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Case Report

Avulsion fracture of the tibial tubercle in a child with testicular feminization syndrome

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ABSTRACT

Apophyseal avulsion fracture of the tibial tuberosity is rare. It often affects athletic adolescent males approaching skeletal maturity. This injury has been reported in children with existing or previously treated Osgood-Schlatter's disease (OSD) and osteogenesis imperfecta. We present an adolescent boy with an avulsion fracture of the tibial tubercle with testicular feminization syndrome (TFS) and have been previously treated for OSD. We report a late presentation of a rare tibial tuberosity fracture with intra-articular extension in a 15-year-old athletic boy with TFS and a previous history of OSD. He was managed by open reduction and internal fixation under image guidance. Young general practitioners should have a high index of suspicion when children present with knee pain and swelling following sporting activities in order not to miss such an injury.

Keywords: Athletic injury, Avulsion fracture, Osgood-Schlatter's disease, Testicular feminization syndrome, Tibial tuberosity

INTRODUCTION

Avulsion fracture of the tibial tuberosity is a rare pathology. It constitutes about 1% of epiphyseal injuries and 3% of proximal tibial fractures.[1] Physically, active adolescent males with well-developed quadriceps muscles approaching skeletal maturity are frequently affected more than females. Osgood-Schlatter's disease (OSD) and osteogenesis imperfecta (OI) have been reported to be associated with this pathology. OSD has been reported to occur in about 23% of the cases, but the exact incidence still needs to be ascertained and no causal relationship has been established.[1-3] The mechanism of injury is associated with extreme quadriceps contraction against a fixed foot and sudden knee flexion against a contracted quadriceps. Both mechanisms can occur during sporting activities such as sprinting, jumping, gymnastics, football, and others. However, the fracture pattern will depend on the extent of skeletal maturity and degree of knee flexion during injury.[1,2]

Several classifications exist but are based on that proposed by Watson-Jones.[1,2] These classifications consider the growth phase and presence of a physis. The Watson-Jones classification describes the fracture relative to the physis proposing three types of fracture (Types I, II, and III), while Ogden *et al*., in 1980, modified it by adding subclass A (non-comminuted) and B (displaced and comminuted) to each type.[4] In 1985, Ryu and Debenham added type IV

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to the list.[5] Then, Mckoy and Stanitski, in 2003, proposed a type V.[6]

The treatment principles involve restoring the extensor mechanism, congruent joint surface, and meniscal mechanism if compromised.[1,2] Treatment can be operative and non-operative. The non-operative option is not very popular but will depend on the level of displacement and classification. The complications associated with this fracture include compartment syndrome, knee stiffness, genu recurvatum, anterior knee pain, non-union, mal-union and growth retardation (extremely rare), and others.[1,2]

Testicular feminization syndrome (TFS), *aka* "Androgen insensitivity syndrome (AIS), is the most common cause of disorder of sex development in 46, XY individuals." This presents clinically as three phenotypical variants: Mild AIS (MAIS), partial AIS, and complete AIS.[7] We report the case of a 15-year-old boy with 2 weeks history of tibial tuberosity avulsion fracture that was under treatment for TFS and had been previously treated for OSD. We have searched the literature and found no reported occurrence of this pathology with sex hormone syndrome. We report a case of a child with this pathology with sex hormone syndrome.

CASE REPORT

We present the case of a 15-year adolescent boy who presented with knee swelling, anterior knee pain, and limping gait on his right leg for 2 weeks. Examination findings were tender prominent tibial tubercle, joint effusion, and patella alta with knee range of motion limitation due to pain. The left knee was normal. He was under treatment for pubertal gynecomastia with hormone replacement therapy. Furthermore, we have previously managed him for OSD. He is an active sportsman who plays both basketball and football. He sustained the injury during a basketball tournament, where he jumped to catch the ball and on landing, he attempted to run forward immediately. He had a sharp pain and heard a snap at the right knee joint, which was associated with anterior knee swelling. He weighs 70 kg and he is 175 cm in height. He self-medicated and received treatment from a traditional bone setter. Radiographs and blood investigations were ordered, which revealed an avulsion fracture displacement of the right tibial tubercle through the physis with an intra-articular fracture of the proximal tibial epiphysis and a high riding patella, suggestive of Ogden type IIIA [Figure 1]. The parent consented to surgical management. Intraoperative findings included anterior-superior tubercle displacement with callus formation and proximal tibial epiphyseal fracture displacement. The intra-articular fracture hematoma was evacuated, and the tibial tubercle cavity filled with callous was curetted for easy reduction. Some fibers' of the patella tendon were torn but not significant to disrupt its function. The tubercle was fixed using a cancellous lag screw with a washer, while the intra-articular extension was fixed with a lag screw under image guidance [Figures 2 and 3]. The knee was immobilized with a rigid brace at 5° flexion and mobilized on bilateral axillary crutches for 4 weeks. He was referred for physiotherapy. He was followed up for 4 months.

At the last visit, his knee range of motion had been restored and he now ambulates without any walking aid. Follow-up radiographs show ossification of the tibial tuberosity and union of the proximal tibial epiphyseal fracture [Figure 4].

DISCUSSION

Avulsion or traction fracture of the tibial tubercle is an uncommon pathology affecting children from 9 to 17 years old with immature physis to physis approaching skeletal maturity.[1,2] The age of our patient corresponds to the time of growth plate closure and maturation of the fibrocartilaginous attachment of the tuberosity. During the transitional phase of closure, the tibial tubercle physis is vulnerable to injury.[1]

Studies and reports have shown that this pathology is more common in males than females.[2,3] However, trauma is more

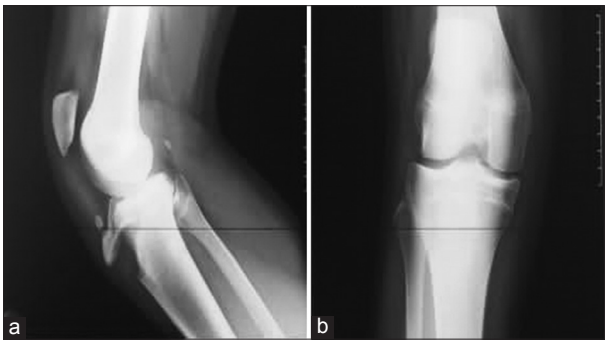

Figure 1: (a and b) Radiograph of the right knee showing displaced tibial tubercle avulsion fracture with intra-articular fracture of the proximal tibial epiphysis.

Figure 2: Intraoperative images post-fixation.

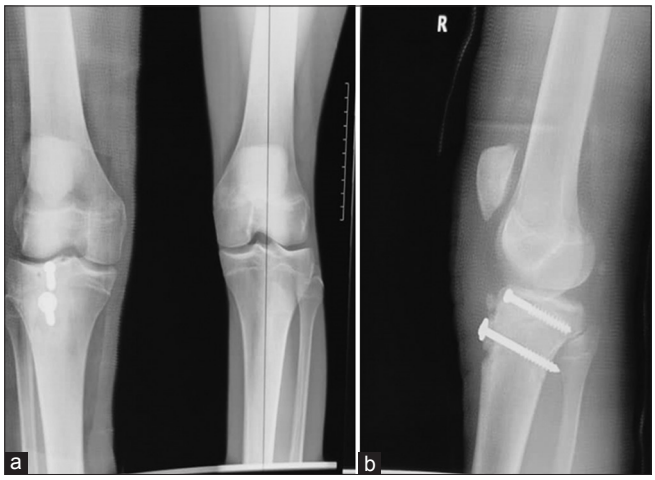

Figure 3: (a and b) Post-operative radiograph showing well-reduced fracture.

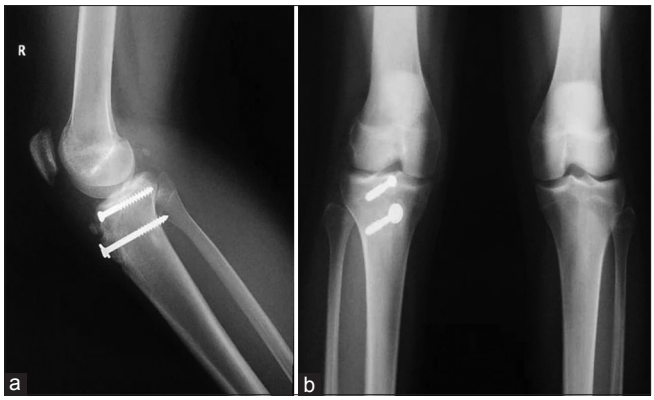

Figure 4: (a and b) Follow-up radiograph showing union with ossification of the tibial tuberosity and proximal tibial epiphyseal fracture.

common in males than females due to their involvement in more contact and risky activities.[8]

In our report, the injury affected the right lower limb. This is similar to that reported by some studies in the USA and Africa.[2,3,9] No explanation was given for these findings in these studies. This may be related to the dominance of the limb.

The index patient had a previous history of OSD. Most authors have consistently reported an occurrence of the injury in children with OSD. However, no causal relationship has been proven.[1-3] He is also being managed for pubertal gynecomastia, which is associated with MAIS.[7] Estrogens play a role in stimulating physeal closure/fusion and cessation of growth. However, androgens, on the other hand, promote longitudinal growth by delaying physeal closure, including the apophysis. The exact mechanism by which sex hormones affect the physis has not been fully understood, but numerous mechanisms have been postulated.[10] This syndrome can be indirectly associated with the pathology, even though no causal relationship has been established.

Our patient sustained the injury during a sporting activity as he plays competitive basketball and football. This is similar to findings reported by other authors.[2,3]

The patient presented late after seeking alternative treatment from traditional bone setters with no remarkable improvement. In most developing countries, late trauma presentation is common and sometimes patients present with complications. This was a finding in a Nigerian study[8] that was different from other studies.[2] Early diagnosis and treatment are the mainstays in obtaining good functional outcomes.

In our report, we repaired both the tibial tubercle avulsion and the intra-articular component with screws, but we did not repair the ligament disruptions because it was negligible. Some authors suggested addressing the three injuries, especially in Ogden type III fractures.[1,2,9]

With reference to the Ogden classification, most of the studies report the prevalence of type III injury.[2,3] Our patient had a type IIIA (fracture displacement of the right tibial tubercle through the physis with an intra-articular fracture of the proximal tibial epiphysis) injury. There is no explanation for the observation, but it could be due to the shear strength of the quadriceps mechanism.

CONCLUSION

From our report, TFS, as well as OSD, can be associated findings in athletic adolescent males with a tibial tubercle avulsion fracture. There has been no proven causal relationship yet, but only time will tell if many cases are reported with these associations. Early diagnosis and treatment are keys to a good functional outcome. Young, upcoming surgeons and general practitioners should have a high index of suspicions, especially if a child presents with anterior knee pain following trauma.

RECOMMENDATIONS

Osteogenesis imperfect, Osgood-Schlatter's disease, and probably TFS can be associated with tibial tubercle fracture, and the parents need to be counseled.

AUTHORS' CONTRIBUTIONS

All authors participated in the report conception. Literature reviews were done by BJA, TU, and EJA. The review of past medical journals and examination of the patient was done by JA, TU, and BJA. The first draft of the manuscript was written in parts by all authors. All authors made inputs to the previous version of the manuscript. All authors have critically reviewed and approved the final draft and are responsible for the manuscript's content and similarity index.

USE OF ARTIFICIAL INTELLIGENCE (AI)-ASSISTED TECHNOLOGY FOR MANUSCRIPT PREPARATION

The authors confirm that there was no use of Artificial Intelligence (AI)-Assisted Technology for assisting in the writing or editing of the manuscript and no images were manipulated using the AI.

ETHICAL APPROVAL

The research and ethics committee of the institution gave a waiver since the patient's privacy was protected, and no trial or new treatment was contemplated.

DECLARATION OF PATIENT CONSENT

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's parent has given his consent for the patient's images and other clinical information to be reported in the journal. The parent understands that the patient's name and initials will not be published, and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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CONFLICTS OF INTEREST

There are no conflicting relationships or activities.

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