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CASE REPORTS

Freiberg's Infraction in Identical Twins: A Case Report

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Freiberg's infraction is an ostechondrosis of a lesser metatarsal head resulting in degeneration of the metatarsophalangeal joint. Several mechanisms have been suggested in its pathenogenesis. Freiberg first described the entity and believed single impact trauma was the underlying cause. Repetitive biomechanical microtrauma is the most widely accepted etiologic theory. Other factors contributing to its development include aseptic necrosis, ischemia, and a congenital predisposition. We present a case report of Freiberg's infraction occurring in identical twins involving multiple metatarsals in various stages of degeneration. One of the twins was affected unilaterally whereas the other twin was affected bilaterally. Both twins had involvement of the second metatarsal on the same side extremity. The occurrence of Freiberg's infraction in identical twins suggests that an underlying congenital predisposition to the condition may play more of a role than previously considered. (The Journal of Foot & Ankle Surgery 44(3):218-221, 2005)

Key words: Freiberg's infraction, osteochondrosis, identical twins

Osteochondrosis of the second metatarsal head was described by Freiberg in 1914 (1). It occurs predominantly in teenaged females during skeletal growth. It is radiographically characterized by flattening of the metatarsal head and eventually progresses into arthrosis of the metatarsophalangeal joint (MTPJ). There are several theories surrounding the pathogenesis of the lesser metatarsal osteochondrosis. Chronic repetitive microtrauma is the most widely accepted theory (2–5). Freiberg believed that the condition results from a single traumatic event (1). However, Kohler did not accept that trauma alone would cause osteonecrosis of the metatarsal head (6). Freiberg later acknowledged that trauma was not the only underlying contributing factor (7). Other authors have attributed this condition to aseptic necrosis of the metatarsal head due to vascular insufficiency (8–13). However, there is no data suggesting a genetic predisposition for the development of Freiberg's infraction.

Several investigators have created classification schemes based on the radiographic features in an attempt to categorize the natural progression of the syndrome (14–17). Smillie's classification is the most recognized and involves 5 stages of degeneration (17). Stage I represents an epiphyseal fracture that is typically not visualized on radiographs. Joint space widening may occur, but is a nonspecific finding. With stage II, the metatarsal head begins to flatten as the dorsal aspect of the joint continues to deteriorate. This characteristic feature of Freiberg's infraction is easily identifiable on radiographs. Structural compromise of the metatarsal head is the hallmark of stage III and results in central joint depression from subchondral bone collapse. The medial and lateral aspect of the metatarsal head remains intact and is visualized as medial and lateral osseous projections. Importantly, only the dorsal aspect of the joint is involved while the plantar aspect remains intact and unaffected. In stage IV, loose bodies are seen about the periphery of the joint; these represent fracturing of the medial and lateral projections. Lastly, stage V is complete degeneration (arthrosis) of the MTPJ and represents the end point of the condition.

We present a case report of Freiberg's infraction occurring in identical twins with various stages of degeneration.

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Although a report on osteochondrosis of the navicular in identical twins may suggest a possible genetic cause for other osteochondrosis in the foot, a congenital association with Freiberg's infraction has yet to be determined and requires further investigation (18). To the author's knowledge, this is the only reported case of Freiberg's infraction occurring in identical twins.

Case Reports

Patient 1

A 23-year-old female competitive amateur marathon runner presented with an 8-year history of right second MTPJ pain that occurred 2 to 3 times per year. There was no history of trauma. Pain was worse with physical activity and caused her to alter her gait. She identified a family history of forefoot pain as her identical twin sister also had similar complaints.

Physical examination revealed a painful second MTPJ with limited range of motion of approximately 30°. A dorsal osteophyte was palpable. There was a mild hallux valgus deformity and a hypermobile medial column. An equinus deformity was present as well. Evaluation of the contralateral foot did not reveal any lesser MTPJ abnormalities, although equinus, hallux valgus, and a hypermobile medial column were present.

Radiographic evaluation of the symptomatic (right) foot revealed flattening and degeneration of the second MTPJ with loose body formation (Fig 1). The asymptomatic foot (left) showed flattening of the third metatarsal head and mild sclerosis within the epiphyseal area.

Patient 2

After Freiberg's infraction was identified in patient 1, her symptomatic identical twin sister was examined. She was also a high-level runner and did not recall a specific traumatic event. Physical examination of the right foot demonstrated a palpable exostosis at the dorsal second MTPJ. Range of motion of the second and third MTPJ was limited; and without pain to palpation or motion. Both extremities had an equinus deformity associated with a hypermobile medial column and mild bunion. Radiographs revealed degenerative Freiberg's infraction of the second and third metatarsal heads (Figs 2 and 3). The second metatarsal head demonstrated dorsal central collapse with an intact lateral projection (stage III). The third metatarsal head demonstrated central collapse with loose bodies (stage IV).

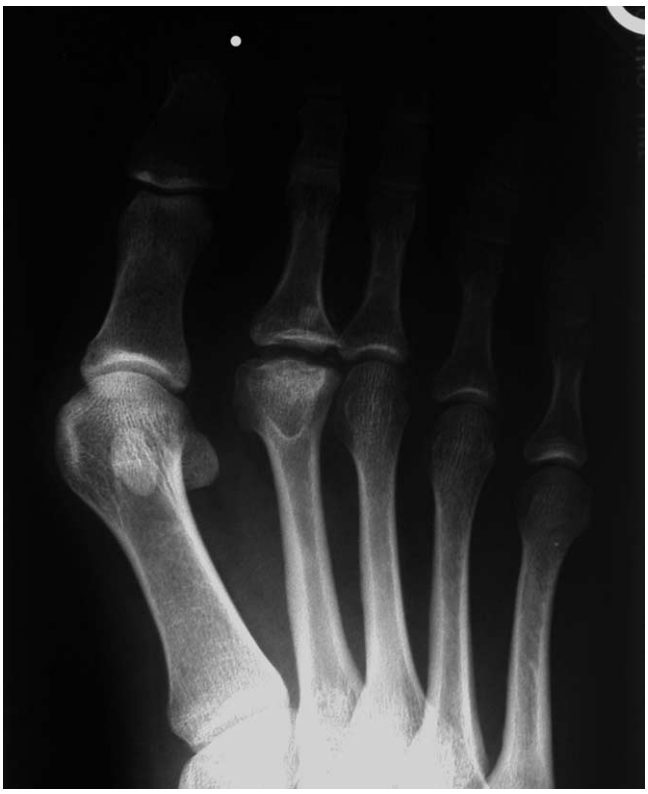

FIGURE 1 Patient 1, right foot. Anteriorposterior radiograph demonstrating degenerative arthrosis of the second MTPJ (stage V). Note the decreased joint space, loose bodies, and phalangeal base deterioration.

Discussion

Freiberg's infraction is a disturbance of skeletal growth (osteogenesis and chondrogenesis) of a lesser metatarsal head and the most common osteochondrosis of the foot (19). It is classified as a primary articular osteochondrosis and involves the secondary ossification center (20). Unlike other osteochondroses, there are strong female predilections; women are 5 to 11 times more likely to develop the condition (14, 15, 19, 21). The second metatarsal is most commonly affected (68% of cases) (21). Any lesser metatarsal is susceptible and multiple metatarsals may be concomitantly involved. Bilateral involvement occurs in 7% of cases (15).

It is thought that the stress of weight-bearing is a major contributor to the increased incidence of osteochondroses of the foot when compared to that of the upper extremity and hand (6). The second and third metatarsals are susceptible to repetitive overload, as seen with a Morton's foot type, including gastrocnemius equinus, hypermobile medial column, short first ray, and/or a long second metatarsal (22, 23). Hansen suggests that gastrocnemius ankle equinus is a pathological driving force for metatarsal overload (24). The tight heel cord produces a strong plantar-flexing force throughout gait. The tibialis anterior and the recruited long

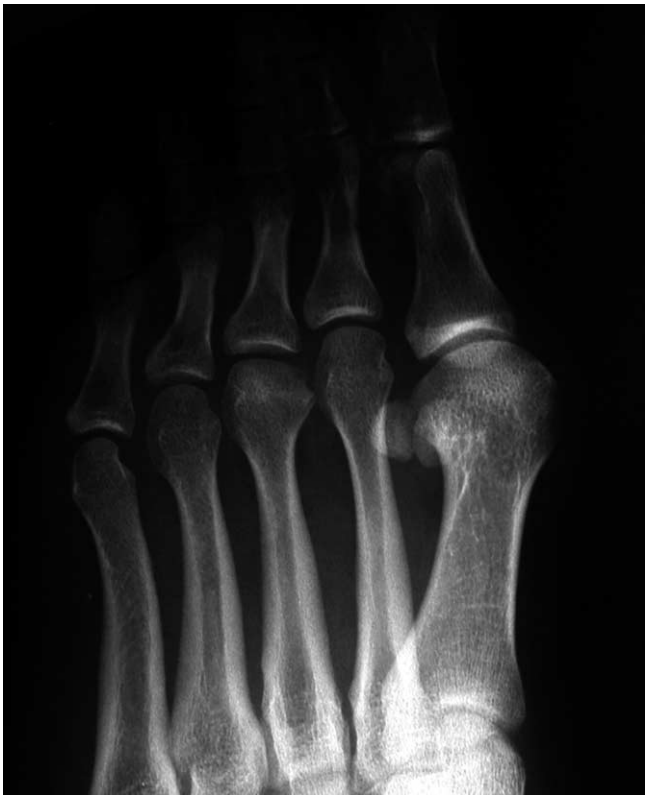

FIGURE 2 Patient 1, left foot. Oblique radiograph demonstrating flattening of the third metatarsal head (stage II).

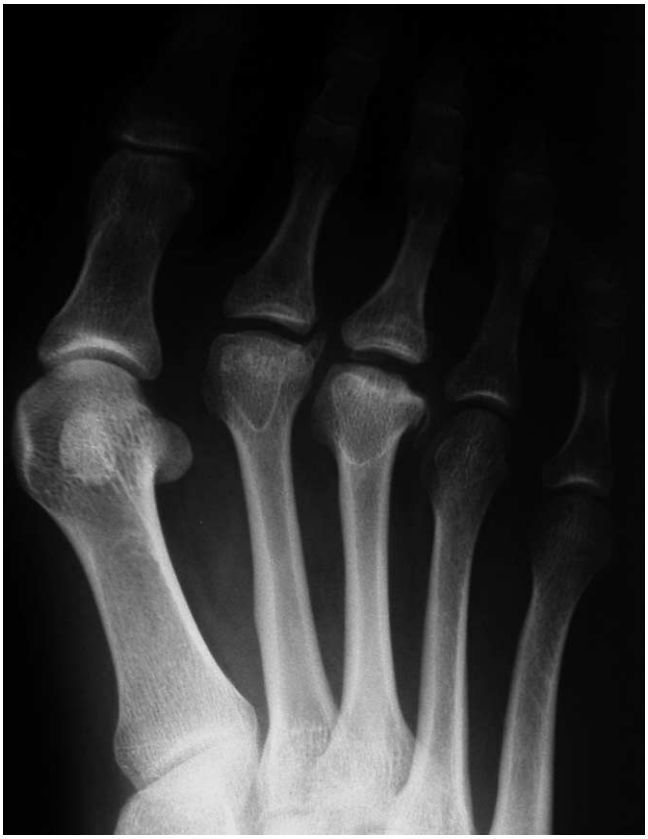

FIGURE 3 Patient 2, right foot. Anteriorposterior radiograph demonstrating multiple metatarsal involvement. The second metatarsal head with dorsal central collapse and an intact lateral projection (stage III). The third metatarsal with central collapse and loose body formation from fracturing of the medial and lateral projections (stage IV).

toe extensors cannot overcome the power of the gastrocsoleal complex. The lumbricals are also overpowered and the overall resultant extensor recruitment gradually produces a claw toe deformity. Eventually, digital contracture exerts retrograde forces on the metatarsal head and is exacerbated by the anteriorly displaced fat pad. Furthermore, insufficiency of the first ray transfers an increased load onto the lesser metatarsals during gait. Overload of the second and third MTPJs is thought to play a role in plantar plate degeneration or rupture, hammer toes, and stress fractures (25–27). Moreover, high heel shoes worn by females may contribute to an increased forefoot pressure and create a situation of dorsal impingement of the base of the proximal phalanx on the metatarsal head (28). All of these biomechanical factors may promote the occurrence of a Freiberg's infraction.

Because Freiberg's disease occurs during skeletal growth, it manifests during the teenage years. Its clinical presentation is variable and poorly correlated with the underlying radiographic features, especially in the early stages of the condition. The incidence of Freiberg's infraction is difficult to ascertain because many patients remain symptom free and do not seek medical attention. Often degeneration of the metatarsal head is discovered during adulthood as an incidental radiographic finding. Some have discounted classifying MTPJ arthrosis in adults as true Freiberg's infraction (19). The only differences may be the etiology and initiating factors causing the joint degeneration, but the treatment remains identical.

Avascular necrosis or vascular insult has been a popular theory for the development of osteochondroses. Validot and Validot maintained that a mechanical arterial spasm that occurs at the joint capsule ultimately compromises the blood supply to the metatarsal epiphysis (8). There is some cadaveric evidence to suggest that a variable arterial network of the second metatarsal head may contribute to Freiberg's infraction. The metatarsal heads are supplied by both the dorsal metatarsal arteries and the plantar metatarsal arteries, originating from the dorsalis pedis and posterior tibial artery, respectively (9). The second metatarsal neck, in particular, is supplied by an anastomotic branch of the first dorsal metatarsal artery (10). Adachi identified a very thin dorsalis pedis artery in 3% of 230 feet (11). Moreover, Huber reported an absent dorsalis pedis artery in 12% of 200 cadaveric feet (12). In another study of 6 cadavers, 2

specimens had an absent second metatarsal artery (13). It is possible that some individuals with Freiberg's infraction may have a vascular anomaly and/or a relative lack of blood supply to the second metatarsal head. Such an association has yet to be identified, probably because arteriographic studies are not practical for this small anatomic area.

Concomitant medial conditions have been reported to occur in association with osteochondroses. These include hypothyroidism, Gaucher's disease, slipped capital femoral epiphysis, renal osteitis, cystic fibrosis, lipidosis, and gout (19, 29–34).

It is not clear why the identical twins in this case presentation demonstrated metatarsals in various stages of degeneration (stage II through stage V) and a different pattern of metatarsal involvement. Patient 1 has bilateral involvement whereas Patient 2 has unilateral multiple metatarsal involvement. The simple presence of Freiberg's infraction in identical twins suggests that there may be a genetic propensity for the condition, especially because both twins have involvement of the second metatarsal of the same-side extremity. The physical demands of marathon running (biomechanical microtrauma) are also suggested to play a role in the development of Freiberg's in these twins and may account for the overall clinical variability. Although 1 case report of Freiberg's infraction in identical twins does not indicate that genetic factors alone are responsible for the condition; it does suggest that genetic factors may be more responsible than previously considered. Further studies exploring the inheritance patterns are needed to clarify the presence of an inherited propensity for the condition.

Conclusion

The exact etiology of Freiberg's infraction remains unclear and is likely a multifactorial process. Congenital factors have been considered, but the genetic contribution and pattern of inheritance remains to be established. This case report suggests that an unknown genetic predisposition may be associated with the development of a Freiberg's infraction.

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