



## A rare case of aneurysmal bone cyst of cuboid bone in a 10-year-old girl

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**Aneurysmal bone cysts (ABC) are rare and they represent 1- 1.4 % of all primary bone tumors. ABC of cuboid bone are extremely rare, especially in children. Very few cases have been reported in the literature since 1967. We present a case of pathological fracture of cuboid bone due to an aneurysmal cyst in a 10-year-old girl. Surgery was performed, which included open biopsy with aspiration and intralesional curettage . Bone defect was then filled in by fully synthetic cancellous bone graft substitute consisting of pure  $\beta$ -tricalcium phosphate. Aneurysmal cyst of cuboid bone with pathological fracture could be successfully treated with pure  $\beta$ -tricalcium phosphate as a bone graft substitute. This procedure is safe with excellent outcome. Follow up to nearly four years did not show any recurrence.**

**Keywords :** aneurysmal cyst ; cuboid bone ; synthetic bone graft.

### INTRODUCTION

The term aneurysmal bone cyst (ABC) was coined by Jaffe and Lichtenstein in 1942 (10,15). It is an expansile cystic lesion and may occur in any bone in the body. Although benign, the ABCs are categorized as locally aggressive lesions that rarely undergo spontaneous healing (7) and can cause extensive weakening of the bone and impinge on the surrounding tissues. Primary ABCs represent

1-1.4 % of all primary bone tumours, with more than 70 % of the cases occurring during the first two decades of life (8,11). The most common locations are the long bones and spine (3,4,12). However, unusual locations may lead to a false diagnosis (2,15). Main clinical symptom of ABC is painful swelling, although some remain asymptomatic. Even though, there are many typical radiological findings to diagnose ABC, an open biopsy is performed to confirm diagnosis. In children, most pathological fractures are secondary to benign tumours and due to ABC occurs in an one third up to 72% of cases. Literature is void about ABC of cuboid bone in pediatric patients. Only two cases of aneurysmal

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cuboid bone cyst in the pediatric population have been reported since 1967 and one chondroblastoma with an associated aneurysmal bone cyst in cuboid bone (6,13). We present a case of a 10-year-old girl with pathological fracture treated surgically with curettage and  $\beta$ -tricalcium phosphate as a bone graft substitute. Beta-tricalcium phosphate granules improve osteogenesis in vitro and establish innovative osteo-regenerators for bone tissue engineering in vivo (5).



*Fig.1.* — Clinical presentation of the left foot ankle



*Fig.2.* — Pre-operative plain radiogram of the left foot

## CASE PRESENTATION

Our patient was a 10-year-old girl with a history of the painful left foot that was lasting for eight months. The onset of symptoms started after simple fall. The pain was insidious in onset, but gradually increased together with swelling. She was treated in the other institution as a sprained ankle. Local clinical examination showed hazelnut-sized swelling with definite margins and tenderness, on the outside lateral edge of her left foot. Overlying skin was uninvolved (Fig. 1).

First diagnose in our hospital, based on conventional radiographs was an old fracture (Fig. 2). Initial treatment by the junior physician was short leg cast for three weeks, without any improvement after the treatment was finished. After surgeon made suspicion about diagnosis, MRI scan was done (Fig. 3) and it revealed pathological fracture on the base of cuboid bone cyst; cyst was fulfilling a 90% of bone volume, bone edema, hemorrhage, reparatory process, soft tissue edema with no aggressive changes were described.

Once the diagnosis of ABC of left cuboid was made, the patient underwent open biopsy with aspiration and intralesional curettage of the cyst (Fig. 4). After curettage bone defect was filled in by the fully synthetic cancellous bone graft substitute, consisting of pure  $\beta$ -Tricalcium Phosphate ( $\beta$ -TCP) (Fig. 5). In this case we used chronOS® Bone Void Filler, small granule size of 0,7-1,4mm to fulfill the cyst cavity.

The patient passed post-op period without complications. Histopathological examination confirmed diagnose of ABC of cuboid bone. Follow up was at three months intervals during the first year. At 12 months post-op, the patient was ambulating normally with full range of motion at all left foot joints (Fig. 6). Her recent radiography shows very good incorporation of the bone graft substitute also with no signs of recurrence after four years from surgery (Fig. 7).

## DISCUSSION

Location of ABCs in the foot is quite rare and location in the cuboid bone is an exceptional occurrence.



Fig.3. — Pre-operative oblique MRI scan of the left foot



Fig.4. — Aneurysmal cyst after the curettage



Fig.5. — Cyst fulfilled with bone graft substitute



Fig.6. — Plain radiograph after 12 months from surgery

Within the Mayo Clinic case register – 143 foot locations were described out of 238 cases (5.5%)— only six of which were at tarsal level (2.5%) (8,10). A pathological fracture occurs in bone tissue that is pathological, weak and remodeled, with altered or reduced mechanical and viscoelastic properties (3). Our patient was a girl with a misdiagnosis of sprain ankle. After the MRI scan, the right diagnose was made and surgery was performed. The indications for treatment in case of ABC are pain, pathological fracture, the risk of such a fracture, large cyst situated in a weight-bearing area, etc. The treatment depends on the surgeon’s experience but also of

hospital’s opportunities in equipment and medical resources. Selective arterial embolization, intralesional injections (calcitonin, methylprednisolone, ETHIBLOC) percutaneous aspiration and injection of an aqueous solution of calcium sulphate are some of the non-surgical possibilities in the treatment of ABC (12). Intralesional curettage through a cortical window or en-bloc or wide excision are some surgical possibilities. Adjuvant therapy (phenol, methylmethacrylate, liquid nitrogen) which extends the area of treatment beyond that which can be physically excised is an eventuality. Fulfilling of the empty bone space after curettage with bone auto



**Fig.7.** — Plain radiograph after 42 months from surgery

or allograft is also an option. We decided to perform intralesional curettage and to use chronOS® small granules (synthetic allograft) for filling in the cyst cavity. The successful use of  $\beta$ -TCP for ABC in other locations in children has been reported. Age, complication, packing methods and granule diameters have a significant influence on  $\beta$ -TCP degradation. A histological analysis of biopsy showed that  $\beta$ -TCP supported the growth of fibrous tissue, vascular tissue, as well as bone tissue into the implants. Single  $\beta$ -TCP use is an advantageous alternative to allografts for lacunar bone defect repair as it is well incorporated without any adverse reaction to the synthetic material (1,17). The excellent results has been also reported in patients with benign bone lesions treated by  $\beta$ -TCP mixed with autologous bone marrow showed neither recurrent disease nor complications (9). There were no complications and no signs of recurrence in our patient. We have found in the literature that local recurrence rates are high, ranging from 12% to 60% and to decrease these rates after intralesional curettage of ABCs, different adjuvant treatments have been recommended. Liquid nitrogen spray and argon beam coagulation have provided the lowest recurrence rates, but unlike the high-speed burr, these adjuvants are not always available in operating rooms (18). The only parameter affecting partial healing and local recurrence was the proximity of the lesion to the growth plate (4).

### CONCLUSION

Diagnose of ABC of cuboid bone with pathological fracture can be challenging and overlooked.

Although it was diagnosed late the outcome was beneficial for a child. We believe that aneurysmal cyst of cuboid bone could be successfully treated with pure  $\beta$ -tricalcium phosphate as a synthetic bone graft substitute. This procedure is safe, the second operation of autologous bone grafting is avoided and good clinical and functional results are achieved, without recurrence.

### REFERENCES

1. **Altermatt S, Schwöbel M, Pochon JP.** Operative treatment of solitary bone cysts with tricalcium phosphate ceramic. A 1 to 7 year follow-up. *Eur J Pediatr Surg.* 1992 Jun ; 2(3) : 180-2.
2. **Aycan OE, Çamurcu İY, Özer D et al.** Unusual localizations of unicameral bone cysts and aneurysmal bone cysts: A retrospective review of 451 cases. *Acta Orthop Belg.* 2015 Jun ; 81(2) : 209-12.
3. **Canavese F, Samba A, Rousset M.** Pathological fractures in children: Diagnosis and treatment options. *Orthopaedics & Traumatology: Surgery & Research* 102 (2016) S149–S159.
4. **Erol B, Topkar MO, Caliskan E et al.** Surgical treatment of active or aggressive aneurysmal bone cysts in children. *J Pediatr Orthop B.* 2015 Sep ; 24(5) : 461-8.
5. **Gao P, Zhang H, Liu Y et al.** Beta-tricalcium phosphate granules improve osteogenesis in vitro and establish innovative osteo-regenerators for bone tissue engineering in vivo. *Sci Rep.* 2016 Mar 22 ; 6 : 23367
6. **Kashuk KB, Hanft JR, Schabler JA et al.** Aneurysmal bone cyst of the cuboid. *J Am Podiatr Med Assoc.* 1990 Nov ; 80(11) : 588-94.
7. **Luna AR, Fahandez-Saddi H, Garcia AV et al.** Aneurysmal bone cyst in children involving infrequent locations. Report on two cases. *Chir Organi Mov.* 2004 Oct-Dec ; 89(4) : 347-52.

8. **Novais EN, Zimmerman AK, Lewallen LW et al.** Functional outcomes and quality of life following surgical treatment of aneurysmal bone cysts of the pelvis in children. *J Child Orthop* (2014) ; 8 : 281-288.
9. **Ondruš S, Straka M, Bajerová J.** Tricalcium phosphate mixed with autologous bone marrow in the treatment of benign cystic bone lesions in children. *Acta Chir Orthop Traumatol Cech.* 2011 ; 78(6) : 544-50.
10. **Parashari UC, Khanduri S, Upadhyay D et al.** Radiologic and pathologic correlation of aneurysmal bone cysts at unusual sites. *J Can Res Ther* 2012 ; 8 : 103-5.
11. **Rajasekaran S, Aiyer SN, Shetty AP et al.** Aneurysmal bone cyst of C2 treated with novel anterior reconstruction and stabilization. *Eur Spine J.* 2016 Mar 23.
12. **Reddy KI, Sinnaeve F, Gaston CL et al.** Aneurysmal bone cysts: do simple treatments work? *Clin Orthop Rel Res* 2014 ; 472 : 1901-10.
13. **Sessions W, Siegel HJ, Thomas J et al.** Chondroblastoma With Associated Aneurysmal Bone Cyst of the Cuboid. *The Journal of Foot & Ankle Surgery* 2005 ; 44(1) : 64-67.
14. **Schmidt RG, Kabbani YM, Mayer DP.** Aneurysmal bone cyst. *J Am Podiatr Med Assoc.* 1993 Oct ; 83(10) : 595-7.
15. **Unni KK, Inwards YC.** Conditions that normally simulate primary neoplasms of the bone. In: Unni KK, Inwards YC, editors. *Dahlin's Bone Tumors.* 6th ed. Philadelphia: Lippincott Williams and Wilkins ; 2010 : 305-80.
16. **Verrinaa F, Dagninob G, Gulinoc MT et al.** Aneurysmal bone cyst of the cuboid. *Foot and Ankle Surgery* (2003) ; 9 : 193-196.
17. **Wang Z, Guo Z, Bai H et al.** Clinical evaluation of  $\beta$ -TCP in the treatment of lacunar bone defects: a prospective, randomized controlled study. *Mater Sci Eng C Mater Biol Appl.* 2013 May 1 ; 33(4) : 1894-9.
18. **Wang EHM, Marfori ML, Serrano MVT et al.** Is Curettage and High-speed Burring Sufficient Treatment for Aneurysmal Bone Cysts? *Clin Orthop Relat Res* (2014) ; 472 : 3483-3488.