CASE REPORT
PATHOGENESIS AND RADIOLOGICAL FINDINGS IN RARE CASE OF SALTER HARRIS TYPE I DISTAL TIBIAL FRACTURE WITH ASSOCIATED OSTEONECROSIS IN THE PAEDIATRIC POPULATION

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Salter-Harris type I fractures of the distal tibia are commonly seen in paediatrics and management of such fractures follows an algorithm established in the literature. Despite this, osteonecrosis of the distal tibia can subsequently develop. Osteonecrosis or avascular necrosis is cell death that occurs secondary to trauma, metabolic disturbances, sickle cell disease, or medication side effect. It most frequently affects the femur, talus, or humerus, and rarely the tibia. Radiographs and MRI are pivotal in making a timely diagnosis in order to minimize patient discomfort. To the best of our knowledge, there has only been one previous documented case of osteonecrosis following a Salter Harris Type I distal tibial fracture. Here, we present the second such case.

Keywords: Salter Harris Fracture; Osteonecrosis; Distal Tibial Fracture

INTRODUCTION
Osteonecrosis or avascular necrosis is a phenomenon which occurs due to cell death causing architectural osseous collapse. This presents clinically in a patient as arthralgias. Osteonecrosis is most commonly seen as a result of trauma. However, it may also be associated with non-traumatic conditions such as sickle cell anaemia, metabolic disturbances, vascular or rheumatologic disorders like systemic lupus erythematosus, alcoholism, infections, corticosteroid use, hyperbaric events and coagulation defects. Post-traumatic osteonecrosis is most commonly seen in the femoral head, talus, and proximal humerus. Although exceedingly rare, osteonecrosis can also occur with distal tibial fractures in children. One such case has been reported in the literature. The pathophysiology of osteonecrosis begins with an interruption of the blood supply to a localized area of the bone. However, once the infarct becomes established, a central necrotic core develops which is then surrounded by an ischemic zone. Beyond the boundaries of the ischemic zone, the bone marrow is viable. Between the normal viable bone marrow and the ischemic zone, demarcation occurs with development of viable granulation tissue.

Approximately 15% of Salter Harris (SH) I fractures in the paediatric population involve the distal tibial physis. Radiographs can be used initially in the diagnosis; however, they are less sensitive than Magnetic Resonance (MR) Imaging. A plain film radiograph for SH Type I fracture demonstrates widening of the physis and/or displacement with overlying soft tissue swelling. MR findings for SH type I fracture demonstrate an increased T2 signal within the physis. Comparison to other physes in the field of view on the MR sequences can be helpful in making the diagnosis. A linear fracture in the metaphysis and/or epiphysis usually demonstrates decreased signal on T1 and T2 sequences surrounded by marrow oedema.

CASE PRESENTATION
A 12-year-old male presented to the Emergency Department with left ankle oedema and inability to bear weight after trauma. Persistent left-sided gait instability was observed by the parents. The patient reported having left ankle and lower leg pain. On physical exam, strength with dorsiflexion and plantar flexion was 5/5 bilaterally. An antalgic gait was noted, with the foot progression angle externally rotated. Initial ankle radiographs demonstrated widening and irregularity of the left distal tibial physis when compared to the right distal tibial physis, which appeared grossly unremarkable (Figure-1). This was suggestive of a distal tibial SH Type I fracture. A cast was placed over the left leg and the patient was sent home. The patient returned two months later, reporting persistent ankle pain that worsened with activity.

On follow-up examination, there was no point tenderness overlying the distal tibia. Past surgical and past medical history were non-contributory. Follow-up left ankle radiograph (Figure-2), demonstrated interval increase in widening and irregularity of the distal tibial physis. However, on this radiograph there was no evidence of sclerosis, articular surface irregularity and/or collapse or associated fragmentation to suggest any.
radiographic evidence of osteonecrosis. An MRI was subsequently obtained which demonstrated irregularity, widening and increased signal within the distal tibial physis, compatible with SH Type I fracture. However, additional findings included serpiginous heterogeneous sign within the distal tibial metaphysic with surrounding bone marrow oedema suggestive of osteonecrosis/avascular necrosis (Figure-3).

Figure-1: widening and irregularity of the left distal tibial physis, suggestive of Salter Harris type I fracture. Overlying soft tissue swelling is identified. A comparison is made to the right distal tibial physis, which appears grossly unremarkable.

Figure-2: Left Ankle: Follow up radiograph 2 months later demonstrates interval increase in widening and irregularity of the distal tibial physis.

Figure-3: MRI Left Ankle: frontal and sagittal STIR images, sagittal T2 and axial T1 images were obtained.

There is irregularity, widening, and increased signal on STIR and T2 sequences within the distal tibial physis. These findings are compatible with Salter Harris Type I injury of the distal tibial physis. Additionally, serpiginous regions of signal abnormality are also visualized within the distal left tibial metaphysic with surrounding bone marrow oedema, compatible with osteonecrosis.

DISCUSSION
Paediatric fractures require appropriate care to ensure proper bone healing. The SH classification system aids in categorization of the fracture type and guides treatment options. The SH classification system describes fractures based on their anatomical localization within the bone. Fractures are subdivided into five categories. SH Type I fractures involve the physis (or growth plate). Type II fractures involve the physis and metaphysic, while Type III involve the physis and epiphysis of the bone. In Type I and II fractures, the germinal layer of the growth plate and the epiphyseal blood supply are spared, leading to more rapid recovery. Type III injuries tend to occur when the growth plate is partially closed, as in adolescents. Fractures of all three regions (physis, metaphysic, and epiphysis) are
classified as Type IV. Since Type III and Type IV fractures involve the germinal layer, there is potential for permanent damage. Type V injuries result from the crushing of the physe. Distal tibial SH type I fracture is the second most common type of physeal injury of long bones. Fractures of the physeal plate are typically concerning as they can lead to premature physeal closure, shortening, or angular deformity of the bone. Since joint capsule and ligamentous structures are stronger than the physis in the paediatric population, physeal fractures in children are the mechanical equivalent of ligamentous injuries in adults. Imaging characteristics of SH fractures can be evaluated with radiographs and/or MRI, with MR having a higher sensitivity and specificity. On radiographs, type I fractures demonstrate widening of the physe with or without displacement. However, they have low sensitivity and type I fractures may appear normal or show only mild soft tissue swelling. Type II fractures demonstrate a lucent fracture line extending through the metaphysis and into the physe with adjacent soft tissue swelling and joint effusion. Type III fractures demonstrate a lucent fracture line extending through the epiphysis and into the physe with soft tissue swelling and joint effusion. Type IV fractures are visualized as a lucent fracture line extending through the metaphysis, across the physe and into the epiphysis with accompanying soft tissue swelling and joint effusion. Type V SH fractures demonstrate narrowing of the physe. If post-treatment radiographs demonstrate persistent physeal widening >3 mm post reduction, then periosteal entrapment of the fracture is likely and treatment would require open reduction to remove the trapped periosteum.

For evaluation of type I fractures, protocol recommendations include obtaining opposite-side comparison radiographs. CT scans are used to evaluate anatomic extent and degree of displacement of complex SH injuries and are most commonly used with triplane fractures of the distal tibia (type IV). CT can also be beneficial to evaluate focal bony bridging across the physe during healing process, which is most commonly seen in type IV or V fractures. MRI findings of SH fractures include increased T2 signal within the physe for type I injuries. Other SH types demonstrate a linear fracture in the metaphysis/epiphysis that would appear as low T1 and T2 signal with accompanying marrow oedema. Chondral injury, soft tissue swelling, and joint effusion are other findings that can be identified on MR sequences. A pitfall on MR that is sometimes mistaken for an abnormality but is actually a benign finding is the periphyseal oedema zone. This demonstrates a characteristic bone marrow oedema pattern, which is centred around the central aspect of the closing physe in the knee joint. It is usually seen in adolescents and is thought to be related to early stages of physeal closure and could result in pain.

A rare complication of a distal tibial physeal fracture is post-traumatic osteonecrosis. Although the mechanism of post-traumatic osteonecrosis has not been established, there have been a few proposed theories in the current literature. Bone is a highly vascularized connective tissue and receives about 10–20% of the cardiac output. Blood vessel formation is important for osteogenesis, bone remodelling and bone regeneration. Upon damage, osteoblasts deliver nutrients and growth factors to damaged site. It has been shown that osteoblast precursors are localized along the vascular routes of bones, thus establishing a strong correlation of angiogenesis and osteogenesis. Avascular necrosis of the bone is characterized by ischemic injury, necrosis of osteocytes, degeneration of articular surfaces, and onset of osteoarthritis. Bone fracture or dislocation can initiate and contribute to these events. Normally, endothelial progenitor cells (EPCs) play an important role in bone formation and neovascularization. A study published by Feng et al. demonstrated osteonecrosis to be a consequence of diminished number of EPCs or inability of these progenitor cells to migrate. Disruption of vascular supply to the bone is also visualized upon damage to the endothelial cell membrane, which could be secondary to coagulation defects.

The most common anatomic locations of osteonecrosis include the femoral head, talus, and the humeral head. Our case manifests one of very few instances of osteonecrosis occurring at the distal tibial metaphysis of a patient sustaining a SH type I fracture. Diminished vascularization of the distal tibia has been reported previously and may contribute to the eventual development of avascular necrosis. In a study conducted by Menck et al., vascular flow to the lateral aspect of the distal tibia was supplied by only one branch of the tibialis anterior artery in one third of the cases. Lack of collateral vascularization, in combination with injury, is an alternative explanation for the formation of a bone infarct. Although decreased blood supply, as a result of the fracture, may have led to the bone infarct, distal tibial osteonecrosis has a rare incidence as compared to the talus and humerus. Unlike the tibia, the talus and humerus have more vulnerable vasculature, making them more prone to damage.

Based on the literature review, there has only been one other account of distal tibial Salter-Harris I fracture leading to osteonecrosis of the tibial metaphysis in the paediatric population. The injury occurred in a 10-year old boy who suffered a SH type I physeal fracture as a precursor to the onset of...
osteonecrosis. It was hypothesized that an associated proximal microfracture reduced the vascular supply of the metaphysis, which then resulted in osteonecrosis of the particular region. Despite the rare incidence of this complication in the distal tibia, post-traumatic osteonecrosis should be considered in the diagnostic work-up if the patient’s clinical symptoms worsen or progress.

MRI is more sensitive and specific than radiographs for evaluation of osteonecrosis. Radiographic findings of osteonecrosis include sclerosis, fragmentation and articular surface collapse. Particularly within the talus, osteonecrosis can be secondary to post traumatic aetiology, likely in the setting of a talar neck fracture located distal to the posterior facet. Initially, vascularized bone becomes osteopenic with devascularized bone appearing dense. As new bone formation and healing occurs, the devascularized fragment becomes dense. The Hawkins sign is demonstrated by visualizing a subchondral lucency of the talar dome and indicates intact blood flow to the talar body, and usually is associated with a smaller risk of osteonecrosis. This sign can be seen 6–8 weeks after talar neck fracture. Other non-traumatic causes of osteonecrosis of the talus can present as ill-defined sclerosis with serpiginous margins on the X-ray. Mueller Weiss Syndrome refers to osteonecrosis of the navicular bone in adults, whereas Kohler disease is osteochondrosis of the navicular bone in children. These are usually bilateral with radiographic changes occurring lateral in the initial phase of the disease or syndrome. However, with disease progression the lateral aspect of the navicular bone collapses and the medial aspect subluxes medially and dorsally.

MR findings of osteonecrosis include hypointense marrow signal on T1 sequences. However, the STIR (fluid sensitive sequences) will demonstrate bright marrow signal in the early phase and low marrow signal in the late phase. With regards to the talus, the classic appearance of osteonecrosis due to non-traumatic causes will be visualized with serpiginous low signal peripherally with central fat and a double line sign. A nuclear medicine bone scan for osteonecrosis shows focus of decreased uptake in the initial phase with increased uptake in later phase of the disease process. Steinberg et al. have developed criteria to quantify stages of avascular necrosis using the femoral head, one of the most common sites of incidence. Stage 0 refers to suspicion of osteonecrosis that cannot be confirmed by radiographs, bone scans, or MRI. In stage 1, radiographs are still normal, but bone scans and MRI show injury and death of osteocytes in the insulted area. Stage II reveals radiolucency on radiograph, indicating bone resorption, and sclerosis, which suggests new bone formation over dead trabeculae. A radiolucent “crescent sign” indicates Stage III of osteonecrosis, and progression into Stage IV is marked by a flattening of the articular surface, which is irreversible. Joint narrowing and degeneration follows in Stage V and VI. MRI ensures that osteonecrosis is identified and diagnosed at an earlier stage for positive intervention.

As suggested in the study by Pugley et al. delayed diagnosis and treatment of a physeal fracture may lead to inflammation and infarct of the tibia and thus clinical significance of early detection of SH type I fractures is crucial in prevention of osteonecrosis. Treatment for low SH category fractures includes casting, however, higher SH type fractures require open reduction and internal fixation. Treatment of osteonecrosis, prior to onset of fragmentation and collapse can include non-weight bearing management. However, once collapse and fragmentation occur, treatment may require surgical intervention. In our case, despite the initial correct and timely diagnosis of Salter-Harris type I fracture and the subsequent treatment, the injury progressed to osteonecrosis.

CONCLUSION

Osteonecrosis following fracture should be considered as part of the differential diagnosis in the context of worsening symptoms or symptoms resistant to treatment. Knowledge of the radiographic and MRI findings suggestive of osteonecrosis can help clinicians make the diagnosis with increasing regularly and lead to better care.

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