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Glomangiomatosis Concentrated in the Ankle with Varied Appearances: A Case Report

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ABSTRACT

Glomangiomatosis is an exceedingly rare condition. This report details a case of multiple glomangiomas concentrated in the ankle of a 59-year-old male. Despite identical histological findings, the lesions displayed varied clinical features at physical examination, at magnetic resonance imaging, and upon further examination after excision. Unlike solitary glomus tumors, glomangiomas can vary widely in clinical appearance.

Glomangiomatosis, first identified by Bailey (1) in 1935, is a particular type with multiple lesions (2) and an extremely rare soft tissue lesion comprising only 2% to 3% of glomus tumors in adults (3,4). Because of the condition's rarity, identifying the specific clinical characteristics has been difficult. In the present case, multiple lesions were present in a very limited area concentrated around the ankle. Although all lesions had a histologically identical appearance, their features varied widely on physical examination, magnetic resonance imaging (MRI), and additional examination after excision. The present case demonstrates the wide variety of clinical features present in multiple lesions of glomangiomatosis.

Case Report

A 59-year-old male presented with a 30-year history of multiple protruded lesions with tenderness around his right ankle. The primary lesion had developed in the medial lower leg and had gradually enlarged to 8.0 × 3.0 × 2.0 cm in size. The nodule, measuring 1.0 × 1.0 × 1.0 cm, was palpable from the posterior side to the lateral malleolus and was the most recent lesion, manifesting 7 years earlier. Physical examination revealed a total of 5 lesions, of which 2 were medial and 3 were lateral. The size and texture of these 5 lesions varied. The primary lesion was the largest and included induration in a completely elastic soft mass. The last lesion was the smallest and presented as an elastic hard nodule-like fibromatous tumor. The other 3 masses were elastic and soft, similar to lipoma or bursitis. No thrill was present, nor were the veins dilated, and no discoloration or temperature sensitivity was present. The patient had neither a family history of vascular dysplasias nor of Recklinghausen disease. A radiograph revealed calcification, consistent with the induration in the medial primary lesion (Fig. 1*A* and *B*). On T_1-weighted MRI, all 5 nodular lesions were homogeneous and isointense to muscle in the subcutaneous layer of both malleolar areas of the right lower leg (Fig. 1*C*). These appeared heterogeneous, with high-signal intensity on T_2-weighted images (Fig. 1*D*). A gadolinium-enhanced MRI scan demonstrated wide variation in the image intensity of the lesions (Fig. 1*E*). The patient underwent excisional biopsy of all 5 lesions (Fig. 2). The gross appearance of the 5 lesions varied (Fig. 3*A*). The 2 lesions on the medial side had a yellowish appearance, similar to a giant cell tumor. The 3 lesions on the lateral side were similar to hemangioma, bursitis, and a fibrous nodule, respectively. Despite these differences in the gross features, the histologic appearance of the lesions was identical. Microscopic examination revealed dilated, variably sized vessels, with walls surrounded by regular, round glomus cells. No cytologic atypia was present (Fig. 3*B*). Immunohistochemical staining showed that the glomus cells were positive for smooth muscle actin and negative for S-100 protein and CD34. These findings confirmed the diagnosis of glomangiomatosis. The postoperative course was uneventful. At the most recent follow-up visit 2 years postoperatively, no recurrence was identified.

Discussion

In contrast to most glomus tumors, which have been defined as small benign neoplasms and occur in the dermis and subcutis of the

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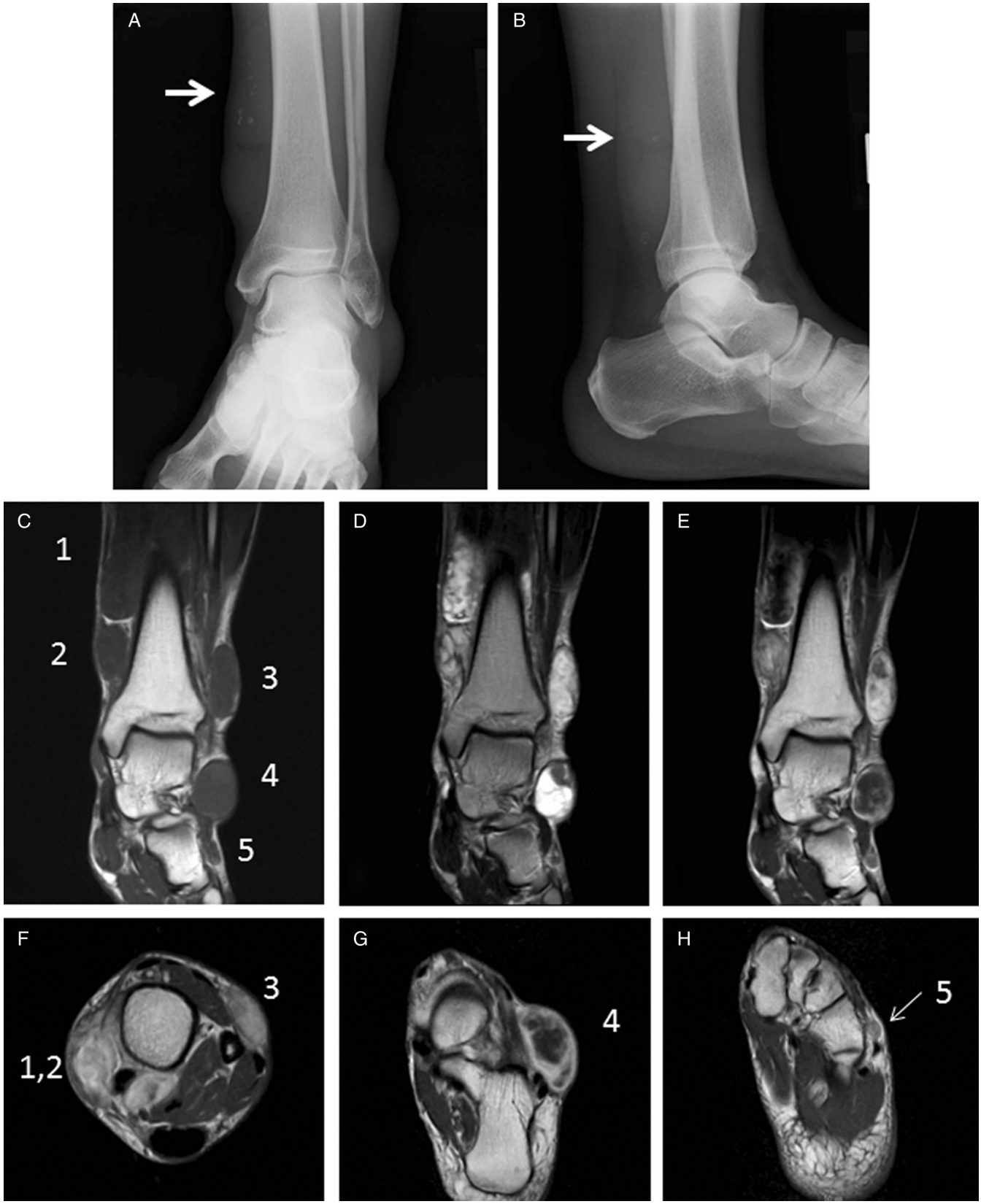

Fig. 1. Diagnostic imaging of glomangiomatosis. A 59-year-old male presented with multiple lesions in his right lower leg. (*A* and *B*) Radiograph revealing calcification in part of the medial lesion *(arrows)*. (*C*) Coronal, T_1-weighted magnetic resonance image demonstrating 5 nodular lesions (numbered serially from 1 to 5) with homogenous isointensity to muscle in the subcutaneous layer of both malleolar areas of the right lower leg. (*D*) These lesions appeared heterogeneous with high-signal intensity on T_2-weighted imaging. (*E*) Gadolinium-enhanced magnetic resonance image showing these lesions were enhanced with varied intensity. (*F–H*) Axial views of gadolinium-enhanced magnetic resonance imaging.