CASE REPORT

Aneurysmal bone cyst of medial cuneiform and a novel surgical technique for mid-foot reconstruction

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SUMMARY

Aneurysmal bone cyst of the foot is extremely rare and the involvement of medial cuneiform has never been reported in the literature. In this report, we describe a 15-year-old boy who presented with a 6-month history of pain and swelling in his left foot. Radiograph demonstrated a lytic lesion in the medial cuneiform extending on to the middle cuneiform, the navicular bone and the base of the first metatarsal. En bloc resection of the lesion was performed using a dorsal longitudinal incision along the first ray. Tricortical iliac crest graft was harvested and shaped to fill the defect. Two drill holes were made and the tibialis anterior tendon was attached to the graft. Prepared, morcellised allograft was placed along the junction of autograft and host bone. At 1-year follow-up, the patient was pain free, the medial arch of the foot was maintained and the graft had united with the host bone.

BACKGROUND

Aneurysmal bone cyst (ABC) is an eccentric, osteolytic lesion with an expanded, remodelled 'ballooned' cortex that is usually diagnosed in the second decade of life.¹ ABC involving the bones of the foot is rare. In a review of 897 cases of ABC from 12 published studies, the incidence in foot bones was 6.3%.² In Mankin's series of 150 ABCs over 20 years, only eight cases were noted in the foot.³ Moreover, the involvement of medial cuneiform bone has never been reported in the literature, to the best of our knowledge. We report a rare case of ABC arising from medial cuneiform and a novel surgical technique employed in treating this patient.

CASE PRESENTATION

A 15-year-old boy presented with a 6-month history of pain and swelling in his left foot. The pain and swelling were progressively increasing and the pain had worsened over the past 2 months. There was no history of trauma, fever or any other constitutional symptoms. Clinical examination revealed a 5 cm×3 cm swelling over the medial aspect of his left foot about 5 cm anterior to the medial malleolus. The swelling was tender, bony hard, immobile, arising from underlying bone with ill-defined margins.

INVESTIGATIONS

Radiograph of the foot demonstrated a lytic lesion in the medial cuneiform extending on to the middle cuneiform, the navicular bone and the base of the first metatarsal (figure 1). MRI revealed a well-defined multiloculated expansile lytic lesion

Figure 1 Anteroposterior and oblique radiographs of the left foot in a 15-year-old boy demonstrating a lytic lesion in the medial cuneiform extending on to the middle cuneiform, the navicular bone and the base of the first metatarsal.

with multiple thin septations and fluid levels suggestive of ABC (figure 2). A core biopsy was performed which showed a giant cell lesion with ABC-like areas.

DIFFERENTIAL DIAGNOSIS

- Giant-cell tumour (GCT) with secondary ABC
- Primary ABC

TREATMENT

A working diagnosis of GCT with secondary ABC was made. The patient and his parents were explained regarding the various treatment options and they opted for en bloc resection.

En bloc resection of the lesion was performed using a dorsal longitudinal incision along the first ray. The lesion was found to involve the medial and

Figure 2 T2-weighted sagittal section MRI of the same patient demonstrating a well-defined multiloculated expansile lytic lesion with multiple thin septations and fluid levels, suggestive of aneurysmal bone cyst.

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middle cuneiform, base of the first metatarsal and the distal part of the navicular bone. The tibialis anterior tendon was cut after applying stay sutures proximal to its insertion. A marginal wide excision of the lesion was performed. Tricortical iliac crest graft was harvested from the left iliac crest and the graft was shaped to match the defect. The graft was placed in such a way that the concave inner table of the graft faced plantarward and the thick crest margin formed the medial border to reconstitute the medial arch of the foot. K-wires were placed to secure the graft in position. It is important to harvest graft from the ipsilateral iliac crest so as to match the three-dimensional (3D) structure of the medial longitudinal arch. Two drill holes were made just medial to the thicker medial border of the graft. The tibialis anterior tendon was passed through the holes and sutured back to itself so as to create a sling. Care should be taken to avoid excessive tension to the tibialis anterior tendon. Prepared, morcellised allograft was placed along the junction of autograft and host bone.

OUTCOME AND FOLLOW-UP

The excised tumour was sent for histopathological examination on which a definitive diagnosis of ABC was made. H&E-stained sections showed blood-filled dilated spaces separated by connective tissue septae containing spindle cells, new bone (figure 3A, $\times 40$) and few osteoclastic giant cells (figure 3B, $\times 400$). The patient was kept non-weight bearing for 6 weeks. Follow-up radiograph performed at 6 weeks showed early signs of graft union. At this time, K-wires were removed and partial weight-bearing mobilisation was initiated on a short leg walking

cast. Complete weight bearing was initiated at 10 weeks postsurgery after confirming union of the graft clinically and radiologically (figure 4). At 1-year follow-up, the patient remained pain free, the medial arch of the foot was maintained and the graft had united with the host bone. There was no evidence of recurrence. Active dorsiflexion was possible up to 35° and the power was 5/5 as per Medical Research Council grading (figure 5).

DISCUSSION

ABC of the foot remains a diagnostic and therapeutic challenge because of the rarity of its occurrence, the complex anatomy of the foot and its resemblance to other giant cell containing lesions.⁴ Of the 238 ABCs treated at Mayo Clinic between 1910 and 1979, only 6 patients had ABC involving the tarsal bones.⁵ Casadei *et al*,⁶ in their review of 257 benign bone tumours of the foot, mentioned that ABC of small tarsal bones are rare compared with GCT at this site. ABC needs to be differentiated from GCT, giant cell reparative granuloma, Brown's tumour of hyperparathyroidism and telangiectatic osteosarcoma. Furthermore, ABC can occur secondarily in other tumours such as GCT, osteoblastoma, angioma and chondroblastoma.¹ Histopathological examination of the lesion is essential before proceeding with a definitive treatment and the diagnosis may remain elusive until excision biopsy is performed, as demonstrated in our case.⁴

The aetiopathogenesis of ABC remains unclear. The vascular theory proposed by Lichtenstein⁷ is the most popular theory until now.⁷ According to this theory, venous hypertension causes collection of blood within the bone and ballooning of its cortices that can rapidly enlarge and erode into adjacent structures.

Figure 3 H&E-stained sections showing blood-filled dilated spaces separated by connective tissue septae containing spindle cells, new bone (A, $\times 40$) and few osteoclastic giant cells (B, $\times 400$).

Figure 4 Postoperative radiograph of the foot showing union of graft with the host bone. Drill holes made in the autograft to reattach the tibialis anterior tendon are visible.

Figure 5 Postoperative clinical picture demonstrating the maintained medial longitudinal arch and active dorsiflexion at 10 weeks following surgery.

The venous hypertension is caused by a poorly understood vascular insult resulting in either venous thrombosis or anomalous arteriovenous malformation. However, recent evidence suggests that primary ABC is a neoplastic disorder caused by a genetic alteration. Oliveira *et al*⁸ noted USP6 and CDH11 gene rearrangements in 36 of 52 primary ABCs. This genetic alteration was absent in the so-called secondary ABC.

Multiple modalities have been described to treat ABC including intralesional injection, embolisation, curettage with bone grafting and resection with reconstruction.^{4–9} The treatment of choice for most ABC of the foot is curettage with bone grafting.¹⁰ Mankin *et al*,³ in their review of 150 cases of ABC, found recurrence rate of 22% in patients treated with curettage and bone grafting as opposed to 5% recurrence following resection. Chowdhry *et al*¹⁰ reviewed the results of curettage in 14 patients with ABC of the foot and they reported 21% recurrence rate following this procedure. They concluded that ABCs of the feet will respond to treatment similar to ABCs that occur elsewhere in the body. Our patient and his parents took an informed decision to undergo a wide excision as they preferred a surgery with the least chance of recurrence.

Restoring the structure and functions of the mid-foot following resection is a challenging task because of its complex anatomy. The longitudinal and transverse arches need to be reconstructed and major tendons, such as the tibialis anterior tendon in our case, need to be reattached so as to avoid functional disability. Some authors have used free vascularised fibular autograft for reconstructing the medial arch of the foot following wide excision of malignant bone tumours involving the metatarsals.^{11–12} Ahn *et al*¹³ described a technique of mid-foot reconstruction using iliac crest autograft in a patient with GCT of medial cuneiform. The graft was fixed to host bone using plates, and the tibialis anterior tendon was reattached to the navicular bone through drill holes. However, the patient developed wound dehiscence over the plate at 6 weeks which required a rotation skin flap. Plate removal was performed at 12 months following the index procedure to relieve continuous skin irritation. Our technique of mid-foot reconstruction differed from that of Ahn *et al* in several ways. Iliac crest graft was harvested from the ipsilateral side to match the 3D geometry of the medial longitudinal

arch. Minimal hardware (K-wires) was used for fixing the graft to avoid wound dehiscence. The tibialis anterior tendon was reattached to the graft (instead of the navicular bone) closer to its original insertion for effective functioning. Morcellised allograft was added to assist union and to fill the mismatch in shape between the defect and the graft.

There have been reports of ABC arising from the cuboid and metatarsal bones but involvement of medial cuneiform has never been reported.^{14–15} Moreover, this case report highlights the importance of reconstructing the arches of foot following en bloc resection of mid-foot tumours.

Learning points

- ▶ Primary aneurysmal bone cyst can occur in bones of the foot including the medial cuneiform.
- ▶ Ipsilateral tricortical iliac crest autograft is a viable alternative to autologous fibular graft for mid-foot reconstruction.
- ▶ Reattaching the tibialis anterior tendon using drill holes made in the autograft is important to prevent foot drop.

Contributors SAK was the chief surgeon and DJ was the primary assistant surgeon of the surgical team that performed the surgery. VSK was part of the surgical team and authored the manuscript under the guidance of co-authors. ARM was the pathologist who reported the excision biopsy specimen.

Competing interests None.

Patient consent Obtained.

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