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RESEARCH ARTICLE

ANEURYSMAL BONE CYST OF THE 2ND METACARPAL IN CHILDREN : A CASE REPORT

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Abstract

Aneurysmal bone cysts are benign, rare tumorthat mainly affects childrenand young adults. It a large lytic lesion that appear most often around metaphyseal bone, principally located in the long bones, pelvis, and spine and rarely in other anatomical district such as the hand.The lesions are locally aggressive with high recurrence rates, treated with radical excision and filling of the residual cavity with bone substitute. The patient has shown no signs of recurrence. However, the patient showed an excellent outcome with a satisfactory active range of motion and grip strength. Many treatment modalities have been reported in literature, However, controversy exists in the literature regarding optimal treatment, including radiation, curettage and bone grafting, cryotherapy, and excision. Due to its rarity in the hand, no evidence-based treatment regimen has been established.

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Introduction:-

Aneurysmal Bone cyst is a rare benign bone tumor, described firstly by Jaffe and Lichtenstein in 1942,accounts for 1-2% of all primary bone tumors [1]. It is predominantly a disease of children and adolescents with a slight female preponderance. It shows predilection [2,3] for long bones and vertebral column, rarely involves the bones of hand less than 5% of all ABC occurs in long bones of hand [4].

Pathogenesis of ABC is obscure. Various other theories about origin of ABC makes it true neoplasm. Lichtenstein suggests that persistent local disturbance in hemodynamics causes marked increase in venous pressure and leads to development of dilated engorged vascular bed [5,6].

As with most bone lesions, pain and swelling ofseveral months' duration were the most frequent complaints.

Four radiological stages of aneurysmal cyst are described in the literature as: Initial, active, stabilization, and healing [7].

Histologically, ABC appear as lytic lesion multicystic, with cavernous spaces stuffed with blood. The walls of cysts contain fibroblasts and thin strips of bone. The tumors are separated from the surrounding tissue by a thin layer of periosteal new bone [8].

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Although, various options for the treatment of aneurysmal bone cyst have been reported in the literature [9], controversy exists regarding optimal treatment. Due to its rarity in the hand, there is no established evidence-based treatment regimen.

We report a case of aneurysmal bone cyst of 2nd metacarpal, managed with complete resectionwith allograftand later followed.

Case report:
14-year-old girl, with no particular history, consulted in May 2022 for swelling of the left index finger that had appeared one year before her consultation, associated with pain, without inflammatory signs opposite, the anamnesis found no previous trauma, it was progressively increasing in size with no limitation of finger mobility.

The initial examination revealed a girl in good general condition, with a 3 cm painless and hard mass located on the left 2nd metacarpal bone. The skin was normal, with no inflammatory signs, and the rest of the clinical examination was unremarkable.

Standard radiographs of the left hand, face and profile (figure 1), revealed a well-limited central metaphyseal-diaphyseal multilacunar osteolytic image of the distal 2nd left metacarpal, blowing out the bone margins without cortical rupture and without surrounding periosteal reaction. CT images(figure 2, 3) showed a well-limited epiphyseal/diaphyseal distal mass of the 2nd left metacarpal, blowing the bone and causing cortical thinning with bone lysis, measuring 23x21mm and extending over 40mm, without periosteal reaction, with respect for the adjacent soft tissues, suggesting a chondroma or aneurysmal cyst.

Due to the extensive nature of the tumor, radical excision was carried out (figure 4), with curettage of the bone cyst and filling of the residual cavity with bone substitute, and immobilization with an anterior palmar splint for pain relief; the post-operative course was unremarkable,

Histological analysis showed a tumor proliferation of spindle-shaped cells arranged in interlaced bundles, with an ovoid nucleus showing discrete cytonuclear atypia with fine chromatin and a discrete nucleolus. Numerous osteoclastic giant cells are associated. Overall, a histological appearance compatible with an aneurysmal bone cyst.

The evolution was good, with no complications, and the patient was seen again one year later, with no pain or recurrence. Finger mobility preserved (figure 5); standard follow-up radiograph shows no recurrence (figure 6).

Figure 1:- Pre operative Xray showing multilocular expansile lytic lesion with ballooning of 2nd metacarpal.

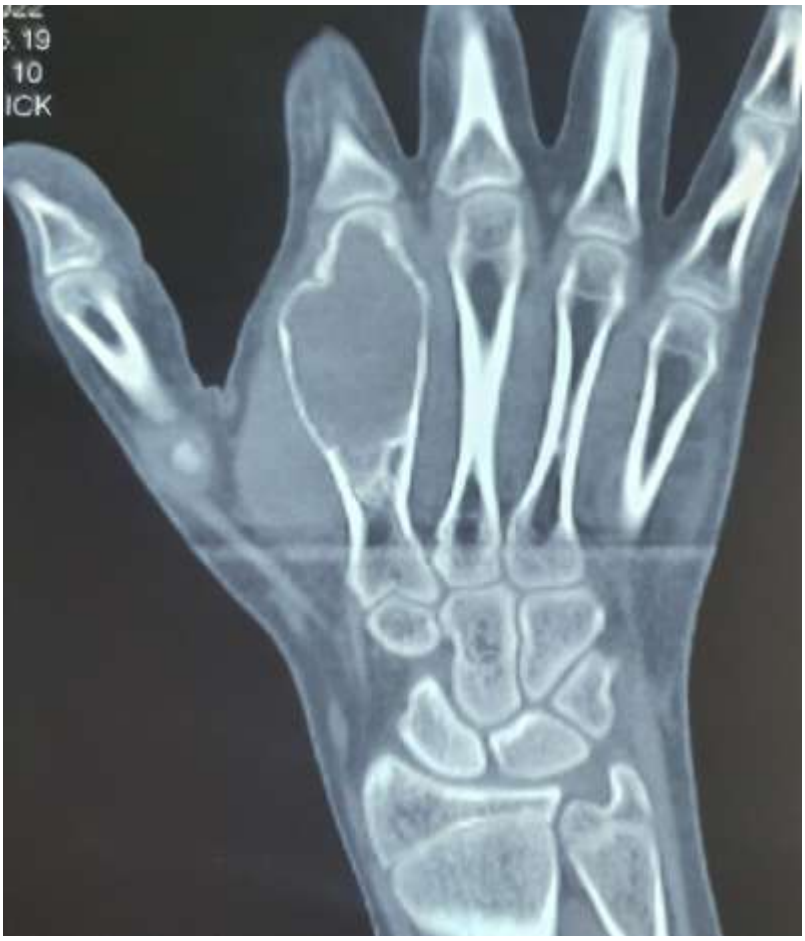


Figure 2:- CT frontal imaging showing a distal mass of the 2nd left metacarpal.

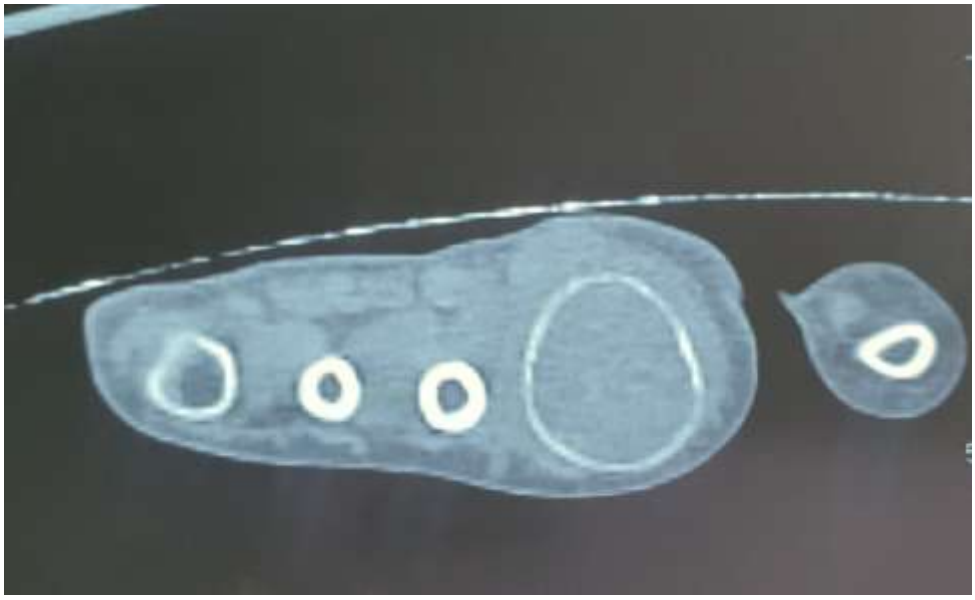


Figure 3:- CT transversalimaging showing a distal mass of the 2nd left metacarpal.

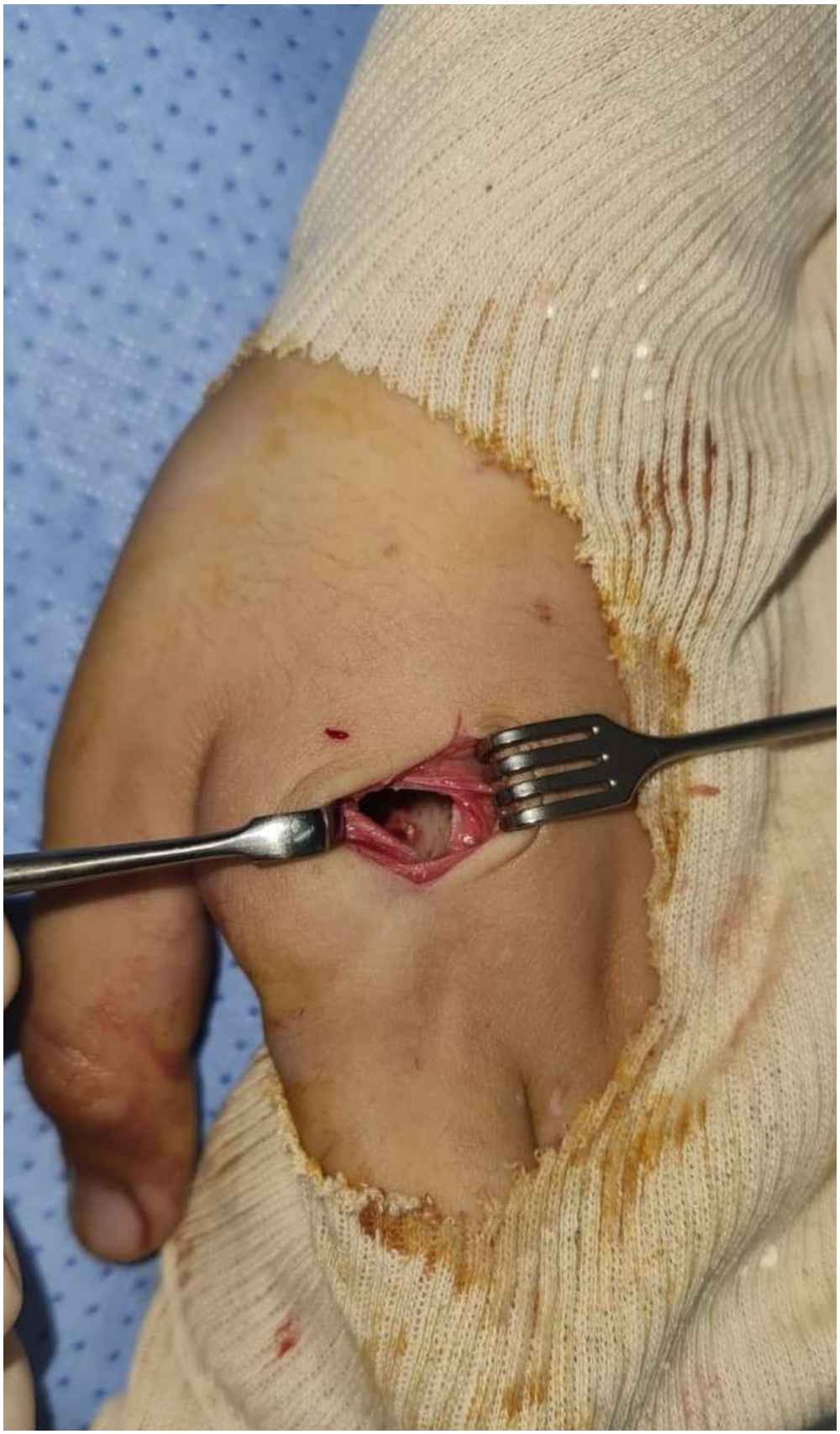


Figure 4:- Intraoperative photograph demonstrates the excision of the ABC.

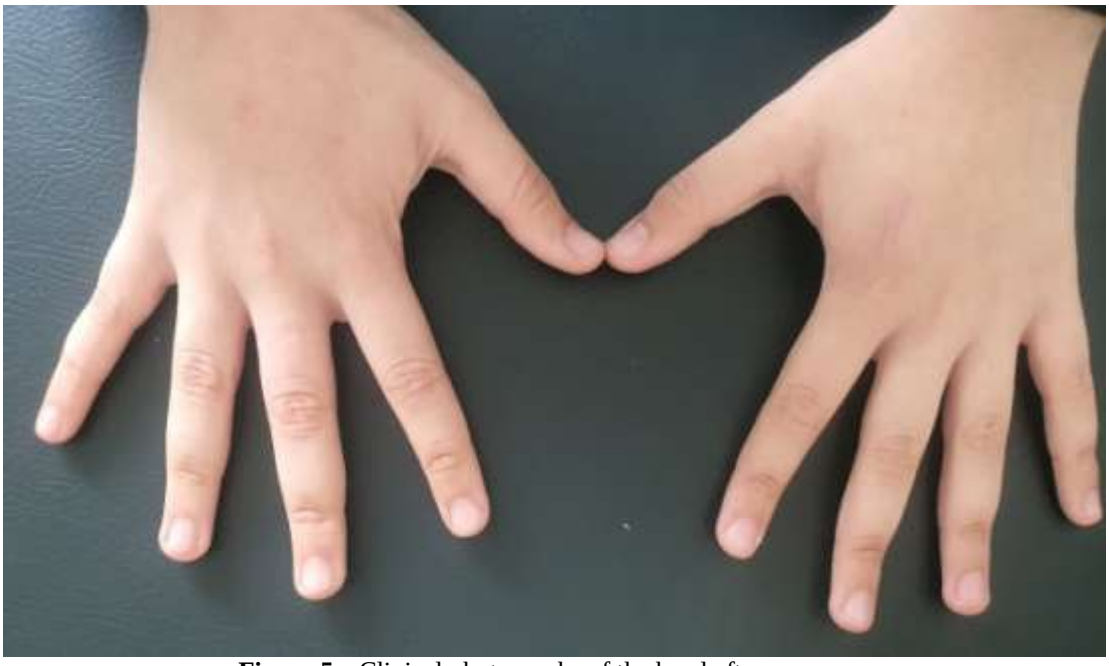
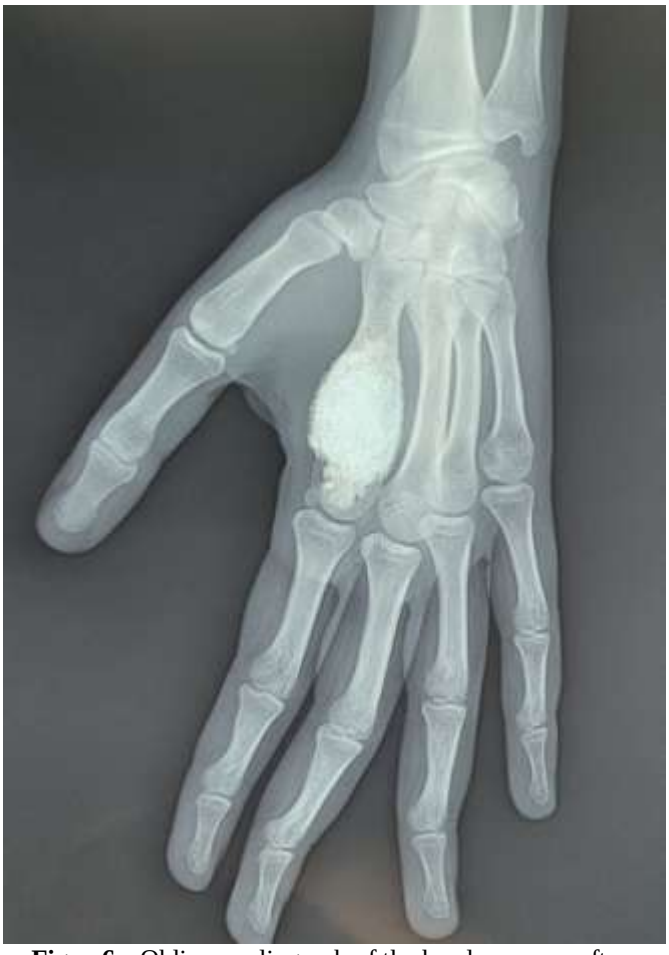


Figure 5:- Clinical photographs of the hand after onr year.



Figue 6:- Oblique radiograph of the hand one year after.

Discussion:-

The most frequent sites for ABC are the long bones of the lower limb, then the upper limb, followed by the axial skeleton and finally the flat bones. Localisations on the hands and feet are rarer, and often limited to tubular bones [10].

The positive diagnosis is based on the patient's history, clinical examination and radiographic findings, with an image of osteolysis expansive, blowing away the cortical bone without rupturing it. CT and MRI provide a more precise diagnostic approach, demonstrating the absence of soft-tissue invasion, and sometimes the presence of liquid levels in MRI, which is not specific to aneurysmal cysts. Differential diagnosis is mainly represented by giant cell tumors and enchondromas [11].

In 1985, Capanna et al. proposed an interesting classification for juxtaepiphyseal KOAs of long bones (Fig. 10) [12]:
- type I: central, non-blowing, metaphyseal or metaphysodiaphyseal ;
- type II: central KOA that has extended across the entire width of the bone, with a blowing appearance. This type is generally seen on the metaphysis or metaphysodiaphyses of small calibers long bones (fibula, radius, ulna) or on flat bones;
- type III: eccentric, intraosseous KOA, often metaphyseal;
- type IV: subperiosteal KOA, extraosseous, often diaphyseal
diaphyseal (rare) ;
- type V: subperiosteal KOA, intra- and extraosseous, often
often metaphyseal.

The goal of treatment is the eradication of the lesion, preservation of function without recurrence. controversy exists in the literature regarding treatment. Many modalities have been reported such as radiation, curettage and bone grafting, cryotherapy, and excision.

The treatment of aneurismal bone cyst is controversial. Different propositions have been reported like Curettage alone or combined with adjuvant therapy[13].

However, different studies have reported unacceptably high rates of recurrence and have recommended more aggressive resections, leading to challenging reconstructions. [5, 14].

Athanasian et al reported that curettage is the essential step, which alone can lead to a good result [11]. However, the systematic association of an autograft seems preferable, as it often leads to better results than curettage alone, and enables a filling that provides a better mechanical foundation, even in small lesions [5,12]. More advanced lesions that are inaccessible to intralesional treatment, or located in the subchondral bone, should be considered for resection in conjunction with a vascularized or non-vascularized fibula graft [13,14].

Due to its rarity in the hand, no evidence-based treatment has been established.

Despite the functional loss frequently encountered after resection, this option is sometimes the only one recommended [15] in order to minimize the risk of recurrence. In the long bones of the hand, more aggressive conservative treatments such as cryotherapy or cement, often used for other skeletal localizations [16], seem in the case of hand bones to be reserved for lesions that are resistant to curettage-grafting, or that are very advanced from the outset [17]. These treatments have their own complications, affecting the surrounding soft tissues and leading to neurapraxia, burns or necrosis, the more easily the lesion is distal. Some authors, however, report convincing results with cryotherapy as a first-line treatment, underlining the adverse effects of cryotherapy on autografting, necessitating a first stage of curettage with cryotherapy and cement filling, followed by a second stage of autograft surgery [17].

Pallapati etal[18] reported a non-vascularized metatarsal transfer as treatment of a large bone defect. They show favorable results in 9 patients after a mean follow-up of 44 months. Thetreatmentconsists on matched metatarsal autograft harvested from the foot aiming to preserve movement and function of the MCP joint.

In other study, it has been shown that bone defects that are left empty heal just as well as when filled with a bone substitute and defect protection alone was sufficient to allow for healing even of critical size defects [19].

Our case shows that good results can be achieved although the tumor involved the growth plate and had destroyed the entire diaphysis of the metacarpal, and also the efficacity of resection and allograft bone substitute.

Conclusion:-

An en block resection of aneurismal bone cyst of metacarpal with allograft with bone substitute is a good choice for treatment for Aneurysmal bone cyst of hand bone with good result in our patient. It gives structural construct with full return of functional activity.

Declaration of competing interest

The authors have no conflict of interest to declare.

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