



Case Report:

A Recurrent Aneurysmal Bone Cyst of Cuboid Bone with Soft Tissue

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Abstract: Aneurysmal bone cyst of cuboid is a rare benign tumor and at times it can be aggressive. Here we would like to report a case of recurrent ABC of cuboid with soft tissue involvement managed by resection reverse sural artery island flap and fibula graft. Only 3 cases of ABC cuboid were reported in the literature till date.

Key Words: Aneurysmal bone cyst; Cuboid; Reverse sural artery island flap; Fibula graft

Introduction:

Aneurysmal bone cyst (ABC) is a rare benign locally aggressive bone lesion which involves extremities. The other rare sites involved by ABC are skull, mandible (4%), clavicle and ribs (5%) and feet (3%).[1] In the feet the most commonly involved site is metatarsals.[2] Till date only 3 cases of cuboidal ABC were reported in the literature to the best of our knowledge.[3-5] This case adds to the literature a recurrent cuboidal ABC managed successfully.

Case Report:

A 41 years old female presented with painful swelling in the left foot on the lateral aspect one year back for which she underwent curettage initially in a peripheral hospital. She developed recurrent swelling and pain with restriction of movement after 6 months of initial surgery. An X-ray radiograph of the foot showed a lytic lesion destroying the 1st, 2nd metatarsals, lateral cuneiform, talus and calcaneum. She underwent wider resection (2nd, 3rd, 4th, 5th metatarsals, talus, lateral, intermediate cuneiform were excised) and the wound was closed primarily. She was later referred to us for further management.(Fig.1) Review of the previous histology slides confirmed the diagnosis of ABC classic variant.

She was planned for delayed reverse sural artery island flap and fibula graft. We performed delayed resection after 2 weeks as we required a skin flap of 14x 10 cms and involved the upper 3rd of the leg. The flap was raised based on sural nerve, median sural artery and short saphenous vein which form a subdermal plexus and hence the deep fascia to be included with the flap and the wider pedicle to be maintained

as shown in Fig.2. An avascular fibula graft was harvested from the opposite leg and was used to replace 2nd and 3rd metatarsals which were stabilized by 2 mm- Kirschner wire directed from proximal to distal through the toe in a retrograde fashion through the graft to the talus. In the postoperative period foot elevation was maintained and patient was put on low molecular weight heparin for 5 days. The flap developed venous congestion after 6 hours of surgery for which the wound is opened and the ligated end of the short saphenous vein and one of the perforator of short saphenous vein were opened to drain out the venous congestion. The wound was resutured after 4 days. The patient was put on non weight bearing below knee plaster cast for 6 weeks and the Kirschner was removed after 12 weeks with gradual weight bearing and patient was able to resume full weight bearing by 20 weeks. Six months after surgery there has been no recurrence.



Fig. 1a: Antero-Posterior radiograph of left foot showing lytic lesion involving cuboid bone suggestive of aneurysmal bone cyst. 1b: Recurrent ABC causing destruction of 3rd, 4th, 5th metatarsal, cuneiform bone with soft tissue involvement
1c: Post wide excisional status of ABC of left foot

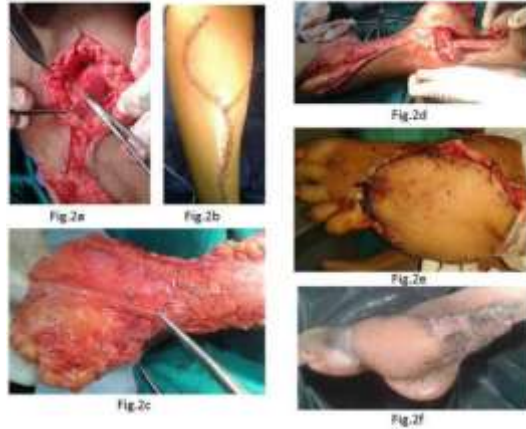


Fig. 2a: Ligation of perforator of medial sural artery 2b: Incision of the flap for delayed reverse sural flap 2c: The flap is incised completely after 2 weeks and a delay procedure was performed with the sural pedicle 2d: An intraoperative photograph after reconstructing 2nd and 3rd metatarsal with autologous fibular graft bridging the osseous gap after removal of tumor fixed to talus 2e: Sutures removed due to venous congestion of the flap 2f: A well healed wound and flap after 4 months of surgery.

Discussion:

Aneurysmal bone cyst was first described by Jaffe and Lichtenstein in 1942 and it is considered as a reactive process of unknown etiology.[6] It is been defined by WHO as a benign osteolytic lesion with blood filled spaces intervened by connective tissue septa containing trabecular or osteoid with occasional osteoclastic giant cells. Various hypotheses have been proposed as an etiological factor in development of ABC. Some consider it as result of vascular malformation and others consider it due to increased vascular pressure leading to dilatation and rupture of vessels.[7] One more hypothesis is that ABC is due to a traumatic pathology.[8] There are 4 phases in the development of ABC namely the inactive, stabilization, active and aggressive phases.[3] Histologically there are two variants, the classic and solid types. The classic type is associated with more recurrences than the solid type. Various modalities of treatment were proposed like curettage alone or with adjuvants (High speed burr, phenolisation, argon beam coagulation, cryosurgery, etc.) or resection of the bone. In one study of ABC of feet, curettage alone lead to a recurrence rate of 21%. Various reasons have been proposed for the increased recurrence of ABC like location of the lesion, size, mitotic index and phase of the development of ABC.[3] Till date only 3 cases of ABC of cuboid were reported. This is the first case of recurrent ABC of cuboid bone managed by resection and reconstruction using a delayed RSIF.

Conclusion:

ABC of cuboid is a rare entity and it can be aggressive with soft tissue involvement. They may need aggressive resection and reconstruction to decrease the chance of recurrence.

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