

CASE REPORTS

Iselin's Disease: A Case Presentation of Nonunion and Review of the Differential Diagnosis

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The confusing pathology of the proximal fifth metatarsal can make Iselin's disease difficult to distinguish from Jones', avulsion, or stress fractures, or os vesalianum. While Iselin's disease appears to be rare due to the scarcity of cases in the medical literature, this may be due to misdiagnosis. The case report discusses the course of treatment for a young male with Iselin's disease that developed into a nonunion. The differentiating factors of the above-mentioned fractures are discussed. (The Journal of Foot & Ankle Surgery 38(6):409–416, 1999)

Key words: apophysitis, avulsion, Iselin's disease, Jones' fracture, osteochondrosis, os vesalianum, stress fracture

Since Iselin first described a traction apophysitis of the proximal fifth metatarsal, only nine cases have been reported in the world literature (1–6). A single case of nonunion in a 20-year-old basketball player was reported by Canale and Williams (3). While Iselin's disease appears to be rare due to the scarcity of literature, it probably occurs more frequently. The complex pathology of the proximal fifth metatarsal may lead to the misdiagnosis of Iselin's disease. The more commonly treated disorders when dealing with this confusing area of bone pathology are Jones', avulsion, and stress fractures. The cases presented herein and review of the literature provide an opportunity to better understand the differential diagnosis of Iselin's disease.

Iselin's disease (ID) is one of the 11 osteochondrities, excluding the sesamoids, found in the foot (Fig. 1).

Osteochondrosis is a condition in which the primary or secondary ossification center in a formerly normal growing bone undergoes aseptic necrosis followed by gradual resorption of dead bone with regeneration and recalcification (7). The condition can be broken down into three main categories: articular, nonarticular, and physeal osteochondroses (8). The nonarticular category is further subcategorized into three etiologies: 1) the site of the tendon attachments, 2) the site of ligament attachments, and 3) the site of impact. ID is unique in that it falls under all three of these subcategories. The proximal fifth metatarsal is the site of three ligament attachments: the lateral band of the plantar fascia, and the plantar and dorsal fourth to fifth metatarsal ligaments. Two tendon attachments, the peroneus brevis and the peroneus tertius, insert in this area (Fig. 2) The tuberosity of the base of the fifth metatarsal is a prominence on the lateral aspect of the foot that is vulnerable to many types of impact.

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Case Presentation

A 17-year-old boy presented with localized pain of 3 months' duration on the lateral aspect of the left foot. The sharp pain worsened with activity, especially running and jumping. Some relief was felt during rest and the pain was

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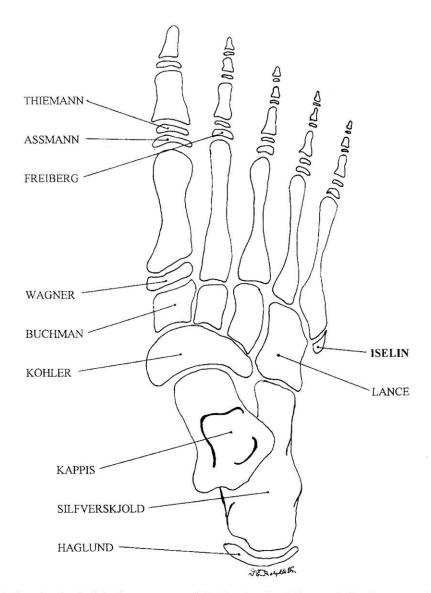


FIGURE 1 An illustration showing Iselin's disease as one of the 11 osteochondrities, excluding the sesamoids, found in the foot.

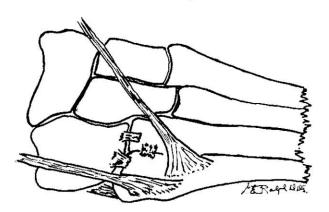


FIGURE 2 An illustration showing the proximal fifth metatarsal as the site of three ligament attachments — the lateral band of the plantar fascia, and the plantar and dorsal fourth to fifth metatarsal ligaments — and two tendon attachments — the peroneus brevis and peroneus tertius.

the worst when wearing dress shoes. The patient had no history of trauma or surgery to the affected foot.

On physical examination, a large prominence at the lateral aspect of the left foot appeared to be originating in the area of the cuboid and proximal fifth metatarsal (Fig. 3). The prominence was smooth, indurated, and nonmovable. There was no increase in skin temperature in the area. Some superficial hyperkeratotic tissue was noted on the lateral-plantar area and thought to be caused by friction pressure. Pinpoint tenderness was elicited with palpation and pain was experienced with resisted eversion. Muscle strength was decreased when testing the peroneal muscles most likely secondary to pain. All other strength and neurovascular exams were within normal limits.

Medial oblique, anterior-posterior, and lateral radiographs of the left foot were obtained. They showed bony development of a mature adult. Soft-tissue swelling was

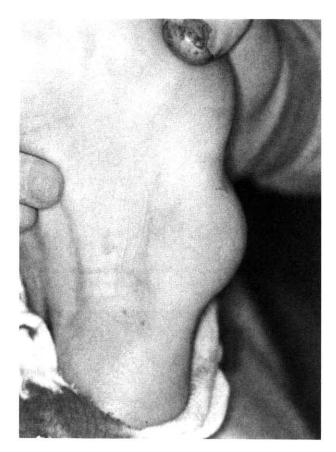


FIGURE 3 Large nonmovable, smooth, and indurated prominence on the plantar-lateral aspect of the left foot.



FIGURE 4 Preoperative radiographs of the left foot showing an oblique radiolucent line with fragmentation of the epiphysis secondary to a former traction apophysitis.

noted at the proximal fifth metatarsal with enlargement of the styloid process. An oblique radiolucent line with fragmentation of the epiphysis was also observed in the area in question. It was thought to be a fracture possibly secondary to a former traction apophysitis (Fig. 4).

The patient was treated conservatively by placing him in a postop shoe and instructing him to abstain from any running, jumping, or pursuing other aggravating activities. Following 5 months of various conservative treatments consisting of postop shoes, Cam walkers, nonweightbearing status, and decrease in activity, there was no long-term pain relief and the soft-tissue swelling did not resolve. Serial radiographs taken at intervals over 5 months showed nonunion of the apophyses and some widening of the radiolucent line was apparent. All attempts at treating the patient conservatively failed and NSAIDs provided only short-term pain relief. Unique to this patient, bone maturity had been reached and thus the apophyses could not fuse, causing the traditional methods to fail. The nonunion which developed secondary to ID added to the complexity of the case.

The patient elected to undergo surgical excision of the bony fragment at the site of nonunion. Under general anesthesia, a semi-curved incision was made over the deformity.

Intraoperatively it was determined that excision of the proximal portion of bone would be preferred to attempting fixation to the base of the fifth metatarsal. The location of the excised bone would not interfere with the function of the peroneus brevis pull and removal would not affect stability or gait. Dissection was carried to the insertion of the peroneus brevis. The peroneus brevis tendon insertion engulfed the entire fifth metatarsal base. Minimal

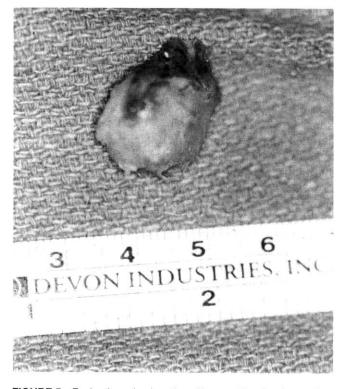


FIGURE 5 Excised proximal portion of bone at the site of nonunion.

disruption did not affect the tendon's function. The tendon was cut longitudinally and a small portion was freed from the bone. A synchondrosis was also noted and was easily separated with the use of a Freer elevator. A scalpel was used to release the fragment from the peroneus brevis tendon (Fig. 5). A small bursa sack was also excised from the affected area. The tendon was reapproximated, and the site was flushed and closed. Postoperative radiographs show total excision of the proximal bony fragment (Fig. 6).

The patient was placed in a postoperative shoe, and placed on nonweightbearing status for 2 weeks. Because only minimal disruption of the insertion of the peroneus brevis occurred during surgery, the patient was allowed partial weightbearing after 2 weeks of nonweightbearing status. At 4 weeks, the patient was then permitted total weightbearing status in normal shoe gear as tolerated. Eight weeks postoperatively, the patient was back to full activity with no restrictions. At 1 year follow-up, the patient is fully functional without limitations.

Discussion

In the past, overuse injuries in children were very rare. With the growth of organized sports in today's society, especially in younger age groups, overuse injuries are becoming more frequent. The traction apophyses, consisting of columns of growth cartilage that unite tendon with bone (peroneus brevis to the base of the fifth metatarsal), is a site of active growth in a child. Injuries at the site may result from one episode of macrotrauma or from repetitive microtrauma. The result of the trauma is pain, swelling, and only occasional bony and cartilaginous overgrowths referred to as apophysitis (9). In the growing child, the apophyseal growth cartilage is considered the weakest site for ligament and tendon attachment (10). Rapid growth periods in a child or adolescent can cause muscle-tendon tightness. The tightness is subject to repetitive overload, seen in sports training or competition, and allows that potential for tiny avulsion fractures to develop at the weak apophyseal growth cartilage. When this occurs at the bases of the fifth metatarsal, traction apophysitis or ID occurs.

ID should be considered when evaluating an adolescent or preadolescent who is active in sports. Boys are more likely candidates for ID because they have delayed maturation of the secondary growth center and are generally subject to increased trauma and stress through childhood (11). In ID, the secondary center of ossification at the base of the fifth metatarsal appears in girls at about age 10 and in boys at around age 11. Fusion occurs approximately 2 years later. Differentiating between Jones', avulsion, and stress fractures and ID is best done using radiographs in conjunction with the history and clinical



FIGURE 6 Postoperative radiographs showing total excision of the proximal bony fragment at the sight of nonunion.



FIGURE 7 A small fleck of bone that is oblique to the fifth metatarsal shaft and on the lateral aspect of the tuberosity is a variant found in most children. When correlated with clinical findings, ID should be considered.

symptoms. Patients with ID generally present with a tender prominence of the proximal fifth metatarsal which causes the most pain during activity and has varying degrees of relief at rest. A history of trauma to the area is usually absent. However, in some reported cases, an



FIGURE 8 Radiograph of a Jones' fracture transversing the junction of the diaphysis and metaphysis without extension distal to the fourth and fifth intrametatarsal articulation.

inversion injury was thought to be the beginning of the symptoms and thus cannot be ruled out as an initiating event (3). Any type of repetitive inversion of the forefoot will cause stress at the base of the fifth metatarsal and may lead to this condition. The foot will first become painful during running, especially in lateral maneuvers, and cutting and jumping motions. Examination will show an enlargement in the affected area that is painful on palpation. Resisted eversion produces discomfort, as do extremes of plantar flexion and inversion (2). Generally, there is pain over the insertion of the peroneus brevis tendon with very little or no erythema, edema, or ecchymosis. On radiographs, a normal variant will be noted in most children. The small fleck of bone is oblique to the fifth metatarsal shaft and on the lateral plantar aspect of the tuberosity (Fig. 7). This radiographic finding must correlate with clinical symptoms to make the diagnosis of ID. A medial oblique view will provide the best view (3). If radiographs are normal, but clinical signs suggest ID, a technetium bone scan will often show an increased uptake over the apophysis (2).

A patient with a Jones' fracture will present with an acute forefoot injury. A history of an inversion injury



FIGURE 9 Radiograph showing a stress fracture greater than 1.5 cm from the metaphysis-diaphysis junction showing evidence of bony callous.

that includes maximum loading over the lateral aspect of the foot is the most common mechanism of injury. This usually occurs while the ankle is plantarflexed and an adducted force is applied to the forefoot. Examination will reveal edema of the lateral aspect of the foot that is usually localized to the base of the fifth metatarsal. The degree of ecchymosis will depend on the time between when the injury occurred and the treatment was rendered. Radiographically, a Jones' fracture is transverse at the junction of the diaphysis and metaphysis without extension distal to the fourth and fifth intrametatarsal articulation (Fig. 8) (12, 13). Anatomically, a Jones' fracture corresponds to the area intermediate to the insertion of the peroneus brevis and tertius tendons (14).

A stress fracture of the proximal fifth metatarsal is a diaphysial pathological fracture located at the proximal 1.5 cm of the shaft (15). It is often misdiagnosed as a Jones' fracture. Stress fractures are the end result of repetitive cyclic forces. They begin with microfractures and become complete fractures if reparative processes cannot keep up with continued cyclic forces (Fig. 9). A stress

fracture can present without an acute traumatic episode and have an insidious onset of symptoms. Physical and radiological presentations of stress fractures differ greatly from those who have ID. Stress fractures do not have a large prominence at the lateral base of the fifth metatarsal. On radiography, the stress fracture is more distal and in the transverse plane.

The avulsion fracture occurs when the proximal portion of the fifth metatarsal is fractured off secondary to a violently contracting peroneus brevis muscle during sudden inversion of the foot (16). In a study done by Richli in 1984 using cadaveric specimens, it was shown that the lateral band of the plantar aponeurosis is a more likely structure causing tuberosity avulsion fractures (17). The avulsion fracture is very common with lateral ankle sprains. Radiographically, the fracture line appears in the transverse plane, sometimes it can be slightly oblique (Fig. 10). This fracture most mimics ID radiographically and thus is sometimes misinterpreted.

In addition to the possible fractures, os vesaleanum, an accessory bone found proximal to the base of the fifth metatarsal, must be distinguished from ID (Fig. 11). This

bone is found within the peroneus brevis tendon and considered asymptomatic in the majority of patients. Radiographic differentiation may be difficult because it can appear in many different shapes (Fig. 12). Therefore, history and clinical presentation play an essential role in ruling it out.

The treatment of ID ranges from conservative management to surgical intervention. As with any overuse injury that leads to traction apophysitis, the initial treatment involves eliminating the causative forces, thus allowing the inflammation to decrease in milder cases. Ice and low dye strapping may also aid in decreasing symptoms. If tenderness and inflammation do not resolve, oral nonsteroidal anti-inflammatories (NSAIDs) can be added to the treatment plan. Patients must be cautioned that limited use of NSAIDs is recommended and they cannot consider the medication the only form of relief. If the NSAID is discontinued and symptoms return, a different course of treatment should be employed. In more moderate cases when discomfort returns, immobilization is recommended. This may be done by using an air cast, a walking cast, or possibly a below-knee cast and crutches to achieve nonweightbearing status. When the tenderness is completely resolved, physical therapy



FIGURE 10 Radiograph showing an example of an avulsion fracture of the base of the fifth metatarsal.



FIGURE 11 Radiographic example of an os vesaleanum, an accessory bone found proximal to the base of the fifth metatarsal.

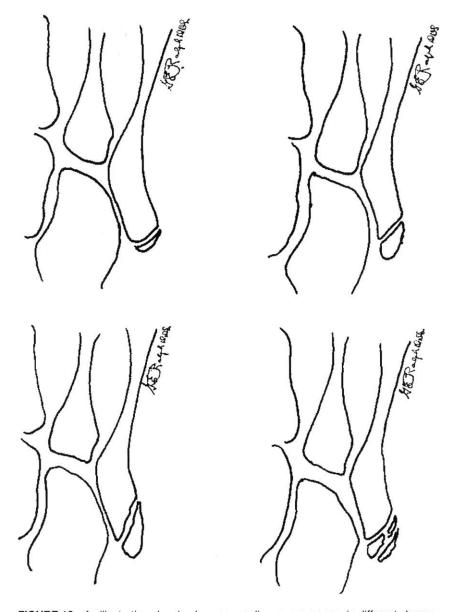


FIGURE 12 An illustration showing how os vesalianum can appear in different shapes.

consisting of strength, coordination, and range-of-motion exercises should be initiated.

If conservative treatment fails, ID can develop into a nonunion and surgical intervention may be indicated. The area of pathology must be treated surgically either by resection or fixation at the nonunion site. Excision of the proximal bony fragment is recommended as long as the excised bone does not interfere with the function of the peroneus brevis tendon or stability of gait.

Conclusion

A traction apophysitis is most commonly seen in young athletic males. It is usually self-limiting, with relief of symptoms occurring with decrease in activity or, in more severe cases, immobilization. It is often misdiagnosed as a fracture. If bony maturity is reached and symptoms continue, it can develop into a nonunion. When all conservative treatments fail, a more aggressive approach must be taken to treat the nonunion. In our case, this meant surgical excision of the proximal bony fragment.

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