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Dysplasia Epiphysealis Hemimelica of Talus Mimicking Posterior Ankle Impingement Syndrome in a Young Male: A Case Report with Review of the Literature

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A R T I C L E I N F O

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A B S T R A C T

Dysplasia epiphysealis hemimelica is a rare skeletal developmental disorder representing an osteocartilaginous tumor arising from 1 or more epiphyses during childhood. We report a case of a young male who presented with complaints of pain and swelling in the posteromedial aspect of the left ankle with the clinical diagnosis of posterior impingement syndrome. Imaging studies, including radiography, computed tomography, and magnetic resonance imaging, suggested the possibility of localized juxta-articular dysplasia epiphysealis hemimelica arising from the talus. The patient was successfully treated by surgical excision. The postoperative histologic findings were consistent with osteochondroma. This case report emphasizes the possibility of dysplasia epiphysealis hemimelica in the differential diagnosis of impingement syndromes around the ankle. The aim of the present study was to familiarize clinicians with this rare entity, because the number of documented cases has been increasing.

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Posterior ankle impingement syndrome is a clinical disorder characterized by posterior ankle pain that is caused or exacerbated by forced plantar flexion. The most common cause of this syndrome is the pathologic features of the os trigonum-talar process. Dysplasia epiphysealis hemimelica (DEH) is a rare, nonhereditary, developmental, skeletal disorder affecting the epiphyses during childhood. The reported incidence is 1 in 1 million people, with fewer than 50 cases involving the talus having been reported to date. DEH can mimic posterior ankle impingement syndrome by producing symptoms of joint impingement. We report the unusual presentation of an uncommon disease (i.e., DEH) that is not widely known by clinicians.

Case Report

An 18-year-old male patient presented to the orthopedic outpatient department with complaints of pain and swelling in the

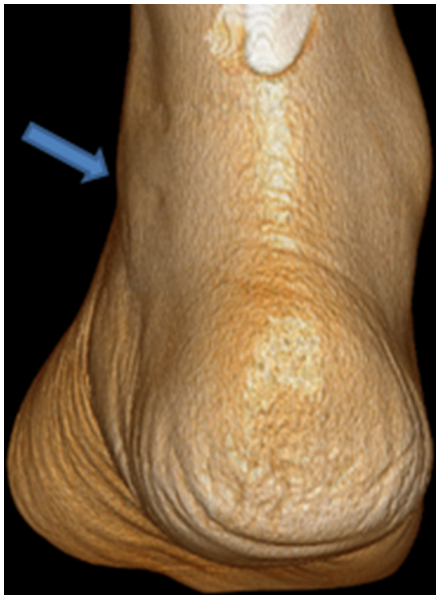

Fig. 1. Virtual image of right ankle showing ill-defined swelling in posteromedial aspect (*arrow*). The overlying skin is normal.

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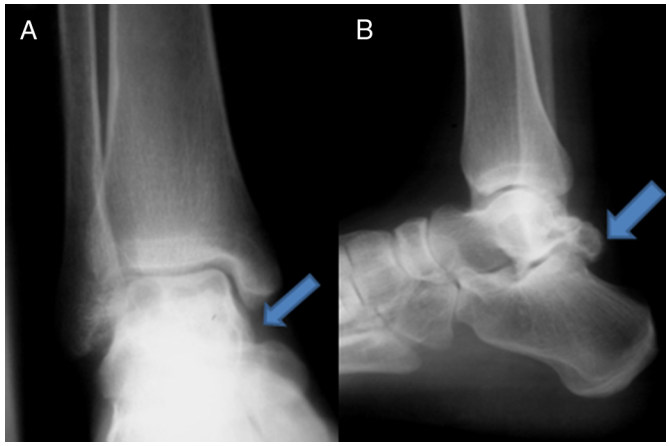

Fig. 2. Radiographs of ankle in anteroposterior (*A*) and lateral (*B*) projections showing an exostosis arising from posteromedial aspect of talus (*arrows*).

posteromedial aspect of his left ankle for a duration of 1 year. The pain was exacerbated by sports activity. He had no history of fever, significant trauma, or symptoms related to the other joints.

The physical examination revealed the presence of an ill-defined, hard swelling over the posteromedial aspect of the right ankle (Fig. 1). Mild tenderness was present, with restricted movement, both plantar flexion and dorsiflexion, compared with the contralateral side. The plantar flexion was more limited and appeared secondary to a mechanical block. Exacerbation of the pain occurred with forced plantar flexion. The overlying skin was normal, with no evidence of any signs of inflammation or sinus. No signs were found of muscle wasting or limb length discrepancy. The routine laboratory examination findings were unremarkable.

Posterior ankle impingement syndrome was diagnosed clinically, and the patient was referred to the radiology department for evaluation. Radiographs of the right ankle in the anteroposterior and lateral projections demonstrated the presence of an osseous outgrowth from the posteromedial aspect of the talus (Fig. 2). On further enquiry, the patient provided a history of a vague swelling since childhood that had gradually increased in size, most significantly in the past 1 year. Initially, the swelling was not associated with pain, but it had been painful for the previous 1 year.

The patient was also evaluated using noncontrast-enhanced computed tomography (CT) and magnetic resonance imaging (MRI) of the ankle. CT showed an osseous outgrowth at the posterior and medial aspect of talus that was juxta-articular in location, with areas of ring and arc-like calcification seen within the lesion (Fig. 3). MRI demonstrated a heterogeneous signal intensity mass protruding from the posteromedial aspect of the talus, with an overlying cartilaginous cap, measuring approximately 2 mm (Fig. 4). Associated edema was present in the adjoining soft tissue.

The clinical presentation of the patient and characteristic imaging findings established the diagnosis of DEH of the talus. Because the patient had presented with pain and impingement, surgical treatment was considered. An arthroscopic approach was not preferred because of the large size of the lesion. Through a posteromedial approach, the tibiotalar joint capsule was opened. The abnormal outgrowth was excised, and an attempt was made to maintain joint congruity. An immediate and significant improvement was seen in plantar flexion, demonstrated perioperatively.

The postoperative histologic examination revealed osteochondroma, confirming the diagnosis. A follow-up examination performed 9 months after surgery revealed the absence of pain and swelling with an improved range of motion (Fig. 5).

Discussion

DEH is an uncommon skeletal developmental disorder representing an osteochondroma arising from an epiphysis. The reported incidence of this entity has been 1 in 1,000,000 (1). The etiology of DEH is unknown, and no definite evidence of genetic inheritance has been found. Historically, it has been referred to by many names by different investigators. It was first described as *tarsomegalie* in 1926 by Mouchet and Belot (2). Trevor (3) used the term *tarso-epiphyseal aclasis* in 1950; thus, this abnormality is commonly referred to as Trevor's disease. Finally, in 1956, the term *dysplasia epiphysealis hemimelica* was coined by Fairbank (4), who reported 14 patients with DEH. The word *hemimelica* is derived from 2 Greek words, *hemi* (half) and *melos* (limb). According to Fairbank (4), DEH will be confined to the medial or lateral half of an epiphysis of a single limb. Approximately 250 cases of DEH have been reported in medical studies since 1957 (5).

The age of diagnosis of DEH is 2 to 14 years, and it is uncommonly seen in adults (6). It is more commonly seen in males and is 3 times more common than in females (7). Most cases of DEH are asymptomatic, but DEH can cause mechanical symptoms depending on the lesion size and location. The disease is usually recognized at a young age because of the antalgic gait, palpable mass, varus or valgus deformity, or limb length discrepancy (8). These signs are produced by alteration of the epiphysis, causing malalignment of the bone and joint. The natural history of the disease is a continuous increase in size until skeletal maturity has been achieved. Surveillance is performed to assess the progression of the disease, not to evaluate for malignant degeneration, which has not been reported.

The disease predominantly affects the lower limb, with upper limb involvement, involving the humerus, ulna, and scapula, seen rarely

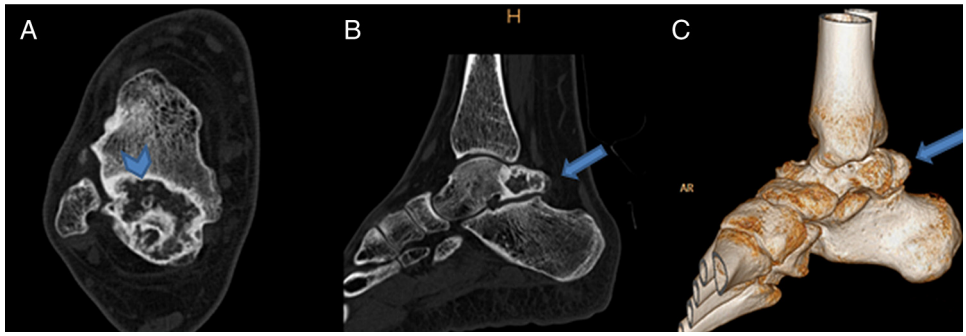

Fig. 3. Axial (*A*), sagittal (*B*), and volume-rendered (*C*) noncontrast-enhanced computed tomography scans of right ankle (bone window) showing osseous outgrowth (*arrows* in *B* and *C*) arising from the posterior and medial aspect of the talus. Ring and arc-like calcifications can be seen within the lesion (*arrowhead* in *A*).

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