Rare disease

Benign fibrous histiocytoma of a metacarpal bone

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Benign fibrous histiocytoma (BFH) is a rare skeletal tumour. Its occurrence in the metacarpal bones is even rarer. The management in this location can be challenging, involving tumour resection and metacarpal reconstruction. The authors present a case of BFH involving the second metacarpal bone.

BACKGROUND

Benign fibrous histiocytoma (BFH) is a tumour whose histology is characterised by both fibroblastic and histiocytic differentiation.¹ It occurs predominantly in the skin.¹ Its occurrence in bone is rare and only a few cases have been described in the literature. We report a case of BFH occurring in the second metacarpal (D2) of the left hand in a middle-aged man.

CASE PRESENTATION

A 35-year-old right-handed male trader presented to us with complaints of constant left hand pain of 5 months' duration and a progressive swelling of the same hand of 4 months' duration. These symptoms resulted in inability to use the hand. There was no preceding trauma to the hand.

Physical examination revealed a diffuse, tender, firm, fixed swelling on the dorsoradial surface of the hand extending to the first and second web spaces (figure 1). The range of motion of the D2 metacarpophalangeal (MCP) and proximal interphalangeal joints was painfully restricted. There was no distal neurovascular deficit and the regional lymph nodes were not palpable.

INVESTIGATIONS

The results of full blood count and erythrocyte sedimentation rate were normal. Chest radiograph was also normal. A radiograph of the hand showed a lytic lesion on the distal third of the shaft of the D2 metacarpal extending into the head just short of the MCP joint. There was soft tissue extension with no periosteal reaction and no sclerosis (figure 2). Fine needle aspiration for cytology showed a probably benign lesion.

DIFFERENTIAL DIAGNOSIS

A provisional diagnosis of giant cell tumour of the D2 metacarpal left hand (Campanacci stage 3) was made. Enchondroma, aneurysmal bone cyst and malignant fibrous histiocytoma were considered as differential diagnoses.

TREATMENT

Two stage surgery was proposed – a wide excision in the first stage and then reconstructive surgery in the second. At surgery (first stage), the mass was found to be unencapsulated but well circumscribed and arising from the D2 and extending into the soft tissue. Wide excision of the tumour was performed under a non-exsanguinating tourniquet. The resection margin included the head of the metacarpal down to its proximal third just distal to its base leaving a 6 cm bone gap (figure 3).

Gross examination showed a globular tumour measuring 7.0×6.0×4.5 cm. Cut section showed a fleshy golden yellow surface (figure 4).

Histological examination

Histological examination showed a tumour composed of proliferating fibrohistiocytic spindle shaped cells admixed focally with giant cells. In some areas, the spindle cells formed a storiform pattern. Also seen were areas with cholesterol clefts, numerous foamy macrophages and several blood channels. There were no features of malignancy and the tumour margin was free (figure 5). These findings are in keeping with a histological diagnosis of BFH of the metacarpal.

Second operation

Two weeks after the first surgery, the patient had a reconstruction plus arthrodesis of the D2 MCP joint using an autogenous subperiosteally resected fibula strut graft 6 cm in length. The graft was anchored with a 6-hole semitubular plate and screws (figure 6). Surgery was performed under an exsanguinating tourniquet.

OUTCOME AND FOLLOW-UP

The patient developed tourniquet palsy in the immediate postoperative period. The hand was splinted in a below elbow volar cast for 6 weeks. Thereafter, active and active-assisted physiotherapy were commenced. Six months after surgery at follow-up in the clinic, the patient had recovered from the palsy with a good range of motion at the wrist and other fingers. Radiograph showed incorporation

Figure 1 Clinical photograph of the affected hand.

of the graft (figure 7). There was no clinical or radiological evidence of recurrence.

DISCUSSION

BFH is a very rare bone tumour. It accounts for approximately 1% of all surgically treated benign bone tumours.² Long before its reported occurrence in bone, it had been known to occur in the dermis, superficial subcutaneous tissue and deep soft tissue.³ Occurrence in bone is most frequent in the ilium, femur, pelvis, humerus, tibia, fibula and ribs, and less frequent in the vertebrae, sacrum, skull and mandible.⁴ In the long bone, BFH can occur in the epiphysis, diaphysis and metaphysis.² Ours occurred in the distal third of the shaft of the metacarpal with an extension into the head of the metacarpal.

BFH constituted 10 out of 11 087 skeletal tumours studied by Dahlin⁵ and by Unni and Dahlin.⁶ All 10 cases were adults between the ages of 23 and 60 years with a male to female ratio (M:F) of 4:3. A median age of 28 years (range 24–52 years) and a M:F of 4:6 were reported by Grohs *et al* in their series.²

Local pain is the main symptom in patients with BFH.⁷ This usually prompts the patient to present early as in our patient. Swelling is not a common presenting symptom.⁷ However, the paucity of soft tissue around the metacarpal region would have accounted for the obvious swelling in our case within a short time of presentation. The radiographic appearance of BFH is commonly that of an osteolytic lesion with sharply defined and sclerotic borders.⁸ Ours exhibited a radiographic appearance devoid of sclerosis; the borders were ill-defined. MRI of BFH, although non-specific, shows T1-weighted images of the same signal intensity as skeletal muscle.⁴

Histologically, BFH is indistinguishable from non-ossifying fibroma and metaphyseal fibrous defect, which are exclusively located in the metaphyses of the long bone.⁹ Their typical radiographic appearance as well as their occurrence in children and adolescents distinguish them from

Figure 2 Preoperative plain radiograph of the lesion.

BFH. Although clinical and radiological features are used to distinguish BFH from non-ossifying fibroma and metaphyseal fibrous defect, histology is needed to distinguish it from tumours such as giant cell tumour, aneurysmal bone cyst, osteoblastoma, enchondroma and malignant fibrous histiocytoma as they may have similar clinical and radiological features as BFH.¹⁰ Such was our own experience, as the clinical diagnosis of giant cell tumour made was different from the definitive histological diagnosis.

Grossly, BFH has cystic and solid components with the cysts containing yellow or haemorrhagic fluid.⁸ The components are well demarcated but unencapsulated. The specimen from our patient was entirely solid with no cystic component.

Microscopically, the fibroblasts form a fascicle or a storiform pattern.¹¹ The matrix consists of collagen including reticulum, foamy histiocytes, multinucleate giant cells of the Touton type, collection of lymphocytes and scanty plasma cells. Within the matrix are cholesterol clefts. The cholesterol content is responsible for the yellow colour. Immunohistochemistry shows widespread reactivity for vimentin and CD 68 indicating a histiocytic differentiation.⁹ The aggressive form of BFH may show features of atypia and increased mitoses and must be distinguished from low grade malignant fibrous histiocytoma. No correlation has been noted between this aggressive histological form and incidence of recurrence.¹² Soft tissue extension, as noted in our case, is not a common finding in BFH.

Curettage and bone grafting, marginal excision and wide excision are the treatment modalities for BFH.⁹ Local recurrence is said to be rare. Three out of eight patients treated with curettage and grafting had local recurrence in

Figure 3 Postoperative (first stage) plain radiograph showing the extent of resection.

Figure 4 Cut section of the resected specimen.

the Clarke *et al* series.⁸ Local recurrence even after wide excision has been reported. A local recurrence rate of 5–25% is reported in the literature with the recurrence rate related to the size of the tumour.^{1 2}

Figure 5 Histopathological appearance of benign fibrous histiocytoma of the second metacarpal embedded in paraffin, stained with H&E and viewed at 40 $\times$. The left insert shows fibrohistiocytic tumour with bland nuclei; the right insert shows cholesterol clefts with inflammatory reaction.

There are no reliable reports in the literature of malignant change with BFH, although Unni and Dahlin in their study of 10 cases described one patient with pulmonary metastasis 2 years after a local recurrence in the distal femur in which amputation was performed.⁶ There is no literature to support the role of adjuvant chemo and/or radiotherapy in the treatment of BFH.

In our patient, we elected to perform a staged procedure to limit the duration of surgery and prevent tourniquet related complications. However, the patient did have tourniquet palsy after surgery which may have been due to increased pressure as the tourniquet time was well below the absolute maximum of 3 h for limb surgery.¹³ Moreover, there is no objective way of controlling the pressure in the non-pneumatic tourniquet used in this case. After resection, MCP joint reconstruction can be achieved by metatarsal substitution, a combined iliac crest and metatarsal head graft or prosthetic replacement.¹⁴ However, in our

Figure 6 Immediate postoperative (second stage) plain radiograph.

Figure 7 Plain radiograph at 6 months after the operation showing gradual incorporation of the graft.

patient, because of lack of experience with these treatment modalities and the low functional demands of the patient, we performed a local resection followed by reconstruction using a non-vascularised fibular graft with arthrodesis of the MCP joint.

Although no local recurrence has been found after 6 months of surgery in our case, a long-term follow-up is essential.

Competing interests None.

Patient consent Obtained.

Learning points

- ▶ Benign fibrous histiocytoma of the metacarpal can occur.
- ▶ Its radiographic features may be atypical.
- ▶ Management follows the long established protocol for treating benign musculoskeletal neoplasms.

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