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Juxtacortical aneurysmatic bone cyst of the trapezium: A case report and review of the literature

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Abstract

Objective: Here we describe a case of an ABC of the trapezium that, to our knowledge, has been mentioned in the literature only twice.
Case description: We describe the case of a solid variant of juxtacortical aneurysmatic bone cyst affecting the trapezium in a 38-year-old healthy male patient complaining about pain and reduction of range of motion. The excision, after completion of diagnostic workup with advanced imaging (CT scan and MRI), led to complete regression of symptoms within two weeks and a recurrence free period of five years.
Conclusion: Aneurysmatic bone cysts are rare, benign bone tumors representing 2% of all bone tumors. Only 3-5% of cases occur in the hand, the carpal bones being the least affected. The knowledge of this rare tumor is important to properly address treatment and postoperative care.

Keywords: Aneurysmatic bone cyst, Trapezium, Carpal bones, Bone tumor, Hand surgery.

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Introduction

Aneurysmatic bone cysts (ABC) represent a rare entity among the benign bone tumors (approximately 2% of all bone tumors) while involvement of the hand has been reported in only 3-5% of the cases (1,2). The carpal bones are less involved than the metacarpals and the phalanges (3). The ABC is reported to have a high recurrence rate, however in the isolated cases involving carpal bones no recurrence of the lesion was found (**Table 1**). Here we describe a case of an ABC of the trapezium that, to our knowledge, has been mentioned in the literature only twice (4,5).

Table 1: Summary of the described ABC cases involving carpal bones

Reference	Bone involved	Side	Patient age (year)	Gender	Trauma	Treatment	Recurrence	Follow up (months)
Barbieri et al, 1984 (18)	Hamate	L	30	F	No	Curettage, autologous bone graft	No	6
Lin et al, 1984 (19)	Hamate	NA	16	M	NA	Excision	No	15
Frassica et al, 1988 (5)	Trapezium	R	20	M	NA	Excision	No	176
Mankin et al, 1995 (20)	Lunate	R	48	F	No	Tumor and bone excision, scapho-capitate arthrodesis	No	6
Platt et al, 1995 (21)	Capitate	R	14	F	Yes	Conservative	No	16
Kabukcuoglu et al, 2003 (22)	Hamate	L	18	M	NA	Curettage, autologous bone graft, excision of hamate	No	36
Sakamoto et al, 2006 (3)	Capitate	R	15	M	Yes	Curettage, autologous bone graft	No	48
Tuzuner et al, 2006 (4)	Trapezium	R	19	M	NA	Tumor and bone excision, suspension arthroplasty	No	11
Mavrogenis et al, 2010 (23)	Hamate	L	34	F	No	Curettage, autologous bone graft	No	12

NA: Not available, R: Right, L: Left, M: Male, F: Female

Case description

A 38-year-old male patient presented with pain in the base of the right thumb for 9 months. Conservative measures using splinting, topical and systemic analgesia as well as hand therapy could not alleviate his symptoms. 10 years earlier the patient had suffered a right thumb distortion, which had been treated conservatively with tapes resulting in a complete regression of symptoms over time. The left dominant patient was otherwise healthy and working in an office. Clinical examination showed a painful and immobile swelling dorsal to the first carpo-metacarpal joint, a pain related restriction of active range of motion in thumb flexion and abduction as well as both ulnar and radial deviation of the wrist. Plain radiographs showed an amorphous cystic bone expansion on the radial margin of the trapezium (**Figure 1**). Further diagnostic imaging confirmed the thinning of the trapezium cortex in the computed tomography (CT) scan (**Figure 2**) and magnetic resonance imaging (MRI) (**Figure 3**) showed the lesion to be calcified with no involvement of the extensor tendons. There was no evidence of cancellous bone involvement. An excisional biopsy was performed (**Figures 4 and 5**).

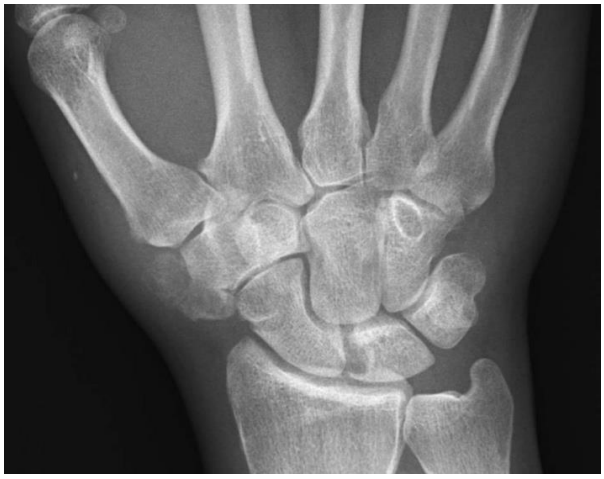

Figure 1: Preoperative plain posteroanterior radiograph of the right, non-dominant, wrist presenting the exophytic bony lesion radial to the trapezium.

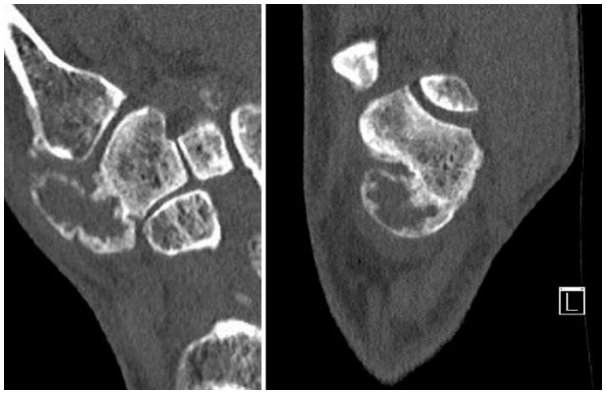

Figure 2: Coronal and sagittal view of the CT scan (at 0.5 mm thickness) showing a thinning of the trapezium cortex and the osseous mass.

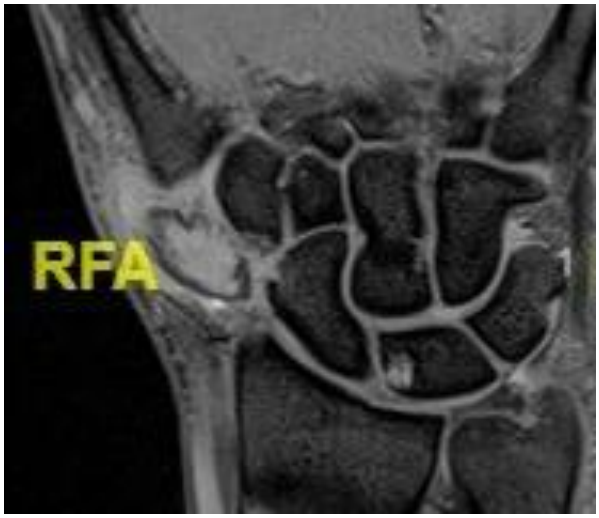

Figure 3: T2-weighted coronal MRI of the juxtacortical aneurysmatic bone cyst of the trapezium.

Histologically, a well-capillarized and undirected spindle cell proliferation was observed with numerous multinucleated giant cells of osteoclast type (**Figure 6**). Fluorescence in situ hybridization (FISH) analysis of the USP6 gene revealed a rearrangement in the tumor cells and the diagnosis of aneursymatic bone cyst (solid type) was made.

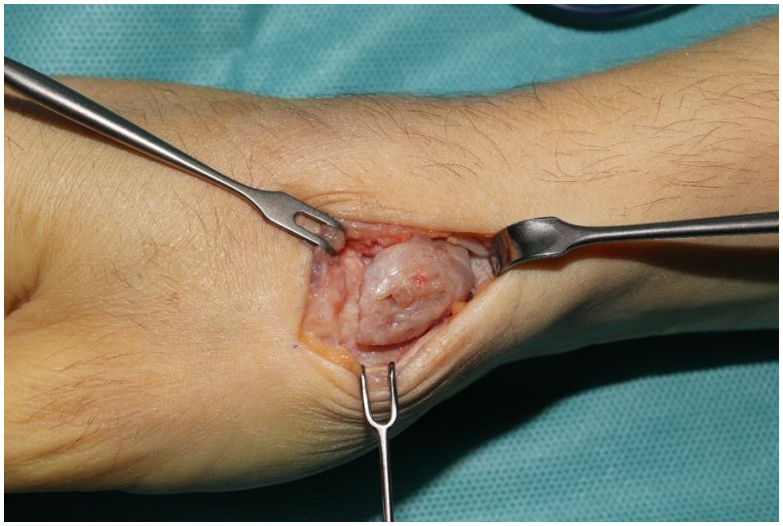

Figure 4: The intraoperative aspect of the tumor before its excision. Macroscopically no signs of infiltration were observed, and the hardness of the tumor resembled the hardness of the trapezium itself.

The patient made a full recovery after two weeks of postoperative splinting and could resume his activities without suffering from pain or discomfort. Five years postoperatively there has been no clinical or radiological sign of a recurrence.

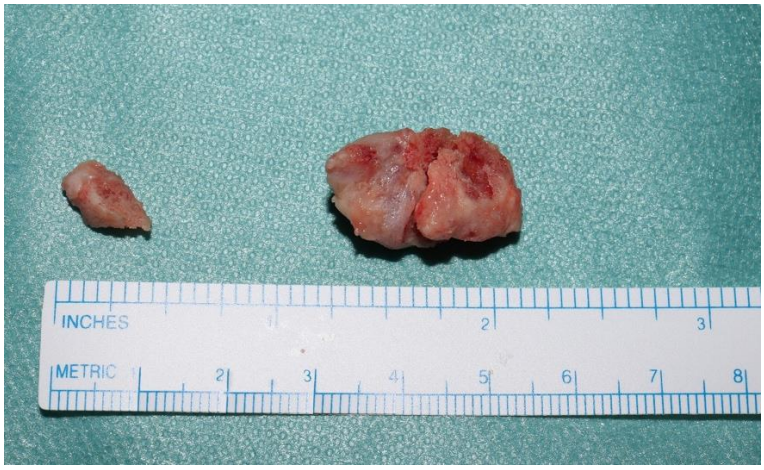

Figure 5: The excised tumor

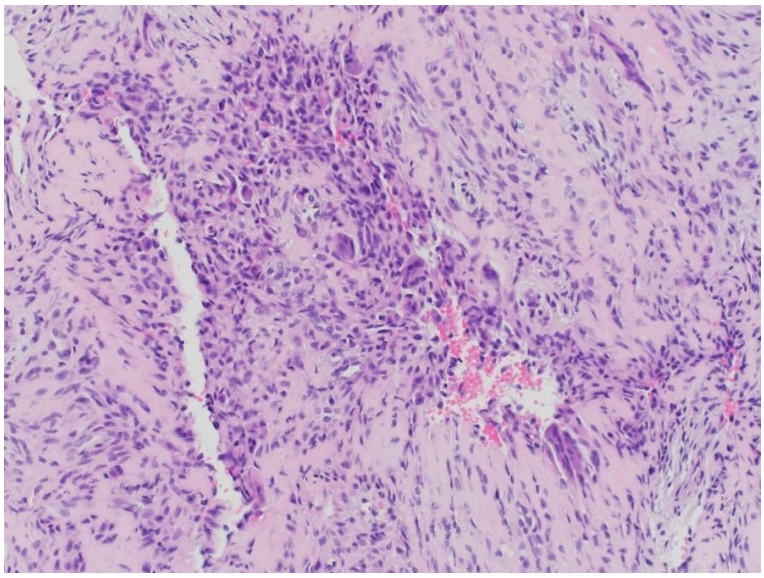

Figure 6: Hematoxylin and eosin staining of the ABC at 200x magnification. The giant cells are rather small (average <10 nuclei per cell) and do not dominate the histologic picture as in a conventional giant cell tumor of bone (immunoreaction against H3-3A G34W furthermore negative). At higher magnification, the cellular pattern remains monomorphic and without high grade atypia or atypical mitotic figures. No necrosis is observed.

Discussion

ABC has been first described by Vam Arsdale in 1893 as an ossifying hamartoma and later renamed into an aneurysmal bone cyst by Jaffe and Lichtenstein in 1942 (6). The etiology is still unknown and pathogenesis is poorly understood. The theory behind the primary ABC is a vascular anomaly as a venous insufficiency or an arteriovenous malformation that would contribute to the buildup of the cyst, which would eventually fill with blood and erode the bony structures. The ABC however also contains cytogenetic rearrangements of the USP6 gene present in the spindle

cells that make the ABC behave like a neoplasm with volume expansion and relatively high recurrence rates (7,8). Hemorrhage into preexisting bone lesions such as eosinophilic granuloma, chondroblastoma, osteoblastoma, non-ossifying fibroma, chondromyxoid fibroma and giant cell tumor was formerly considered as secondary ABC, but should no longer be used due to specific genetic alterations present in primary ABC (2,8,9). Trauma has a controversial role in its etiopathogenesis and has not been directly related to the ABC.

The ABC is more commonly found in long bones' metaphyses either centrally, eccentrically or subperiosteally (8). Sakamoto and colleagues (3) concluded from observing previous ABC cases of the hand in a total of 41 patients, that 37% were affecting the phalanges, 46% the metacarpal bones and 17% the carpal bones. One case described an ABC involving the radial index sesamoid of the hand[10]. In **Table 1** we summarize the described cases of ABC involving the carpal bones so far. In the location of hands and feet, the solid ABC was formerly listed separately in the WHO classification as a giant cell lesion of small tubular bones. It is usually an intraosseous lesion but may occasionally occur in unusual and juxtacortical locations as in our case.

Enneking (11) classified the ABC in different stages according to the staging system for benign bone lesions: stage I: quiescent and clinically non-evident, stage II: active and stage III: aggressive. Bertoni (12) and colleagues described a solid variant of an ABC in 1993 that was also the type of ABC affecting our patient. The histopathological presentation of the solid ABC has been well described by Sanerkin et al. in 1983 (13) with abundant fibroblastic proliferation, scattered collections of osteoclast-type giant and spindle cells, osteoblastic differentiation with osteoid production, degenerative calcifying fibromyxoid tissue and small aneurysmal sinusoids.

The diagnosis of a solid ABC includes the clinical evaluation usually with signs of tenderness and swelling over the lesion. Plain radiographs show bony expansion, lytic lesions, cortical destruction and periosteal reaction (14). This can be supported by a CT and/or an MRI that may show a fluid filled lesion, as found in the case presented here. The histological diagnosis is often difficult because of overlapping features with malignant presentations such as telangiectatic osteosarcoma (8,12). Yet the presence of the USP6 gene thanks to the FISH analysis confirms the diagnosis of the aneurysmatic bone cyst, as in this case.

Similar to the case of our patient, the ABC typically affects younger patients, up to the third decade of life while both genders are equally involved (14).

Most of the ABC have been treated surgically either with curettage and autologous bone grafting, tumor or eventually bone excision and even amputation. Cryotherapy has been described in a single case report of a recurrent aneurysmal bone cyst of the phalanx where it treated the recurrence with success (15). The curettage alone led to a higher recurrence rate in Stages II and III (11,14). The recurrence rate of ABC generally ranges from 20% to 70% (1,16). In the hand a recurrence rate of 29% (3) was observed. The tendency of recurrence in younger patients was seen more frequently in patients with open physes (17). In the hand specifically the phalanges presented the highest recurrence rate (53%), followed by the metacarpal bones (21%) while no recurrence was seen in the carpal bones (3).

In our case we opted for an excisional biopsy of the tumor alone leaving the trapezium intact. Our patient presented full recovery two weeks after the operation. The diagnosis was confirmed both histologically and with the use of molecular-genetic testing. No recurrence occurred as was confirmed clinically and radiologically on a follow up five years postoperatively.

Conclusions

The aim of this case report was to add the description of an ABC of the trapezium being treated successfully with its excision. ABC is a rare benign tumor, where the carpal bones are the least frequently involved among all known locations while literature supports the lack of recurrence in the carpal bones affected by ABC. Recognizing it in the clinical practice is important to properly address its treatment and postoperative care.

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Contributions
Research concept and design: EZV, MS, MB
Pathology reports and expertise: DT
Radiology reports and expertise: MM
Writing the article: EZV
Critical revision of the article: MS, MB
Final approval of the article: EZV, MS, MM, DT, MB
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