

Symptomatic Os Subtibiale Following Injury: A Case Report of Failed Conservative Treatment, Leading to Differential Diagnosis Made Intraoperatively

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Learning Point of the Article:

This case report demonstrates the difference between fracture of medial malleolus with an anteriorly located Os Subtibiale, which continues to present a learning challenge for most orthopaedic physicians as these cases are extremely rare and often asymptomatic

Abstract

Introduction: Os subtibiale is a rare accessory bone found adjacent to the distal tibia and is mostly asymptomatic. Differentiating it from a medial malleolar fracture is challenging. Most cases of acute ankle trauma that presents with an Os subtibiale are initially diagnosed as fractures by treating physicians at emergency centers. It is therefore critical for an orthopedic surgeon who gets referral for such cases to consider complete history of such patients to understand the cause of their symptoms. The goal is to avoid unnecessary surgical treatment and to realize that a symptomatic Os subtibiale can be successfully managed by conservative means. Nonetheless, certain cases may require surgical treatment if conservative treatment options fail. This is a case report of a patient that was diagnosed as a case of symptomatic Os subtibiale intraoperatively, after conservative options were unsuccessful.

Case Presentation: An 18-year-old patient with an injured right ankle was referred to our clinic after failed attempts to resolve his symptoms conservatively. Based on our initial clinical and radiographic examination, the patient was continued on a conservative treatment plan assuming, it was a case of unsuccessful fracture union. After almost a year of failed conservative treatments, surgery was offered to the patient, with the understanding that the bone fragment might either be fused or completely removed based on intraoperative diagnosis. At surgery, it was observed that the bone fragment had the characteristic of an Os subtibiale. It presented as a round, smooth structure with well-formed cortical boundaries, and minimal attachment to the distal tibia. A diagnosis of symptomatic Os subtibiale was made intraoperatively, which was then successfully excised using standard orthopedic instrumentation. The patient healed uneventfully and reported a pain free, normal ankle range of motion at latest follow-up of 18 months.

Conclusion: Accurate diagnosis of Os subtibiale continues to present a learning challenge for most orthopedic physicians as these cases are extremely rare and often asymptomatic. Our patient failed to respond to conservative treatment, which led to surgery and intraoperative diagnosis of Os subtibiale that was the pain causing agent.

Keywords: Os subtibiale, medial malleolus, symptomatic tibial accessory bone.

Introduction

The presence of accessory ossicles within the foot and ankle anatomy is known to be normal variants of bone development that is usually derived from the failure of union of secondary ossification centers adjacent to the main bony mass, and these are usually asymptomatic. In a recent review article by Keles-Celik et al. [1], they studied 24 unique foot and ankle accessory

ossicles reported in the published literature. In some cases, these accessory ossicles may be symptomatic enough to hinder normal activities of daily living. Most often, this can be attributed to fractures, dislocations, and irritation or impingement of adjacent soft tissues related to the accessory ossicle [1]. It is therefore important that we thoroughly understand the anatomy and clinical significance of accessory

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Figure 1: Plain frontal and sagittal radiographic images showing the possibility of an Os subtibiale. Note that in these images, it is difficult to ascertain the anterior or posterior location of this accessory bone.



Figure 2: Reconstructed 3-dimensional images of both right and left foot following a computed tomography scan. The right ankle displays an anteriorly placed bone fragment that appears to be centrally attached to the distal aspect of the tibia. It appears to have smooth and rounded exterior, with fully formed cortices.

ossicles to minimize the frequency of misdiagnosis, in an attempt to provide the best possible treatment to a symptomatic patient.

The Os subtibiale is one such an accessory ossicle found in anatomical relation to the distal tibial bone, that is, the medial malleolus. It is very rare, having an estimated prevalence of 0.7–1.2% in the general population [1]. Diagnosing it on plain ankle radiographs, especially following an episode of injury, is challenging and most often confused with medial malleolar fractures. This leads to administration of treatments that are

misguided, which results in incomplete or no resolution of symptoms. Researchers have reported that there may be clues to distinguishing an Os subtibiale from a medial malleolar fracture. First, although reported to be unilateral in some cases, it is mostly bilateral in occurrence. And second, it usually displays smooth and rounded edges compared to irregular appearance associated with a fractured medial malleolus.

In this article, we report a case of a teenager who presented with a symptomatic right ankle following a twisting injury that resulted from a sports activity.



Figure 3: Intraoperative image showing an anterior mid-line incision used to gain access to the distal tibial.



Figure 4: Intraoperative image showing a smooth and round shaped bone fragment attached to the anterior-distal aspect of the distal tibia.



Figure 5: Image showing the excised Os subtibiale along with a scale to gauge its size.

Case Presentation

An 18-year-old male patient presented with symptoms of pain and swelling localized on the medial aspect of his right ankle joint, specifically at the medial malleolus. The patient noted a recent history of a twisting injury of his right ankle 6-months before presentation at our specialty orthopedic clinic. The injury occurred while playing football. Initially, he was treated by his general practitioner, who assumed it to be a severe sprain injury and had placed him on a course of ice compress and anti-inflammatory medication followed by a below knee plaster cast for a period of 3 weeks. Following this treatment protocol, the patient was able to walk but reported on-going pain while walking once the plaster cast was removed. No radiographic imaging was done at the time and was therefore not available for review and comparison.

On further examination, swelling was observed over the distal aspect of the medial malleolus, along with tenderness over deltoid ligament of the right ankle. There was no bone related tenderness at the distal tibia. Ankle joint movement was also observed to be normal. We proceeded with a thorough radiograph examination including standard X-rays and 3-dimensional computed tomography (CT) scanning of the right ankle, which revealed the presence of a small bone fragment located anterior and distal to the medial malleolus (Figs. 1, 2). This fragment was not free floating and rather appeared to be attached to the distal tibia. Furthermore, it was a uni-lateral presentation as the patient's left ankle anatomy did not reveal the presence of a similar bone fragment and was rather normal in appearance (Fig. 2). Based on our findings as well as the findings of previously published reports on this topic, a benefit

of doubt was provided, and it was assumed to be a case of acute trauma, leading to a small fracture of the anterior aspect of the distal tibia.

At first consultation, the patient was explained of his diagnosis and offered a conservative treatment protocol that included a compression bandage and anti-inflammatory medication. Since this had minimal effect on his symptoms, he was placed in a plaster cast for period of 6 weeks thereafter. Following removal of the cast, the patient's symptoms re-occurred, and at that point, he had been on a conservative treatment regimen for almost 10 months with no significant clinical benefit. Based on this, it was considered that this might be a case of differential diagnosis, with the possibility of it being the case of a symptomatic Os subtibiale. At that stage, a surgery appeared to be the best option to help this patient with his symptoms. It was noted that intraoperatively, this surgery may involve either fixation of the bone fragment if it was deemed to be a fracture, or removal of the same if it appeared to be an Os subtibiale. The patient was then offered a surgical treatment option to which he consented and agreed to proceed as necessary.

On admission to the operating theater, the patient was positioned supine and administered general anesthesia. An upper thigh tourniquet was used on the patient's right leg in this case. To gain access to the distal tibia, a 4–5 cm anterior mid-line incision over right ankle joint was performed (Fig. 3). The surrounding soft-tissue structures, superficial nerve, and vascular structures were carefully protected and retracted to gain exposure to the distal tibia, which revealed the presence of a smooth and oblong bone fragment attached to the anterior-distal aspect of the medial malleolus, as was observed in the X-ray and CT imaging (Fig. 4). Based on its appearance, and the lack of bony fusion like characteristics with the adjacent distal tibia, this bone fragment was deemed to be an Os subtibiale. In addition, the margins of the distal tibia marked with a surgical marker, without considering the Os subtibiale, gave the appearance of a normal medial malleolus. It was also observed that it was pressing directly on the deltoid ligament, which explained the on-going pain that the patient experienced while walking. The deltoid ligament was released and the Os subtibiale was then surgically excised using a small 3 mm osteotome and a small bone rongeur. It was measured to be approximately $2 \times 11 \times \text{cm}^3$ in size (Fig. 5). Following excision, the surrounding bone and soft-tissue structures were carefully examined to ensure that there were no other anatomical bone or soft tissues abnormalities that could be contributing to the patient's symptoms. On confirmation, the surgical area was thoroughly irrigated with saline, and standard surgical procedures were used to close the incision in a layer by layer fashion. A standard surgical dressing was applied over the operated site, and the foot was placed in a heavy compressive

bandage for the next 2 weeks. This was followed by ankle mobilization and stretching exercises as tolerated by the patient. At his latest follow-up of 18 months, the patient reported to be pain free and displayed normal range of motion at the ankle joint compared to his non-operated, contra-lateral left foot. He also consented to participating in this study.

Discussion

Given the history of acknowledging the existence of Os subtibiale [2, 3], combined ironically with the publication of very few peer reviewed articles that are currently indexed in PubMed makes it important to discuss the recent findings on this subject matter. A PubMed search using the term “os subtibiale” resulted in only 20 hits (in October, 2020), of which only 14 were in English, with the oldest of these being published in 1986 by Coral [4], who reportedly misinterpreted an Os subtibiale for a fracture, and subsequently fused it using k-wires and tension band wiring. Although Coral’s [4] patient recovered well, he did not discuss in that article what he would have done to resolve the patient’s symptoms had he correctly diagnosed it as an Os subtibiale and not a fracture. He did state though that in his case, the fixation and immobilization were probably unnecessary. The choice between conservative treatment versus surgery to treat symptomatic Os subtibiale patients continues to be a dilemma for an orthopedic surgeon even today.

In contrast to Coral’s [4] finding of the Os subtibiale on the posterior aspect of the medial malleolus, it was located on the anterior aspect in our case but with similar size and shape characteristics. In a subsequent radiographic study, Coral [2] reported an incidence rate of 2.1% for anteriorly placed ossicles sized between 4 mm and 8 mm. He argued that only ossicles found in proximity to the posterior colliculus are true accessory bones, that is, the Os subtibiale, whereas those found in proximity to the anterior colliculus may result from an unfused ossification center or a post-traumatic ossification and are thus not classified as an Os subtibiale. The present author would like to argue this discrepancy in the definition and would like to counter that any ossicle found in the distal tibial, that is, subtibial, region, displaying the size, shape, and characteristics akin to that of an accessory bone should be classified as an Os Subtibiale, as observed in the present case report.

In case reports by Park et al. [5] and Turan et al. [6], they surgically treated a 28-year-old male and a 35-year-old female patient, respectively, suffering from posterior tibial tendon (PTT) dysfunction due to impingement caused by a posteriorly located Os subtibiale. The patient recovered following removal of the Os subtibiale and reported no PTT-related symptoms following treatment. In the case report by

Madhuri et al. [7], they reported a case similar to ours, albeit with a posterior Os subtibiale. Their 25-year-old male patient displayed a tender deltoid ligament and recovered following a short course of conservative treatment without the need for further intervention. However, they fail to mention any follow-up beyond 1 week. The case report by Bellapianta et al. [8] supports the present authors definition and classification of Os subtibiale as stated in the paragraph above. The patient in their case, a 20-year-old male soccer player, presented with bilateral, anteriorly located Os subtibiale and talocalcaneal coalition. He too was initially diagnosed as a case of non-healing fracture of medial malleolus. Following failure of conservative treatment, he was treated with arthroscopic debridement surgery, which helped relieve his symptoms. The authors noted that he may require further surgery; in case, his symptoms were to resurface.

In an interesting three patient case report by Kim et al. [9], they misdiagnosed two of three patients as having an old fracture, and tried to fuse the Os subtibiale to the medial malleolus. In one case, the bones did not fuse, but the patient was relieved of their symptoms. Learning from their first two cases, in the third patient that presented with similar clinical and radiographic findings, they decided to diagnose it as a case of Os subtibiale and excised it surgically. They noted that patient recovery was much easier and sooner in this patient, who was pain free at 13 months of follow-up and had returned to playing sports. Recently, Aydin [10] was successful in differentiating a secondary center of ossification from an Os subtibiale in his case report of a 9-year-old male patient who had suffered from acute trauma. He was treated conservatively and healed within 10 days with no need for additional treatment. Iliev et al. [11] recently reported a case similar to ours with an 18-year-old male patient who presented without injury but with symptoms of pain and swelling over his left medial malleolus. The patient did not respond to conservative treatment and was eventually treated with surgical excision of the Os subtibiale that resulted in complete resolution of symptoms at 8 months of follow-up. These authors also classify Os subtibiale into Type I and Type II based on the size, that is, Type I being sized less than 7 mm and Type II being sized greater than 7 mm. Per this classification, our patient falls under the category of Type II, which they report to be more likely to cause symptoms. Finally, Topal et al. [12] have also reported a three patient case report, in which they were able to accurately identify Os subtibiale in their patients who were referred to them by the emergency department as cases of medial malleolar fractures. In two cases, they were able to treat the patients conservatively, while the third patient also presented with a slightly proximal medial malleolar fracture in addition to the presence of the Os Subtibiale, who was treated with closed reduction and splinting. The patient healed uneventfully.



Conclusion

This case report demonstrates the difference between fracture of medial malleolus with an anteriorly located Os subtibiale, which continues to present a learning challenge for most orthopedic physicians as these cases are extremely rare and often asymptomatic. In our case, the patient failed to respond to conservative treatment for almost a year, which led to the thought of a possibility of a differential diagnosis that was confirmed intraoperatively following excision of the Os subtibiale accessory bone, which was the pain causing agent.

Clinical Message

Although not a major risk or health issue, Os subtibiale can and does impact a patient's ability to ambulate pain free, and both conservative and surgical treatment options should be discussed with the patients to provide them with the best possible solution for the symptom(s) they present with, which is mostly pain. Given the lack of knowledge available on this topic, it is important to realize that accurate diagnosis of a symptomatic Os subtibiale may only be possible through a detailed imaging study or an intraoperative examination. If surgery is deemed necessary, an attempt should be made to excise the Os subtibiale rather than attempting to fuse it to the distal tibia, as that can lead to more complications, requiring further intervention.

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