distal humerus (AO type 13) contributed 21.36% of all fractures (Table III). Peak incidence of this fracture was between age four to six in both males and females, and it was much less common in older girls. More boys sustained tibia/fibula and clavicle fractures across all age groups. Clavicle fracture in girls is less common with no fracture recorded after the age of eight. Femur fracture mostly occurred in children within the first two years of life, and there was no femur fracture in girls after six years of age.

Home remained the most common place of

Table II : Common fractures by anatomy and gender.

<table>
<thead>
<tr>
<th>Anatomical Sites</th>
<th>Male, No. (%)</th>
<th>Female, No. (%)</th>
<th>Total, No. (%)</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Radius/Ulna</td>
<td>109 (35.28)</td>
<td>33 (10.68)</td>
<td>142 (45.95)</td>
<td>0.0038</td>
</tr>
<tr>
<td>Humerus</td>
<td>55 (17.80)</td>
<td>31 (10.03)</td>
<td>86 (27.83)</td>
<td>0.1677</td>
</tr>
<tr>
<td>Tibia/Fibula</td>
<td>19 (6.15)</td>
<td>8 (2.59)</td>
<td>27 (8.74)</td>
<td>0.0492</td>
</tr>
<tr>
<td>Clavicle</td>
<td>15 (4.85)</td>
<td>7 (2.27)</td>
<td>22 (7.12)</td>
<td>0.1320</td>
</tr>
<tr>
<td>Femur</td>
<td>9 (2.91)</td>
<td>4 (1.29)</td>
<td>13 (4.20)</td>
<td>0.2707</td>
</tr>
<tr>
<td>Others</td>
<td>13 (4.20)</td>
<td>6 (1.94)</td>
<td>19 (6.15)</td>
<td>0.1750</td>
</tr>
</tbody>
</table>

Table III : Grouping of fractures based on AO Classification.

<table>
<thead>
<tr>
<th>AO Classification</th>
<th>Anatomical sites</th>
<th>Male No. (%)</th>
<th>Female No. (%)</th>
<th>Total No. (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>23</td>
<td>Radius/Ulna (distal)</td>
<td>57 (18.45)</td>
<td>16 (5.18)</td>
<td>73 (23.62)</td>
</tr>
<tr>
<td>13</td>
<td>Humerus (distal)</td>
<td>44 (14.24)</td>
<td>22 (7.12)</td>
<td>66 (21.36)</td>
</tr>
<tr>
<td>22</td>
<td>Radius/Ulna (shaft)</td>
<td>40 (12.94)</td>
<td>14 (4.53)</td>
<td>54 (17.48)</td>
</tr>
<tr>
<td>41, 42, 43, 44</td>
<td>Tibia and Fibula</td>
<td>19 (6.15)</td>
<td>8 (2.59)</td>
<td>27 (8.74)</td>
</tr>
<tr>
<td>06</td>
<td>Clavicle</td>
<td>15 (4.85)</td>
<td>7 (2.27)</td>
<td>22 (7.12)</td>
</tr>
<tr>
<td>11 &amp; 12</td>
<td>Humerus (proximal and shaft)</td>
<td>11 (3.56)</td>
<td>9 (2.91)</td>
<td>20 (6.47)</td>
</tr>
<tr>
<td>Others</td>
<td>Head, Trunk, Hand, Pelvis and Foot</td>
<td>13 (4.21)</td>
<td>6 (1.94)</td>
<td>19 (6.15)</td>
</tr>
<tr>
<td>21</td>
<td>Radius/Ulna (proximal)</td>
<td>12 (3.88)</td>
<td>3 (0.97)</td>
<td>15 (4.85)</td>
</tr>
<tr>
<td>31, 32 &amp; 33</td>
<td>Femur</td>
<td>9 (2.91)</td>
<td>4 (1.29)</td>
<td>13 (4.21)</td>
</tr>
</tbody>
</table>
injury (35.60%), followed by parks (21.04%) and schools (16.50%), and this pattern was similar in all five most common fracture sites. Injury on the road contributed only 9.39% of all fractures. Fall from level above bed height (fall from furniture or stairs) was the most common mechanism of injury followed by fall from below bed height (slip or trip when running). Only two (0.6%) children were seen by traditional healers before getting medical consultation. None of the fractures were caused by non-accidental injuries. There was a slightly higher number of fractures recorded in the month of March in 2010, but otherwise the rate has been generally similar across the months.

**DISCUSSION**

Male predominance has been reported in most childhood fracture studies, and our male to female ratio of 2.47:1 is similar to a larger survey conducted in Hong Kong\(^4\). Many studies reported that overall rate of fracture increases with age, and usually reaches a peak at about 12 years in girls and 14 years in boys\(^3,11\). In our study, increase in rate of fracture was mainly seen in boys with an early peak at around 3 years of age, but we were not able to show a second peak due to the age limit of our study. Interestingly, male predominance can be observed as early as age 3, unlike most other reports where it was only observed in older children and was attributed to hormonal changes at adolescence\(^3,11\). This may be due to social, cultural and environmental factors in Asia where girls are expected to behave in a more feminine pattern starting from early childhood. Upper limb fractures accounted for 74.43% of all fractures in our study. Both sides were equally affected although left sided fractures were slightly more in the upper limbs (Table I). In most other studies, fractures in the left outnumbered the right in the upper limbs, while in the lower limbs they were generally similar\(^3,12\). Mortensson et al\(^13\) has shown that irrespective of handedness, left arm was used more often to break the fall.

Radius/ulna were the most common bones (45.95%) to be fractured in our study, followed by humerus (27.83%). When we compared specific locations of fractures, distal radius and ulna was the most common fracture site (23.63%) consistent with most other reports\(^3,2,3,4\). Our second commonest fracture site was distal humerus (21.36%), followed by radius / ulna shaft (17.48%). Cheng et al\(^4\) from Hong Kong reported similar site of second most common fracture, but in other reports the site varied from small bones fractures in the hand\(^3\) to forearm shaft fractures\(^1\). The percentage of AO type 23 distal radius / ulna fracture (23.63%) was only slightly higher than distal humerus fracture (21.36%). However, when we reclassify distal forearm fractures according to distal one third of the forearm length, a method which is more commonly referred to in trauma literature, the

<table>
<thead>
<tr>
<th>Type of fracture</th>
<th>AO classification</th>
<th>Thirds of radial length</th>
</tr>
</thead>
<tbody>
<tr>
<td>Distal</td>
<td>73 (23.62%)</td>
<td>92 (29.77%)</td>
</tr>
<tr>
<td>Shaft</td>
<td>54 (17.48%)</td>
<td>30 (9.71%)</td>
</tr>
<tr>
<td>Proximal</td>
<td>15 (4.85%)</td>
<td>20 (6.47%)</td>
</tr>
<tr>
<td>Total</td>
<td>142 (45.95%)</td>
<td>142 (45.95%)</td>
</tr>
</tbody>
</table>

Table IV: Grouping of forearm fractures based on AO and equal thirds of radial length
percentage increased to 29.77% (Table IV). Considering the fact that distal forearm fractures have distinct characteristics with respect to the mechanism of injury, age, gender pattern\textsuperscript{11} and that extensive literature has been published on its management\textsuperscript{1,2,3,4}, it may be more relevant for AO type 23 fracture to include area beyond the metaphysis.

Age and gender distribution for distal radius fractures in this study corresponded to type I curve pattern described by Rennie et al\textsuperscript{11}, showing increasing incidence with age and male predominance. However, it was not possible to show the decline in incidence after adolescence due to the age limit of this study. Age distribution for distal humerus fractures in this study showed a unimodal curve pattern with a peak in early childhood similar to the study by Landin\textsuperscript{9}. More recently, Rennie et al reported a similar curve pattern but further described that this fracture affects both boys and girls equally at all ages\textsuperscript{11}. In our study, boys with supracondylar fracture outnumbered girls by a factor of more than 2 after the age of 2 years. This is probably due to socio-cultural and environmental factors of this region that also contributed towards the overall lower incidence of fractures in girls starting from younger age as mentioned earlier. Fracture of the tibia/fibula and clavicle showed male predominance throughout childhood, and the differences between gender were rather constant. We did not find an increasing difference with increasing age as reported in other studies.\textsuperscript{1,9,14,15} Femur fractures were predominantly located in the shaft, comprising 4.2% of all our fractures. This is comparable to the rate of 4.6% reported by Cheng et al from Hong Kong\textsuperscript{4}. More than half of all femur fractures occurred within the first two years of life but the number was too small to draw any conclusion.

The most common place of fracture occurrence according to our study was at home (35.60%), which is defined as area in the house and its surrounding vicinity\textsuperscript{15}. This figure is lower than other studies conducted in India which have reported that between 47.0%-60.8% of fractures occurred at home\textsuperscript{9,16}. Most studies from developed countries focussed their attention on injuries in the playground, park, school and road\textsuperscript{15,17,18} and injuries at home were not commonly described or analysed. Public parks and playgrounds are not readily available in many developing countries and many children would spend more time in or around their homes. As expected, fractures as a result of injuries on the road are not common in children and in our study it only contributed 9.39% of all fractures (Table IV). The most common mechanism of injury (48.22% of all fractures) for nearly all fracture sites was fall from a level more than bed height. This is possibly due to jumping or falling from furniture in the house or facilities in the playground. This is in contrast to the report from a large epidemiology study on childhood fractures conducted in Edinburgh, Scotland, where the most common mechanism of injury (37.4% of all fractures) was fall from level below bed height\textsuperscript{11}.

The practice of traditional healers is common in many developing countries. A recent study in India reported that 36.0% of children with fractures seek traditional treatment before coming to the hospital\textsuperscript{9}, and another report by Nwadinigwe et al\textsuperscript{19} from Nigeria reported a rate of 27.6%. The low rate of initial treatment by traditional healers in this study (0.6%) reflected improving levels of health education and
CONCLUSION

In an urban population in Malaysia the incidence of fractures in girls changed very little after 2 years of age and increase in the overall fracture rates with age was mainly contributed by boys. Distal radius/ulna was the most common fracture site followed by distal humerus. The most common place where fractures occurred was at home and the most common mechanism of injury was fall from a level higher than bed height. Prevention of childhood fractures in developing countries should focus on safety of home environment and further studies should be conducted on this aspect.

ACKNOWLEDGEMENT

We would like to acknowledge contributions by Norfazlin Rashidi and Noor Fadzilah Razali in data analysis and manuscript preparation.

REFERENCES


Awareness among the urban population of our country on the available treatment in modern hospitals. Seasonal variation in incidence of fractures has been reported in many studies conducted in temperate countries, as well as Hong Kong. In the tropics the amount of sunshine and rainfall is rather constant and variation in temperature is also minimal. This would explain the lack of fluctuation in fracture rates in our study throughout the year.

Although many features of age and gender distribution in common childhood fractures were similar to that of other studies in developed countries, observations like lower incidence of fractures in girls from a younger age onwards, place where injury occurred and mechanism of injury would allow the authorities to design more effective strategies to reduce the risk of these potentially preventable problems in children in developing countries. Further studies are needed to investigate home environments to facilitate specific recommendations to reduce the risk of childhood fractures. Studies in rural environment would also be very helpful but these efforts are commonly restricted by limitation in human and financial resources in these regions.

Limitation of this study includes incomplete representation of all the fractures in children especially rib and skull fractures. We also did not include multiple fractures so that analysis of injury risk in severe trauma would not be over represented. Percentage of fractures in various anatomical sites reported in this study is therefore not based on absolute numbers of fractures that occurred.


INTRODUCTION

Paediatric hip fractures are rare and comprise only about 1% of all paediatric fractures. Usually they are the result of high energy trauma like fall from height or road traffic accidents but pathological fractures are also often reported. Management of these fractures is demanding as a number of complications have been documented which can have significant socio-economic and psychological impact on a growing child. Progressive morbidity in a growing child with respect to functional status can impose significant burden on self as well as community.

CLASSIFICATION AND EPIDEMIOLOGY OF THE INJURY

Delbet's classification as described by Colonna is the time tested and widely accepted system in grouping paediatric femoral neck fractures. The main advantage of this system is that it has prognostic significance apart from being descriptive in nature. It is important to remember the classification for further discussion of the management protocol.

Type I: Transphyseal fractures resulting in separation of the capital femoral physis are grouped in this category. It is further subdivided into types IA and IB depending upon the absence or presence of femoral head dislocation.

Type II: In transcervical type, the fracture line passes between the physis and the intertrochanteric line and is intracapsular. They are the most common (45% to 50%) type of femoral neck fractures.

Type III: Cervicotrochanteric fractures are the second most common type of femoral neck fractures (34%) and are located at or slightly above the anterior intertrochanteric line. It is both intra and extra capsular due to the nature of capsular insertion.

Type IV: Intertrochanteric fractures are rare and account for only 12% of femoral neck fractures.

SURGICAL AND APPLIED ANATOMY

Ossification of proximal femoral physis starts at about 4 months in girls and 5 to 6 months in boys and the trochanteric ossification center appears at about 4 years of age. Changes in vascular supply to femoral head with age had been studied extensively using postmortem injection and micro angiographic studies. It has been observed that there in no significant vascular contribution by vessels in ligamentum teres. In the early period of life, major blood supply to the femoral head comes from metaphyseal vessels traversing the femoral neck, which are branches of the medial and lateral circumflex arteries. By 4 years of age as the cartilaginous physis develops, the metaphyseal supply gradually reduces and ceases.

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By the time the metaphyseal supply reduces, the lateral epiphyseal vessels which bypass the physeal barrier become the primary blood supply. It was observed by Ogden that the medial circumflex artery branches into 2 sets of lateral epiphyseal vessels namely, posterosuperior and posteroinferior\textsuperscript{7,9}. Retinacular arterial system originates from the medial circumflex artery at the level of intertrochanteric groove. The retinacular vessels penetrate the capsule and reach proximally under the retinacular folds supplying the femoral head peripherally and the physis. They are the predominant and important vessels supplying the femoral head\textsuperscript{10}. The main difference between adult and paediatric femoral head supply is that the paediatric femoral head lacks the intra-osseous vascular channels due to the presence of physis which plays a significant role in development of osteonecrosis and prognosis of the injury\textsuperscript{10}.

**Management Options**

**Type 1:** Transphyseal fractures occur in young children after significant trauma\textsuperscript{11,12}. They are different from unstable SCFE (slipped capital femoral epiphysis) which occurs following minor trauma superimposed on a weakened physis from combination of factors like obesity, subtle endocrinopathy etc. Apart from high energy trauma, other reported causes for traumatic physeal separation have been child abuse, iatrogenic (following closed reduction of hip dislocation) etc\textsuperscript{13, 14, 15, 16}. The main factors governing management are age of the patient, presence or absence of femoral head dislocation and stability of reduction obtained. In children under two to three years of age with undisplaced or minimally displaced fractures, management is mainly conservative and spica cast immobilization results in a more predictable outcome\textsuperscript{15,16,17,18}. The limb should be immobilized in mild abduction and neutral rotation in order to prevent the fracture getting displaced into varus and external rotation.

Displaced fractures in this age group must be reduced by gentle traction, abduction and internal rotation. If the fracture locks on and is stable, spica cast is applied without any internal fixation. Close follow-up with repeated radiographs within days will be required to look for re-displacement. In case of unstable fracture, internal fixation with small diameter smooth pins (2 mm) that crosses femoral neck into the physis is recommended\textsuperscript{18}. Sen et al have reviewed the literature and reported excellent prognosis of these injuries in children younger than 3 years of age, in spite of severe displacements\textsuperscript{17}.

If the child is more than 2 years old, internal fixation is recommended even in undisplaced fractures. The reason for recommending internal fixation is to avoid late displacement\textsuperscript{17,18}. Transphyseal fixation is recommended for better stabilization. The choice of implants is smooth pins in young children and cannulated screws in older children and adolescents. As the proximal femoral physis contributes to only 13% of limb length, it is reasonable to trade off for superior fixation in older group (> 10 years of age) without worrying much about limb length discrepancy. Type IB fractures are initially managed by a single attempt at closed reduction and if it fails, immediate open reduction and internal fixation is recommended. The surgical approach is from the side to which the head is dislocated. The fixation is protected in a spica cast except in older children who are reliable and have large
Threaded screws crossing the physis. Transphyseal internal fixations, when used, can be removed after fracture healing to promote further growth\textsuperscript{15, 16, 17}. Transphyseal injuries without dislocation have better prognosis when compared to those with dislocation. In cases of dislocation, the outcome is poor due to complications like Osteonecrosis (ON) and premature physeal closure\textsuperscript{11, 12}.

The significance of early management of type I injuries has been emphasized by many studies and an operative intervention later than 24 hours after the initial trauma may increase the risk of complications such as ON\textsuperscript{19}. Other than factors like initial displacement which play a major role in prognosis, there is not much evidence on the role of factors like the postoperative protocol, e.g., the time interval of limited weight-bearing or an additional cast fixation.

**Type II and Type III:** Intracapsular fractures mandate anatomic reduction and internal fixation. In children under 5 years of age with undisplaced, completely stable type II and III fractures, spica casting and close follow-up for varus displacement may still be an option\textsuperscript{6, 20, 21}. But now internal fixation is recommended even for undisplaced fractures because the risk and consequences of late displacement can induce significant morbidity and considerable socio-economic burden when compared to the risks involved in percutaneous screw fixation\textsuperscript{6, 22, 23}. It is important to obtain anatomical reduction and to internally fix displaced fractures to minimize the risk of late complications. The main complications with immobilization alone are coxa vara and non union\textsuperscript{11, 21, 24}. The rates of these complications are significantly lowered with anatomical reduction and internal fixation\textsuperscript{11, 25, 26, 27}.

For fixation of fracture, 2-3 screws can be used depending on the size of child’s femoral neck and the most inferior screw is inserted in a way that it goes along the calcar\textsuperscript{18, 23}. Unnecessary drill holes in the subtrochanteric region are to be avoided to prevent iatrogenic weakening and subtrochanteric fracture. Fixation across the physis is recommended in type II fractures\textsuperscript{6, 18, 26} for better stabilization. The sequelae of growth arrest due to closure of capital physis and overgrowth of trochanteric physis are not so common and debilitating when compared to complications of inadequate fixation like nonunion, pin breakage and osteonecrosis. If possible, the screws should be inserted short of physis in type III fractures. Paediatric hip compression screw can provide a more stable fixation of distal cervico-trochanteric fractures in children more than 5 years of age. Spica casting is routine except for older reliable children where screws cross the physis\textsuperscript{25}.

**Type IV:** In this pattern the injury is extra capsular and not many complications were reported when compared to other fracture patterns. Nonunion is rare and osteonecrosis occurs in 5% of patients\textsuperscript{1, 6, 11, 21, 28}. Their management protocol is simple and most widely accepted. They are less likely and less documented to be complicated. Most intertrochanteric fractures are treated closed (spica cast) and results are good regardless of initial displacement\textsuperscript{13}. Instability following reduction and multiple injuries are indications for internal fixation. A paediatric hip screw provides the most rigid fixation for this type of injury\textsuperscript{18}.
COMPLICATIONS

Fractures involving the neck of femur deserve focused study because of the significantly high complication rate and the burden of lifetime morbidity imposed by them. Potential complications from the fracture and its treatment include osteonecrosis, coxa vara, non union, delayed physiolysis, growth abnormalities, premature physeal closure, infection, slipped capital physis after internal fixation, heterotopic ossification and limb length discrepancy.

Osteonecrosis (ON) is the most feared and frequent complication (30%) following femoral neck fractures and is also one of the important reasons for poor outcome in the follow-up period. ON was increasingly noted in displaced type IB, II and III fractures. Moon and Mehlman in their meta analysis found the incidence of ON from type I to type IV being 38%, 28%, 18% and 5% respectively. Moon and Mehlman and many others are of the opinion that occurrence of osteonecrosis is directly related to fracture displacement which leads to disruption or kinking of the blood vessels supplying the femoral head. The thick fibrous capsule covering the hip joint is considered less likely to tear than in adult hip fractures. The possibility of intracapsular haemorrhage and tamponade effect reducing blood supply to femoral head due to the tough hip capsule has also been discussed.

Literature favors early management of femoral neck fractures within 24 hours of injury and anatomical reduction and internal fixation. The reason behind this approach may be prevention of additional damage to the tenuous blood supply. Varshney et al, in their study on 21 patients with delayed presentation of more than 24 hours, concluded that the significant variable affecting outcome following delayed presentation and delayed fracture fixation is ON of the femoral head. They also blame open method of reduction as a significant factor resulting in disturbance of blood supply, whereas fracture type, age, and sex do not have significant contribution. Sen et al were also of the opinion that apart from degree of displacement, surgical approach influences outcome and it was concluded that the surgical approach must preserve any remaining blood supply. Contrary to the above mentioned studies, a retrospective review of 27 fractures (15 type-II and 12 type-III displaced fractures) in children younger than 16 years by Song et al had a different conclusion. They recorded that anatomical Open Reduction and Internal Fixation (ORIF) is a more reliable treatment modality than Closed Reduction and Internal Fixation (CRIF) for displaced fractures, producing good results and reducing the rate of ON and other complications. They opined that both capsulotomy and ORIF are effective in preventing ON and further complications. The role of capsulotomy in reducing intra-articular pressure caused by fracture haematoma has equivocal support, some favoring while others not finding significant effect. There are not much evidences on the relation between other factors like age, sex, type of implant used and the development of ON.

Ratliff had classified ON into 3 types. Type I involves the whole head and is the most severe and most common type with worst prognosis. Type II displays partial involvement of the head.
and type III involves the neck from the fracture line to physis. Patients usually become symptomatic in a year, but some cases may remain asymptomatic for up to two years. Little and his colleagues used bone scan for early diagnosis of ON. They also concluded that intravenous bisphosphonates significantly improved the outcome at 3 years follow-up. Sen et al had documented the implication of MRI in the early diagnosis of ON following this injury. There are studies mentioning that if MRI does not show any evidence of ON within 6 weeks of injury, the chances of future development of this complication are remote. According to recent trend, it has been cited as the best prognostic imaging modality. There are no clearly effective and established treatment methods for management of established posttraumatic ON in children and the results are usually poor in over 60% of patients. Delayed weight bearing is still the only treatment outlined for ON diagnosed early. Currently, studies on the role of redirectional osteotomy, core decompression, vascular fibular grafting etc are in the pipeline. Reformation of the upper femoral epiphysis is known especially in types II and III AVN, but is an exception rather than the usual course.

Coxa vara : Approximately 20–30% of patients with fracture neck of femur develop coxa vara. The incidence of this complication is significantly lower in patients who were treated by internal fixation after anatomical reduction. The higher incidence of coxa vara following inadequate stabilization had already been documented by Lam et al and Togrul et al and was mainly attributed to malunion although ON and premature physeal closure may also be a cause for the deformity. In extreme cases there may be limb shortening or inefficient abductor functioning. Remodeling of an established malunion may occur in children less than 8 years of age or with neck shaft angle greater than 110 degrees. Older patients may require subtrochanteric valgus osteotomy. Eberl et al in their follow-up of 22 patients (7 type II and 15 type III injuries) concluded that loss of reduction is the
main factor contributing to coxa vara and also mentioned that even screws are not sufficient in holding the reduction. They recommended interlocking implants for fixation but with not much evidence. Also the importance of crossing the physis for adequate purchase and stability in type II fractures was emphasized.

**Premature physeal closure**: With the incidence of 28%, the risk of premature physeal closure increases with violation of physis or when ON is present. A lot of discussion has been made on whether or not to cross the physis while fixing these fractures. In type II fractures it is considered safe to prefer stability when compared to crossing the physis. The reason being that proximal femoral physis contributes only 13% of lower limb length; hence shortening is not significant except in young children. The sequelae of premature physeal closure is considered less significant and manageable than the complications like loss of fixation and its sequelae. Also there are experimental studies that mention that crossing the growth plate leads to premature closure if the pins are drilled from the epiphysis into the metaphysis and not when the drilling is from metaphysis to epiphysis. But further evidence will be required to substantiate such findings. Only significant limb length discrepancy of 2.5 cms or more (projected at maturity) will require treatment.

**Non-union**: The incidence of nonunion is 7% and is a rare complication. It is seen in types II and III fractures and is not so common in types I and IV. The main reason for nonunion is failure to obtain or maintain an anatomic reduction. If no or minimal healing is seen by 3 to 6 months, the diagnosis of nonunion is established. Treatment consists of rigid internal fixation or subtrochanteric valgus osteotomy. Neto PF et al studied the management of nine children with post-traumatic pseudoarthrosis neck femur (5 type II and 4 type III fractures) and concluded that valgus osteotomy with or without insertion of bone graft is a fairly successful treatment modality in managing these cases.

**Other complications**: The incidence of infection is nearly 1% and is not higher than any other closed fractures treated surgically. Two of the case series had reported chondrolysis as a complication, which may be attributed to penetration of hardware into the joint. Gopinathan et al reported a rare case of SCFE following internal fixation of type II Delbet fracture which was successfully managed by transphyseal fixation and fibular grafting. There are also reports on heterotopic ossification following type I injuries. Heterotopic ossification was by and large reported in children younger than 3 years of age.

**CONCLUSIONS**

The significance of this injury is not only its rarity, but the number of possible complications that may follow compounding the initial injury. From available literature there is enough evidence that an orthopaedic surgeon must consider this injury as an emergency and must intervene at the earliest, possibly within 24 hours. Young children less than 3 years of age with type I injuries and those with stable type IV injuries can be treated conservatively. In type II injuries it is better to cross the physis while fixing the fracture to avoid complications that follow inadequate stabilization. Fixation can be done with smooth...
pins, cannulated partially threaded cancellous screws or paediatric dynamic hip screw depending upon the age of the child and fracture configuration. The fixation may be protected by spica cast immobilization in all but the most mature and cooperative adolescents. The role of capsulotomy is still a controversy with some for and few against it. If stable acceptable anatomic reduction is not achievable by closed means, the fracture should be treated by open anatomical reduction and stable internal fixation. ON is the most common and feared complication. Fracture displacement and delay in reduction are the proven two important contributing factors. MRI may become a vital investigation in early diagnosis of ON and therapeutic measures apart from avoidance of weight bearing are under trial. Although significant progress had been made in the management of these injuries, the rarity of this fracture will keep us waiting for accumulation of more evidence based results.

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INTRODUCTION

Developmental Dysplasia of Hip (DDH) is one of the most common congenital disorders responsible for significant disability in children. Though the ultrasound screening for DDH using Graf’s method in some parts of the world has made early diagnosis and treatment possible, considerable number of patients present as neglected DDH around the world. Despite the advances in the diagnosis and management of the DDH, the basic etiopathogenesis of DDH and acetabulum development remains unexplored. There is ample evidence that glycosaminoglycans and proteoglycans play functional roles in chondrocyte development. Heparin sulfate (HS) and Heparin Sulfate Proteoglycans (HSPGs) have been implicated in morphogen signaling during cartilage development. EXT1/EXT2 association is required for the expression of biologically relevant N-acetylglucosamine (GlcNAc) and glucuronic acid (GlcA) co-polymerase activity. To assess the acetabulum development at early age, rat acetabulum samples were procured at newborn, day 3, day 5, day 7, day 10, day 17 and day 24 of their life. Histology, immunostaining for HSPGs and Reverse Transcription-initiated Polymerase Chain Reaction (RT-PCR) for EXT1, EXT2 and GAPDH were carried out.

The results showed that EXT1, EXT2 and HSPGs expression increased in acetabulum in the first week of life, peaking at day 7 and then decreased gradually, which might indicate that EXT1/EXT2 correlate with the acetabulum development and shape modeling in early stage.
EXT2 along with the histopathology and HSPG expression during early postnatal period in developing rat acetabulum cartilage.

Materials and Methods

Animal:

SD (Sprague Dawley) rats were used in this study. The animal research committee of Fudan university has approved all experimental protocols and animal handling. The pregnant rats were bought from Shanghai laboratory animal center. The experiment was carried out in the animal laboratory of pediatric research center, Children Hospital of Fudan University. The feeding condition was specific pathogen free [SPF], each pregnant rat in one cage with steroid food and water, light/dark cycle each 12 hours on 25°C. After the rats delivered the infants, the acetabulum from both the sides were harvested at newborn, day 5, day 7, day 10, day 17 and day 24 respectively. At each different time point 6 rats were enrolled in the experiment randomly. The rats were sacrificed by CO₂ inhalation method.

Histological section

After euthanasia, the acetabula were dissected out and fixed for 24–48 h in 10% neutral buffered formalin and decalcified in 10% formic acid and 10% neutral buffered formalin at 25°C for 5–7 days. Sections (3 μm) were cut onto 3-aminopropyltriethoxysilane-coated slides (Sigma, Poole, Dorset, UK), deparaffinized in xylene, rehydrated and stained with Hematoxylin and Eosin (H&E) staining.

Reverse Transcription-initiated Polymerase Chain Reaction (RT-PCR):

Total RNA was isolated from acetabular cartilage at different time points by the RNA Extraction kit (Invitrogen, Carlsbad, CA, USA). RT-PCR was performed with 10 μg of total RNA using superscript II reverse transcriptase kit (Invitrogen) as described previously. Glyceraldehyde 3-phosphate dehydrogenase (GAPDH) was amplified as a control. The following primer pairs (forward and backward, respectively) were used: EXT1, GGATTGTTCGTCCTACCGCAGTAT and ATATCCCCAGCGCTCCCTTTGAGTT; EXT2, GCTGTGAAGTGGGCTAGTGTGAGC and ATACTTACTTGCATCTCGTG; GAPDH, ACTCCACTCAGGG- AAATTCAACGG and AGGGGCCGAGATGATGACCC. Amplification was performed as follows: denaturation at 95 °C for 45 seconds, annealing at 61°C (GAPDH) or 56 °C (EXT1 and EXT2) for 45 seconds, and polymerization at 72 °C for 1 minute; 15 cycles for GAPDH and 34 cycles for EXT1 and EXT2. The PCR products were separated by 1% agarose gel electrophoresis.

Immuno-histochemistry:

Immunostaining for HSPGs was carried out with paraffin sections that were first de- paraffinized and then treated with 5 mg/ml testicular hyaluronidase in 5% normal goat serum (NGS) for 1 hour at 37 °C to concurrently de-mask the tissue and block non-specific binding sites. To generate the HS antigen, sections were treated with heparinase III (Sigma) at 5U/ml for 2 hours at 37 °C. Sections were then incubated with anti-HSPGs monoclonal antibody (Abcam, Cambridge, MA, USA) at 1:200 dilution in 3% NGS.
and 5% BSA (Sigma) in PBST (PBS with 0.1% Tween 20) for 3 hours at room temperature. Following rinsing, sections were then incubated with biotinylated anti-mouse IgGs (Abcam) at 1:250 dilution in 3% NGS and BSA in PBST for 1 hour at room temperature. Sections were washed with PBST 3 times and then incubated with ABC reagent for 30 minutes at room temperature. ABC reagent was removed and sections were washed three times in wash buffer for 5 minutes each. DAB showed staining. The images were captured with a light microscope using Image-Pro Plus 5.0 software (Media Cybernetics, Inc.).

Results

1. Histology of rat acetabula at different time points (Fig 1)

The gross morphology and coronal histological sections of the component part of the acetabulum stained with hematoxylin and eosin showed an increase in the depth of the acetabulum during development as the result of interstitial growth in the acetabular cartilage. The epiphyses are seen getting narrowed gradually after day 5.

2. EXT1 and EXT2 expression in acetabular chondrocyte during development

In all developmental time points examined, the amplified PCR bands of EXT1 (474 bp) and EXT2 (447 bp) were detected. The expressional levels increased from newborn to day 7 in EXT1 and EXT2. The peak level of expression was observed around day 5 - day 7. After this period the expression of both EXT1 and EXT2 declined gradually. (Fig 2)

3. HSPGs expression during development

During acetabulum development, the morphology of chondrocytes was round and organized gradually since day 5 compared with
spindle and disorganized in newborn. The expression of HSPGs followed this time pattern. HSPGs expression increased at day 5 and the expression peak was at day 7, then decreased slightly and sustained till 24 days, which might suggest that HSPGs played a role in acetabulum development.

Discussion

In this study we demonstrate EXT1 and EXT2 expression in acetabular cartilage during early development. EXT1 and EXT2 are two genes encoding glycosyltransferases crucial for HS biosynthesis. Although both EXT1 and EXT2 possess N-acetylglucosaminyl- transferase and glucuronyltransferase activities, the heterodimer formation between EXT1 and EXT2 in the Golgi apparatus is required for efficient HS chain polymerization by these molecules. Our RT-PCR results showed that the mRNA expression of EXT1 and EXT2 increased with age from newborn to day 7 and the expression decreased after day 7 and stayed at the same level till 1 month in acetabular chondrocyte. As per our observations, the acetabular morphology modeling occurs at an early age (day 0- day7) in rats, which might indicate that these expressions correlated with the acetabular development and shape formation in early stage.

HSPGs influence the activity of a variety of signaling molecules involved in developmental processes including FGFs, heparin-binding EGF, and members of the Wnt and Hedgehog families. The role of HSPGs in proliferation and differentiation of epiphyses are well studied. HSPGs are key and highly specific developmental regulators due to the structural diversity of their disaccharide polymer, which results from a highly complex series of modifications that includes sulfation, and epimerization. Our data showed HSPGs expression gradually enhanced from newborn to day 7, then expression at same level till 1 month (mature age) in rat acetabular chondrocyte, which suggest that HSPGs might stimulate some signal pathway to regulate acetabular cartilage development. EXT1 and EXT2 in the Golgi apparatus is necessary for assembling HS chain polymerization by these molecules. Further study is being done.

Our data showed the evidence that EXT1 and EXT2 gene expression may have a role in regulating the acetabulum development by HSPGs. 

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Ipsilateral congenital pseudarthrosis of the clavicle and congenital muscular torticollis – A rare association in a child.

Harish Petnikota, Debabratha Padhy, Sunithi Mani, Vrisha Madhuri

Introduction

Congenital Muscular Torticollis (CMT) has a known association with other congenital abnormalities. It presents as a sternomastoid tumor at birth which is gradually replaced by fibrosis and shortening of sternocleidomastoid. Investigations by some authors have shown signal changes in the sternocleidomastoid similar to compartment syndrome in the forearm and leg muscles and they have postulated the sequelae of a perinatal compartment syndrome as etiology.

Congenital Pseudarthrosis of the Clavicle (CPC) is a rare condition. Currently CPC is considered by some authors to result from the failure of fusion of two primary ossification centers due to pressure from a high arched subclavian artery caused by an unduly elevated first rib or a cervical rib. Some form of a vascular etiology is implicated in unilateral pseudarthrosis at the clavicle. As vascular etiology is implicated in both, it is possible that ipsilateral CMT and CPC may be due to the same cause.

We report the concurrent presentation of these two clinical entities in a 3 year old child, successfully treated with cortico-cancellous iliac crest graft and fixation with plate and screws.

Case report

A 3 year old girl presented with progressively increasing, prominent, painless swelling in the region of the right side of her neck and flattening of the face, first noticed at 3 months of age. She had no functional limitation. The child was born at term by a normal vaginal delivery. Her perinatal period was insignificant and developmental milestones were reached at an appropriate age. There was no familial predisposition.

Examination showed a taut right sternocleidomastoid tilting the head to the right and chin to the left and flattening of the right half of the face. There was no strabismus. There was a gap felt in the middle third of the right clavicle with abnormal mobility between the medial and lateral segments. The lateral end of the medial fragment was prominent in the supraclavicular fossa, displaced superiorly by the taut sternocleidomastoid.

Coexisting ipsilateral CPC and CMT have not been documented in the English literature to the best of our knowledge. The etiology, clinical and radiological presentation, relevance of Computerized Tomography (CT) findings and management issues are discussed.
sternocleidomastoid (Fig. 1). There was restriction of right lateral rotation and left lateral bending of the cervical spine.

Plain radiographs of the patient showed pseudarthrosis of the right clavicle at the junction of the middle and lateral third, with proximally displaced medial fragment (Fig.2).

Fig. 2: AP radiograph shows widely separated rounded pseudarthrotic ends and flatness and hypoplasia of the lateral end of the clavicle articulating with acromion

There was a cervical scoliosis to the left side, but no congenital anomalies were noted. A CT scan, in addition to confirming the above, showed hypoplasia of the lateral end of the clavicle, elevation of the first rib, significant flattening of the lateral fragment, no cervical spine abnormality and no obvious abnormality of the great vessels (Fig. 3a & 3b).

Fig. 3a: CT scan 3D reconstruction shows the clavicle ends widely separated with elevation of the medial end, flat lateral end of clavicle with hypoplasia of the acromial end. First rib is elevated.

Fig. 3b: Shows the nonunion to be at the junction of middle and lateral third.

Patient was treated with excision of the pseudarthrosis and the adjacent ends of bone. There was no abnormality of the soft tissue in the region of pseudarthrosis and the gross appearance was similar to a gap nonunion. Internal fixation was done with a contoured quarter tubular plate (AO, Synthes) after a tricortical autogenous bone graft from the iliac crest was interposed in the gap between the fragments. In addition, a unipolar release of lower end of the right sternocleidomastoid was done to bring the medial fragment down for

Fig. 4: 9 months after surgery radiographs show united clavicle and loosening of screws
apposition with the other fragment. After surgery the child was put on head-halter traction for a period of 3 weeks followed by a semi-rigid cervical collar. Stretching exercises for sternocleidomastoid were delayed for 6 weeks because of the fragile fixation.

The fracture united and as the lateral screws were starting to loosen, the implant was removed after 9 months (Fig.4). The bone was found to be well healed at the time of surgery. Part of sternocleidomastoid which was still tight was released at the same time from the proximal end.

The follow up after implant removal showed that the clavicle deformity had improved (Fig.5). Patient had terminal restriction of left lateral bending and right lateral rotation of cervical spine and some facial asymmetry. She retained full function of right shoulder at 16 months follow up.

The unusual association of an ipsilateral CPC and CMT in our case is notable and may shed light on the possible etiology. It is possible to postulate a common vascular etiology for both these conditions. As the second part of the subclavian vessel may be compressed by the elevated first rib, an increase in pressure in the branches of first part of the subclavian artery could predispose to rupture of the muscular branches supplying sternocleidomastoid, leading to a compartment syndrome and torticollis in the perinatal period. Another possible etiology could be a birth fracture of the clavicle going on to pseudarthrosis because of elevation of medial fragment and wide separation in congenital torticollis.

The role of CT scan in congenital pseudarthrosis of clavicle is not completely established as it has been reported in only one case

Discussion

Congenital Pseudarthrosis of the Clavicle (CPC) is an extremely rare condition of unknown cause. The diagnosis is based on the clinical features of a right sided lesion, diagnosed in infancy, without previous fracture and increasing in size with growth, with no major functional loss, and no neurofibromatosis or cleidocranial dysostosis features. Plain radiographs and presence of fibro-cartilaginous ends at the pseudarthrosis confirms the diagnosis. Our patient differed from a classical clinical presentation in having associated congenital torticollis and an atrophic variety of pseudarthrosis.
vessel may be at risk from the surgical procedure because of hypertrophic pseudarthrotic ends of the bone.

Treatment is usually pursued for aesthetic reasons in CPC and in our child the appearance was worsened by associated torticollis. Surgical treatment of a congenital pseudarthrosis of the clavicle includes resection of the pseudarthrotic ends, autogenous grafting and internal fixation. Conservative management is reported to give good functional results, however surgical treatment ensures symmetry and good function with few complications. A block of bone graft was used in our patient to restore the length and to create compressive forces at the host graft interface. The literature shows that the children treated without bone graft do not fare as well and may require a second surgery. The loosening of the implant within 9 months highlights the need for a close follow up and an early implant removal.

Conclusion

This first reported case of congenital torticollis associated with ipsilateral congenital pseudarthrosis of clavicle suggests the possibility of a common vascular etiology. It is necessary to treat both the conditions simultaneously and supplement fixation with bone graft to obtain union and restore length.

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Hyperplastic callus in osteogenesis imperfecta type V- A case report.

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Abstract

Osteogenesis imperfecta (OI) is a disorder of collagen resulting in increased fragility of bone and multiple fractures. The previously proposed Sillence classification had four categories. However a distinct group of type IV has the tendency for formation of hyperplastic callus with or without preceding trauma. These patients do not have blue sclera or dentinogenesis imperfecta but have radioulnar synostosis. This group was then reclassified as OI type V based on clinical, histological and genetic pattern. This hyperplastic callus may sometimes simulate osteosarcoma. Thus, biopsy is needed for diagnosis. However, the hyperplastic callus may resolve on its own, even though it may take years. We report an 8 year old girl of non familial OI type V with hyperplastic callus formation and radioulnar synostosis.

Key words – osteogenesis imperfecta, hyperplastic callus, classification

Introduction

Osteogenesis imperfecta (OI) comprises of a spectrum of inherited disorders of connective tissue with increased osseous fragility. OI was initially classified by Sillence into four categories. In type I OI, the collagen is of normal quality but decreased quantity. The clinical features are blue sclera, hearing loss, dentinogenesis imperfecta and bone fragility that improve with age. Type II is the lethal form in which the patient dies in the intrauterine or neonatal period. Type III has triangular face, blue sclera, hearing loss, barrel shaped chest and progressive respiratory failure. Type IV is characterized by moderate bone fragility, short stature and hearing loss.

Glorieux et al further proposed three new categories based on the genetics, which were previously grouped under type IV. The type V has characteristic mesh like appearance in histology. Other features are radio-opaque band near growth plate, hypertrophic callus and calcification of radioulnar interosseous membrane. Type VI has characteristic fish scale pattern of bone lamellae. Features of type VII are small head, exophthalmos, coxa vara, rhizomelia and progressive respiratory insufficiency. Cabral et al further proposed type VIII, due to mutation of another protein Leprecan. Hyperplastic callus (HPC) formation is a rare complication, characterized by exuberant new bone formation after a fracture, nailing or spontaneously which happens in type V OI. In this report we present an eight year old girl, a case of osteogenesis imperfecta with hyperplastic callus formation and radioulnar synostosis.

Case report

An eight year old girl born by normal vaginal delivery and with normal developmental
milestones presented with progressive swelling in the right thigh of one and half year duration. She also complained of progressive deformity and stiffness of right knee for eight months, due to which she gradually became bedridden. She had two episodes of forearm fractures at two and three years of age following trivial trauma.

Radiography showed large fluffy irregular masses arising from right femur, periosteal reactions in proximal femur diaphysis. There was associated warmth, local hypervascularity and 40° flexion contracture of right knee. The radial head was dislocated anteriorly on both sides. Sclera and teeth were normal.

Radiography showed large fluffy irregular masses arising from right femur, periosteal reactions in

Examination showed a bony swelling with maximum diameter 12 cm arising from posteromedial aspect of right distal femur, in metaphysio-diaphyseal region (figure 1).

Another smaller bony swelling of maximum diameter six cm was there on anterior aspect of left femur, bilateral proximal radioulnar synostosis with radial head dislocation (figure 2) and platyspondyly (figure 3). Laboratory investigations were normal other than mild elevation of alkaline phosphatase (367 IU).

Figure 1:  a) Diffuse swelling in the right thigh b) X ray of the right femur at initial presentation showing two large irregular radiodense masses one from proximal and one arising from distal part of right femur resembling parosteal osteosarcoma. There was also diffuse periosteal reaction present in the whole femur
Biopsy was done to rule out osteosarcoma. Histopathology showed haphazardly arranged lamellae with large spaces, plump osteocytes and scanty mineralization. Thus a diagnosis of hyperplastic callus was made.

Serial casting was done to correct the flexion deformity and patient was mobilized with knee ankle foot orthosis. At the recent follow up the callus size appears to be decreasing.

Figure 2: X ray and clinical picture of both elbows with forearms showing bilateral proximal radioulnar synostosis and radial head dislocation

Figure 3: Lateral view of thoracolumbar spine showing osteopenia and platyspondyly affecting most of the vertebra with normal disc space.
Discussion

Hyperplastic callus remains a rare event in osteogenesis imperfecta since its first report in 1908. It was initially found in type IV of Sillence classification. However later, hypertrophic callus was found to be a striking feature of type V.

Type V OI is a moderate to severe variety characterized by formation of hyperplastic callus, presence of radial head dislocation, radioulnar synostosis, a metaphyseal band at growth plate and normal sclera and teeth. The unaffected bone shows large variation in the collagen fiber thickness and lamellation. Though found to be autosomal dominant in inheritance, no structural mutations of the collagen type I gene has been found.

Clinically, it may simulate osteosarcoma due to rapid growth, pain, local warmth and dilated veins. CT and MRI may show sunburst like calcification, mild edema and strong enhancement of adjacent soft tissue. Biopsy settles the diagnosis showing four zones in actively growing callus. The first zone shows edematous muscle with loose collagen, the second mucoid edema and proliferating fibroblasts, the third chondroid and fibrochondroid tissue with nonanaplastic nuclear polymorphism and the inner most has hyper cellular trabeculae with occasional chondroid islands. Biochemically active growing callus shows elevated DNA content but lower protein. Collagen is over-hydroxylated and markedly reduced in content.

Once diagnosed, the hyperplastic callus needs close observation. The evolution may take months to years and it may resolve on its own. The most common complication is fracture through the lesion.

Even though bisphosphonates and radiation have been tried, an effective treatment is yet to be found. Contractures and short stature may result in significant disability and bracing may be helpful. In our case, serial casting and brace helped the patient with ambulation. Surgical management of the synostosis and radial head dislocation is not satisfactory.

Hyperplastic callus when seen after a fracture should alert the clinician of a moderate to severe variety of osteogenesis imperfecta which presents without the hallmark features of blue sclera and bad dentition. It should be differentiated from a malignant bone tumor. No active management is needed in most of the cases.

REFERENCES:


Foot radiograph shows a large accessory ossicle on the medial aspect of the big toe associated with hallux valgus on the right side. Cervical spine x-ray shows soft tissue ossification under the dorsal skin adjacent to T1 vertebra. Findings of brachydactyly clinically with mild hallux valgus and accessory ossicle are characteristic features seen in children with Fibrodysplasia Ossificans Progressiva (FOP). Blood investigations in this child are within normal limits except for a mild elevation of LDH. These radiographic findings combined with stiffness of the axial skeleton involving the neck are suggestive of FOP, also known as myositis ossificans progressiva or Stone man syndrome.

Fibrodysplasia Ossificans Progressiva is a very rare and debilitating disease caused by the heterozygous missense point mutation of ACVR1 gene located at chromosome 2q23–24\textsuperscript{1,2,3}. It is characterized by a peculiar congenital abnormality of the big toe and abnormal bony masses developing in muscles, predominantly in axial skeleton, eventually leading to thoracic insufficiency and death by second decade\textsuperscript{1,2,3}.

The child presents clinically either in the flare up stage having acute onset with painful, inflamed, bony swelling causing stiffness around joints or in the quiescent stage of the disease in which the stiffness due to the bony masses is the only symptom\textsuperscript{4,5}. Occasionally in the quiescent stage, fibrous bands are seen causing restriction of the shoulders, hips and spine movements leading the surgeon to do excision and biopsy with deleterious effects. Biopsy is non-diagnostic in most instances.

The diagnosis can be confirmed by the mutation analysis for the ACVR1 gene. A high degree of suspicion should be entertained in children with great toe abnormalities even before the onset of ossification. A genetic diagnosis of FOP in children having great toe anomaly even before the onset of ossifications can be made by doing a genetic work up to look for ACVR1 mutation. This can postpone the symptomatic
disease with the help of medical management.

There is no curative treatment available at present for FOP. Management is aimed at prevention of flare ups and thereby new ossifications.

Flare ups and the new ossifications are induced by any kind of injury to the muscles and soft tissues such as trauma, biopsy, intramuscular injections and aggressive physiotherapy. Therefore, elective surgeries and intramuscular injections are contraindicated in these children. Superfluous attempts by some clinicians to biopsy or excise the lesions can be avoided by counselling the parents and the children regarding the nature of the disease and its prognosis.

Flare ups are managed with a short course of prednisolone 2mg/kg/day for 5 days. The cyclic bisphosphonates (alendronate and pamidronate) along with daily mast cell stabilizers (Montelukast) and non-steroidal anti-inflammatory drugs have shown promising results in some studies to maintain the quiescence and prevent further flare ups. Though subjective and objective improvement with medical management is reported, further studies are required for establishing management protocols.

REFERENCES:


Type II Supracondylar Humerus Fractures: Can Some Be Treated Nonoperatively?

Hillard T. Spencer, Dorey FJ, Zions LE, Dichter DH, Wong MA, Moazzaz P, Silva M.


Study type: Therapeutic

Study design: Retrospective analysis of data from a prospective registry of elbow fractures.

Level of evidence: Level III

Purpose: To identify the type II supracondylar fractures humerus which can be managed nonoperatively and to distinguish fracture characteristics which can lead to late displacement and hence warrant surgical management.

Population: 258 children with 259 type II supracondylar fractures humerus who presented to an outpatient walk-in urgent care center.

Intervention: Closed reduction and casting or closed reduction and pinning of the fractures based on the clinical and radiographic parameters.

Control: Comparison between the closed reduction group and the operated group.

Outcomes assessed: Carrying angle, range of motion and complications.

Results: 189 (57.9%) cases were managed nonoperatively and 70 (42.1%) cases underwent operative management. Of the 189 children initially managed conservatively, 39 needed closed reduction and pinning during the course of treatment due to coronal or saggital plane malalignment in follow-up radiographs. At final analysis, there was no difference in the carrying angle, range of motion or complications between the treatment groups.

Authors’ conclusion: Fractures without coronal angulation (based on Baumann’s angle/visual judgement), rotation, major extension of distal fragment (based on shaft condylar angle < 15°) or gross swelling can be managed with closed management.

Remarks: The current trend of fixing all type II supracondylar fractures can lead to unnecessary surgeries in at least 58% of cases. This can indirectly increase the chances of iatrogenic injuries, cost and loss of work hours for the parents. This article presents a cohort of type II fractures which, based on clinical and radiographic criteria, need pinning to prevent complications. Commination of medial column should have been included as a confounder as it affects the stability of the fracture. By stratifying the severity of this injury it would be possible to avoid unnecessary interventions.

Loss of a condyle of the femur or tibia following septic arthritis in infancy: Problems of management and testing of a hypothesis of pathogenesis


Study Design: Retrospective case series supported by an anatomical study

Purpose: To hypothesise the reason for loss of a single femoral or tibial condyle in post septic
sequelae of the knee in infancy and to study the radiological features, management problems and the outcome after surgery.

**Methods:** Retrospective study of records of 8 children (record period of 20 years) with loss of a femoral or tibial condyle following neonatal septic arthritis, including 2 children who underwent surgical procedures for reconstruction of the condyle. Cadaveric dissection of 35 stillborn fetal knees (between 20 to 40 weeks) to look for complete vertical septum dividing the joint. The synovial plicae were studied and graded.

**Results:** The radiological pattern of femoral or tibial condyle damage showed that only a single femoral or tibial condyle was affected. Either the medial or lateral femoral condyle was affected while in the tibia only the medial tibial condyle was affected. In one child, a pedicled patella combined with metaphyseal bone graft separated by fat graft was used to reconstruct the tibial condyle. In the other child, the fibula was transplanted to replace the femoral condyle with its physis. In both case the outcome was not satisfactory.

The cadaveric study showed that 16% of the knees had a well formed, complete infra patellar septum separating the knee into medial and lateral half compartments.

**Authors’ conclusions:** The authors hypothesise that the presence of a complete synovial septum could lead to localized infection on one side of the joint, leading to loss of one side of the condyle. The outcomes of reconstruction of the missing condyle were poor.

**Remarks:**
1) If one half of the joint was involved, it is not explained why either only the femoral or tibial half of the condyle and not the other corresponding condyle was involved in any of the eight cases.

2) Since all these cases were not seen immediately after the infection but after some time, there is a chance of physeal arrest. In the cases shown, there is either a proximal or distal migration of the physeal scar and it is not clear whether there is presence of physeal cartilage with the articular cartilage present on it suggestive of physeal arrest. An arthrogram to look for the joint and the presence of a complete septum would have been informative.

**Open reduction and internal fixation of unstable slipped capital femoral epiphysis by means of surgical dislocation does not decrease the rate of avascular necrosis: A preliminary study**

Cristina Alves, Marie Steele, Unni Narayanan, Andrew Howard, Benjamin Alman, James G. Wright.


**Study type:** Therapeutic

**Study design:** Retrospective study

**Level of evidence:** Level III

**Objective of the study:** To determine the rates of Avascular Necrosis (AVN) after open reduction and safe surgical dislocation and comparison with AVN rates after closed reduction and percutaneous pinning.

Intervention: 6 patients underwent closed reduction and percutaneous pinning and 6 underwent open reduction by surgical hip dislocation.

Outcome measured: Incidence of AVN after open reduction with surgical dislocation technique compared with the AVN rates after standard treatment i.e closed reduction and percutaneous pinning.

Results: 4 patients (66.7%) had avascular necrosis in the surgical dislocation group, while 2 patients (33.3%) had AVN of femoral head in the group with closed reduction.

Authors' conclusion: The rates of AVN do not decrease with open reduction and surgical dislocation technique as compared to the earlier standard of closed reduction and percutaneous pinning for treatment of unstable SCFE.

Remarks: This study reports the experience of a centre which introduced a new surgical technique in the treatment of unstable SCFE. They state that although the surgical dislocation technique has been reported to be safe while treating other hip conditions, their experience is that they cannot confirm that it changes the natural history of unstable SCFE or that it has any advantage over closed reduction when considering the rate of AVN. The authors note that their study population consisted only of truly “unstable SCFE” patients who could not rise from a recumbent position, in whom any hip movement was severely painful and there was free movement between the head and the neck observed during fluoroscopy and at the time of open reduction. Their contention is that the earlier studies had included all “acute” SCFE which were not truly unstable according to Loder’s original criteria, resulting in lower rates of AVN. They also had a control group in the same patient population who underwent closed reduction and percutaneous pinning. They acknowledge that theirs was a retrospective study with four different surgeons performing the procedure and all patients presented late (i.e. more than 8 hrs after the injury). They conclude that this new technique has a significant learning curve and surgeons should acquire adequate training and remain cautious as they adopt this new methodology.

Pes planovalgus deformity surgical correction in ambulatory children with cerebral palsy

Muayad Kadhim, Laurens Holmes Jr., Chris Church, John Henley, Freeman Miller.


Centre: Department of Orthopaedic Surgery, Alfred I. duPont Hospital for Children, Wilmington, USA

Study type: Therapeutic

Study design: Retrospective cohort study

Level of evidence: Level III

Purpose: To compare the effectiveness of subtalar fusion and calcaneal lengthening, and to assess the recurrence in ambulatory children with cerebral palsy.
Radiographic angles. Age at the first surgery was a significant predictor of recurrence after adjustment for the BMI. Consequently, for every one-unit increase in age, there was a 22% decreased risk of recurrence, OR = 0.78, 95% CI (0.62–0.97), p = 0.03.

Author's conclusion: Severe and rigid planovalgus feet can be treated effectively with subtalar fusion. Feet with milder deformity show good results with calcaneal lengthening. Surgery provides good correction in young patients, but there is a higher recurrence rate.

Remarks: This study states that although the indications for calcaneal lengthening osteotomy and subtalar arthrodesis are different, there is no difference in radiological, functional and gait analysis.

Baumann angle and radial - ulnar overlap: A radiographic study to control for the angle of the X-ray beam

J. Lee Pace, Wiater B, Schmale G, Jinguji T, Bompadre V, Krengel W III.

Results: There was a statistically significant change in the preoperative CPPI, medial midfoot pressures and heel impulse in the whole sample, with remarkable improvement at last follow-up (p < 0.0001). There was also significant change in radiographic angles. There was a slight change in stride length and velocity at the last follow-up compared to preoperative status. However, by repeated analysis of variance (ANOVA) with Bonferroni correction for multiple comparisons, there was no significant effect of surgery type on the CPPI [F (1) = 0.043, p = 0.84], heel impulse or radiographic angles. Age at the first surgery was a significant predictor of recurrence after adjustment for the BMI. Consequently, for every one-unit increase in age, there was a 22% decreased risk of recurrence, OR = 0.78, 95% CI (0.62–0.97), p = 0.03.

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Baumann angle and radial - ulnar overlap: A radiographic study to control for the angle of the X-ray beam

J. Lee Pace, Wiater B, Schmale G, Jinguji T, Bompadre V, Krengel W III.
Population studied: 71 radiographs of uninjured normal elbows of children in the age group of 0-12 years (these radiographs had equal representation from boys and girls and had an equal distribution through all age groups).

Intervention:

Measurement of predefined relationships between various radiographic measurement points on AP and lateral views as well as the standard measures of lateral capitello humeral angle and Baumann’s angle on the films was done. After an initial analysis of different radiographic measurements, a significant correlation was found between Baumann’s angle and radial - ulnar overlap (the overlap between the medial border of the radius and the lateral border of the ulna at the level of the radial tuberosity on the anteroposterior elbow x-ray). These measurements were repeated on all 71 radiographs by five different observers.

Outcomes: Relationship between Baumann’s angle and Radial Ulnar Overlap (RUO) on a true AP elbow x-ray.

Results: There was significant linear correlation between Baumann’s angle and Radial - Ulnar Overlap which can represented by: Corrected BA = Measured BA - 12.36 (RUO - 0.34). On a true AP view, the Radial- Ulnar Overlap at the level of the tuberosity should approximate 0.33 and be within the range of 0.1 to 0.5.

Authors' conclusions: RUO is a reliable surrogate measure to control for the angle of the x-ray beam and improve the reliability when measuring Baumann's angle that can be easily applied in the clinical setting. The ability to accurately correct the Baumann angle based on a given Radial Ulnar Overlap gives the physician the confidence to adequately interpret imperfect radiographs in the emergency room setting and minimize repeat radiographs.

Remarks: This study provides a good technique to assess the Baumann’s angle and also decreases the need of repeating x-rays to get the perfect view to measure Baumann's angle. However the authors point out that this technique needs to be validated by a prospective study design in healthy volunteers which may have ethical issues related to it.
An eight year old boy, born of consanguinous marriage, presented with swelling of small joints of both the hands from 4 years of age and mild discomfort on prolonged walking. There was no history of fever or constitutional symptoms. The child walked with a waddling gait. Swelling of interphalangeal joints of hands and 10-15 degrees fixed flexion deformity of proximal and distal interphalangeal joints of the ring, middle & index fingers were noted. There was mild swelling of both the elbows. The range of motion of elbows, hips and ankles were terminally restricted. There was no local tenderness or warmth of any of the joints. Radiographs showed bilateral hip coxa vara, anterior beaking of lower dorsal and lumbar vertebral body, Os trigonum behind the talar body and metaphyseal widening with epiphyseal flattening in the proximal and middle phalanges of both hands (Figure 1). The blood investigations are as follows. What is the clinical diagnosis and line of management?

<table>
<thead>
<tr>
<th>Test</th>
<th>Patient value</th>
<th>Lab normal reference value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hemoglobin</td>
<td>12.9 gm%</td>
<td></td>
</tr>
<tr>
<td>WBC total count</td>
<td>8600/cu mm</td>
<td></td>
</tr>
<tr>
<td>Differential count</td>
<td>N 64% E 5% L 28% M 3%</td>
<td></td>
</tr>
<tr>
<td>Rheumatoid factor</td>
<td>&lt; 10.1</td>
<td>&lt;20</td>
</tr>
<tr>
<td>C- Reactive Protein</td>
<td>&lt; 3.41</td>
<td>&lt;6</td>
</tr>
<tr>
<td>Anti Streptolysin-O</td>
<td>&lt; 50.9</td>
<td>&lt;300</td>
</tr>
<tr>
<td>Anti Nuclear Antibody</td>
<td>NEGATIVE</td>
<td></td>
</tr>
<tr>
<td>Vitamin D</td>
<td>95.47</td>
<td>20-32</td>
</tr>
</tbody>
</table>

Figure 1: Plain radiographs of both hands with wrist, ankle, spine and hips.

ANSWER IN THE NEXT ISSUE
India is a country in south Asia. It is the seventh-largest country by area, the second-most populous country with over 1.2 billion people, and the most populous democracy in the world. That is what I learned in textbook. I did not expect I would have chance to feel it until APOA chose me as a travelling fellow. On September 19th, my traveling started and the first stop was Mumbai.

Ganesh Chaturthi, the elephant god, was the first impressive name to know because it was the first celebration day when I arrived in Mumbai. The birthday of Ganesh is a 10-day extravaganza all across India. Mumbai is a good place to catch the fervor, as thousands of devotees head to the beaches and dip their idols of Ganesh in the sea to the hypnotic backbeat of drumming, singing and dancing in all over the city till midnight.

After two and half hours of observation in the hospital, we went to the second stop, Manipal, by flight to Mangalore first, then by car. I called Manipal, a cute green town. We could see rainforest everywhere. We were kindly invited to the temple that was built five hundreds years ago. I believe that it is one of wonders in the world. At different time point, the sun light shines on a different god bypassing the door. And we stood on the warmest and coldest point of the building. It was an amazing experience. And I ate lunch by hand first time in the temple, which was very traditional south Indian food.

As planned, an overnight train transferred us from Manipal to Vellore on September 26. It was a nice train and our tickets were first class. My companion, Dr Nick, experienced his first attempt on the train in his life. He looked nervous, curious, and funny.

Vellore is one of the oldest cities in south India, and lies on the banks of the Palar river on the site of Vellore fort. Vellore has many colleges, ancient temples, a renowned hospital (the Christian Medical College & Hospital) which we visited. Vellore is very close to Chennai, our fourth stop. We drove three hours on the new highway by car, and then we settled down in a very nice boating club. The river was in front of room window and the birds were flying freely.

We spent the longest time in Chennai, because the travel to Pakistan was cancelled. Chennai is the capital city of the Indian state of Tamil Nadu. The area around Chennai had been part of successive South Indian kingdoms through centuries. We visited the temple that was built in one stone 1300 years ago. Unbelievably we saw a stone stood steadily on one point. We also learnt the meaning of the second largest stone inscription.

Delhi was our last stop. Time was flying. We have been in India almost three weeks. Our experiences were not only the extensive journey and history that we learned and saw, but also lots of friends whom we made.

Thanks APOA organizer a lot and thanks all of the hosts very much in these cities. I had a wonderful time and very good experience in India. I enjoyed here the history, the temples, the scenery, the clinical knowledge and all my friends.
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