Rare Case of Benign Bone Histiocytofibroma of the Proximal End of the Tibia

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Abstract

Introduction: Histiocytofibroma is a mesenchymal tumour located mainly in the skin of the extremities, rarely in the bone (especially in long bones, including the femur, tibia and iliac bone). Little work has described the clinical, pathological and prognostic characteristics of bone histiocytofibromas, making it difficult to differentiate them from other rare lesions such as Giant Cell Tumour (GCT) and nonosseogenic fibroma. Their surgical management is conservative with a low recurrence rate, we report a case of bone histiocytofibroma of the proximal end of the tibia in a young adolescent patient treated by curettage and cement filling.

Keywords: Adolescent • Tumor • Benign Bone Histiocytofibroma • Proximal tibia

Introduction

Benign Fibrous Histiocytoma (BFH) is a mesenchymal tumour located primarily in the skin of the extremities; rarely in bone (especially in long bones, including the femur, tibia and ilium) [1-3].

Bone HFB is a rare skeletal tumour representing approximately 1% of all benign bone tumours operated on [4]. Little work has described the clinical, pathological and prognostic characteristics of bone HFBs, making it difficult to differentiate them from other rare lesions such as Giant Cell Tumour (GCT) and non-osseogenic fibroma. We report a case of bone histiocytofibroma of the proximal end of the tibia in a young patient treated by curettage and cement filling.

Case Report and Medical Observations

She was 16-years-old, with no notable pathological antecedents. She did not report any previous trauma, and for the last 3 years had been presenting with a palpable swelling of progressive and slightly painful appearance on the right knee, with partial functional impotence of the right lower limb, all of

which evolved in a context of apyrexia and preservation of the general state of health.

Clinical examination revealed a firm, poorly limited, palpable, fixed-to-deep-plane swelling on the medial surface of the upper right tibia measuring 5 cm long, with no opposing inflammatory signs.

There was no distal neurovascular deficit, and the regional lymph nodes were not palpable. Knee and ankle joints were free.

Standard radiographs of the knee and the leg face and profile showed a lacunar osteolytic off-centre metaphyseal image with peripheral osteocondensation, 5 cm in diameter, partitioned, respecting the lateral cortex and blowing the medial cortex which is thinned without periosteal reaction opposite, classified as type I according to Lodwick (Figure 1).

An MRI objective was an osteolytic image in heterogeneous iso signal T1 hyper signal T2 with intense enhancement after injection of contrast medium, blowing the cortex, with infiltration of the soft parts opposite. This lesion measured 4 × 3 cm (Figure 2). Laboratory results, including a complete blood count, routine biochemical studies, and measurements of serum calcium, serum phosphorus, and parathyroid hormone were unremarkable. A biopsy was performed that favoured a benign bone histiocytofibroma.

Tumour resection was performed, followed by curettage of the tumour and filling of the residual cavity with polymethylmethacrylate bone cement (Figures 3 and 4).

The macroscopic appearance of the tumour was grey and firm (Figure 3). Histological examination revealed a proliferation of spindle cells without atypia, benign and adopting a storifome pattern with focal dissociation of the bone trabeculae. Histiocytes were involved in this *CD163* immunohistochemically labelled tumour proliferation, without signs of malignancy compatible with the histological appearance of a benign histiocytofibroma of bone. Based on the clinical, radiological, especially anatomopathological and immunohistochemical aspects, we have retained the diagnosis of benign bone HCF. An analgesic immobilization with a knee brace followed the intervention as well as early rehabilitation of the knee without support for 45 days. After six months of follow-up, the functional outcome was excellent and no recurrence was observed after one year of follow-up, but long-term follow-up is essential.



Figure 1. Standard radiograph of the right knee showing a lacunar, metaphyseal off-centre osteolytic image with peripheral osteocondensation, partitioned, respecting the lateral cortex and blowing the medial cortex which is thinned without periosteal reaction opposite.

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Figure 2. Objective MRI an osteolytic lesion in heterogeneous iso signal T1 hyper signal T2 with intense enhancement after injection of contrast material, blowing the cortex, with infiltration of the soft tissues opposite. This lesion measured 4×3 cm.



Figure 3. Intraoperative image showing the tumour cavity and tumour tissue afterwards.



Figure 4. Radiograph of the knee (face and profile) showing the filling of the tumour with biological cement.

Discussion

Benign HCF is a well-established soft tissue tumour that was first described by Stout and Lattes in 1967 [1]. Most commonly, this tumour affects the skin of the extremities [2,3]. It is a rare entity: fewer than 100 cases have been reported in the literature. Approximately 40% of HCFs occur in long bones, most commonly the femur and tibia. Up to 25% of cases involve the pelvic bones, particularly the iliac bone. However, this tumour can involve any bone. In long bones, benign HCF occupies the epiphysis or diaphysis.

Patients ranged in age from 6 to 74 years at diagnosis, with 60% being over 20 years of age, with a slight predominance of females as in our case [5,6].

Little work has described the radiographic characteristics of HFB in the humerus, it is sometimes difficult to distinguish this tumour from other rare tumours with multi-compartmental osteolytic lesions such as TCG. HFB appears in radiography as a very limited, osteolytic, uni- or multilocular lesion with a sclerotic border. It can be centered or eccentric. Cortical extension may be present, especially in flat bones. Periosteal reaction and extension to adjacent tissues are rarely described [4-7]. Hamada et al. reported that the HCF of the femur had a sclerotic border that was a point of differentiation with giant cell tumours [8].

Soft tissue extension is not present. Rarely, the lesion is less well defined, with blurred borders suggestive of malignancy [9]. When the tumour is located at the end of a long bone, it may be central or eccentric and is indistinguishable from a giant cell tumour [10,11]. The diagnosis of benign HCF should be discussed in cases where the clinical and radiographic context is also compatible with that of a non-osseogenic fibroid or giant cell tumour.

Macroscopically, the diameter of lesions is in the order of 3 cm or less, although cases have been reported up to 7 cm [9]. Tumour tissue is usually firm, greyish-white, and often contains irregular yellow to reddish-brown foci. Histopathologically, HCF is a benign lesion of bone composed of spindle-shaped fibroblasts with a storiform arrangement and a variable mixture of small multinucleated giant cells.

Bone curettage and grafting, marginal excision and wide excision are the treatment modalities for benign histiocytofibromas [12,13]. The recurrence rate of HCF is low. Bielamowicz et al. reported that only 11% (2/18) of patients had a recurrence after local exeresis for cervico-cephalic HCF [2]. Metastases had never been reported.

Conclusion

Benign bone histiocytofibroma is a rare and little known benign tumour of bone. Because its microscopic features are non-pathomonic, diagnosis can be difficult. However, by systematically examining the patient's symptoms, tumour location, and radiological and microscopic features, other benign lesions can be ruled out but the prognosis remains excellent after surgical curettage, usually curative resection and filling with acrylic cement.

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