A Novel Technique for the Treatment of a Rare Atypical Osteoid Osteoma of the Talus

Anson Chu, DPM, AACFAS, Stephen A. Brigido, DPM, FACFAS

CS-942

STATEMENT OF PURPOSE

We herein present a case of delayed diagnosis of an atypical talar osteoid osteoma involving an under reported location of the talar body. In addition, we utilize a novel technique of autologous bone grafting mixed with an Acellular Connective Tissue Matrix (ACTM) as a viable treatment option.

LITERATURE REVIEW

Osteoid osteoma is a small benign, well circumscribed tumor of bone that was first described by Jaffe in 1935. (1) It often occurs in the diaphysis of long bones, such as the femur or tibia and represents approximately 10% of all benign bone tumors. (2) It rarely involves juxta-articular surfaces. The tumor is three times more common in males than females, and presents between the ages of 5 and 30. The lesion is often less than two centimeters in diameter. (3) The most common symptom is localized, intermittent pain that worsens nocturnally and is often relieved with non-steroidal anti-inflammatory, especially salicylates. The central nidus has a very active osteoblastic center which leads to high prostaglandin and cycloxygenase-2 activity. Alleviation of pain with NSAIDs is due to the inhibition of prostaglandin synthesis and is nearly diagnostic for osteoid osteoma. (2)

A classic lesion presents with round, lytic, radiolucent nidus surrounded by sclerotic bone in meta-diaphysis. This nidus can be further visualized utilizing through advanced imaging. A computerized tomography is gold standard for identification, location, and size. An MRI is more specific; however, it can be misleading as images can mimic more aggressive lesions. An osteoid osteoma can be identified on bone scan with the “double density sign” characterized by intense focal uptake at the nidus surrounded by area of much less intensity peripherally. An atypical osteoid osteoma is defined by a lesion having radiographic or clinical features other than that of typical lesion. (4)

Incidents of osteoid osteoma in the talus is rare and involves the talar neck more than 90% of the time (5). These tumors are a rare cause for ankle pain and may be misdiagnosed particularly when presented in the setting of concurrent trauma. Treatment options have included conservative care in smaller lesions that are self- limiting, to surgical excision through arthroscopic or open techniques. In larger lesions involving the body and articular surface of the talus, bone grafting has been described to back-fill the void left after surgical excision (6,7). Bone grafting can consist of autogenous, allograft, or bone graft substitutes. Biological augmentation can help facilitate both graft incorporation and healing potential (8).

Recently, the biocompatibility of Acellular Connective Tissue Matrix (ACTM) with human osteoblasts was evaluated. The adhesion, proliferation and osteogenic activity of osteoblasts on ACTM were compared with negative control ultra-low adhesive polystyrene surface or conventional cell culture substrate of tissue culture treated polystyrene. The bench top data demonstrated the following: (1) The presence of CTM particulates is required for the adhesion of human osteoblast (HO) cells. (2) ACTM particulates not only support the cell adhesion but also support the proliferation of HO cells. (3) In comparison with the conventional cell culture surface, HO cells on ACTM-coated surface maintained a better osteogenic activity (9).

CASE STUDY

A 21-year-old Spanish speaking Hispanic male with an unremarkable past medical history use was referred to our clinic for surgical evaluation after failed conservative therapy. Patient reported he initially noted the pain approximately four and half years while living in Puerto Rico and was subsequently placed in a short leg cast with minimal improvement. Patient presented with left medial ankle with insidious onset that waned, unrelieved with rest and ice. Patient denied history of trauma. Upon physical examination, the patient had pain on palpation of the medial gutter and pain noted with active dorsiflexion of the left ankle with anteromedial edema. His plain radiographs were unremarkable and no lesions were identified. All lab work including antinuclear antibody, comprehensive metabolic panel, complete blood count, c-reactive protein, sedimentation rate, rheumatoid factor, Lyme disease antibody, and HLA-B27 were all within normal limits.

A magnetic resonance imaging (MRI) without contrast was completed and demonstrated a bone lesion in the medial aspect of the talar body measuring 1.1 cm x 1.5 cm x 1.5 cm cephalocaudad. The anterior, posterior, and lateral margins were sclerotic and the medial border breached the medial cortex with its articulation to the medial malleolus. (Fig 1) The radiologist recommended another MRI with gadolinium contrast to further evaluate the bone lesion. Postcontrast imaging 3 weeks later revealed minimal enhancement in the periphery of the lesion and surrounding associated structures. This lack of enhancement favored a benign lesion with low possibility for malignancy, however it could not be completely excluded.

Due to the unconfirmed diagnosis with advance imaging, the patient was consented for staged excision and curettage of the lesion. The patient agreed to return for second operation for autogenous bone grafting that would also be augmented with ACTM. The patient initially underwent a medial malleolar osteotomy with excision and curettage of the boney lesion to the medial talar body. The specimen was excised in toto with margins and sent to pathology for gross examination. The osteotomy was fixated with two 4.0mm cannulated screws. Pathology results were consistent with atypical presentation of an osteoid osteoma of the talus with clean margins. The patient was subsequently consented for bone grafting and repair of the talar lesion.

Two weeks following the initial excision and curettage, the patient was brought back to the operating room. An autogenous graft was harvested from the calcaneal body using a powered trephine and was then mixed with ACTM. The prior medial malleolar osteotomy was taken down by removing the 4.0mm screws and the bone lesion site in the talus was identified. The lesion was again curetted to healthy bleeding cancellous bone base. The autogenous bone and ACTM graft were packed into the talus to backfill the lesion to the level of the cartilage. The medial malleolar osteotomy was again fixated with 4.0mm screws and a five-hole medial malleolar locking plate (Fig 2). The patient was placed into a posterior splint with instructions to remain strictly non-weight bearing. Follow up imaging with computed tomography at four months demonstrated incorporation of the graft without evidence of boney lesions (Fig 3). The patient returned to the clinic for a twelve month follow up and remains asymptomatic.

FIGURES

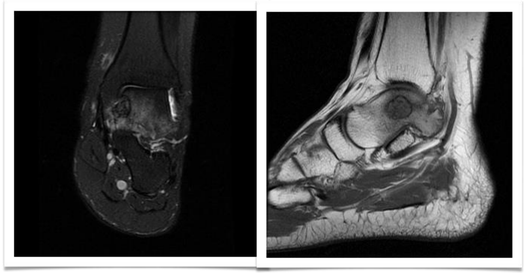

FIGURE 1

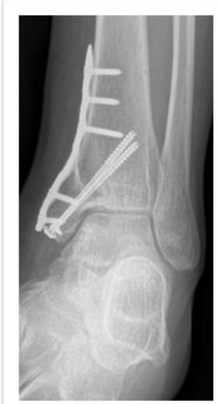

FIGURE 2

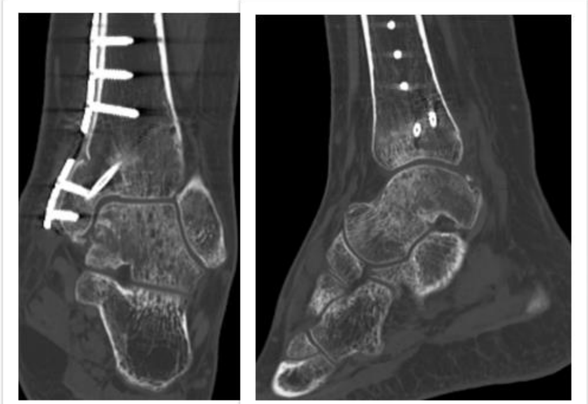

FIGURE 3

ANALYSIS & DISCUSSION

Osteoid osteoma typically presents with local pain that is most severe at night and that can be relieved by non-steroidal anti-inflammatory drugs. Depending on the location of the bony lesion, patients may present with local swelling and tenderness, bony deformities, gait disturbances or muscle atrophy (10,11). Because of an often-misleading clinical presentation, the diagnosis of periarticular osteoid osteoma will be a challenge for most clinicians (12). When clinical symptoms and pain progress and fail to respond to medications and conservative care, surgical treatment options need to be considered. Treatment options include open or arthroscopic excision of the lesion, and are dictated according to location and size (12).

This case study details our treatment of a rarely reported atypical osteoid osteoma involving the talar body. To our knowledge, examples of these tumors being treated in the talar body are rare, and there are no cases of surgical excision and repair utilizing an ACTM. Extended curettage is the most common mode of the treatment of benign bone tumors with a reported success rate as high as 90% (13). Failure of bony ingrowth, and pathological fracture have led to recommendations for the defects to be back- filled with grafts or substitutes. Bone graft substitutes like calcium phosphate and hydroxyapatite are available but their interference with inflammatory cells and immunological reaction is concerning, and their efficacy is also questionable (14,15).

Autografts are free of disease transmission or immunological reactions and have ideal properties of osteogenesis, osteoinduction and osteoconduction. However, the potency of these properties can be affected by the biological make-up of the host both in age and health. To help overcome these variables, augmentation with an extracellular matrix may facilitate cell attachment and bone formation. Bench top data supports that an environment rich in ACTM is a tremendous substrate for cellular proliferation and ultimately a release in growth factors to support healing (9). Our treatment of an osteoid osteoma utilizing an acellular connective tissue matrix and autogenous bone grafting led to a successful outcome, and an expedited return to activity. We believe that the isolated case studies involving ACTM being utilized in foot and ankle surgery are promising. These early results warrant further investigation and should be examined in higher level studies to prove their efficacy in bone healing.

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