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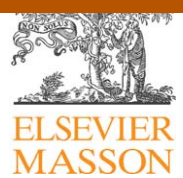

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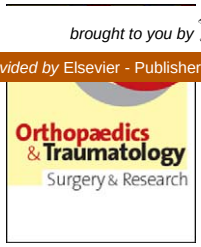

CLINICAL REPORT

Hamatum osteoblastoma

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Summary We report the case of an osteoblastoma of the hamate bone that was successfully treated by curettage. This tumor is very rare in a carpal bone and only nine cases have been reported in the literature. Pathological examination is mandatory before treatment due to the lack of distinctive clinical and radiological features. Osteoblastomas are benign, but potentially aggressive bone tumors. Treatment of the lesion may either be a conservative ''intralesional resection'' or radical ''wide en bloc resection''. The latter option, which has non-negligible functional consequences in the wrist, should be reserved for recurrence after curettage but may also be considered a primary immediate alternative for aggressive forms.

Introduction

Osteoblastomas are primary bone tumors first described and named by Lichtenstein [1] and Jaffe [2] in 1956 in two different articles after an initial publication by Dahlin and Johnson [3] in 1954 who described this entity using the term ''giant osteoid osteoma''. Osteoblastomas only represent 1% of all bone tumors (Healey and Ghelman [4]) and they are extremely rare in the hand (Manguini [5]). We report a case of osteoblastoma of the hamate bone, in which the location of the lesions affected the symptoms, and with a favourable outcome after simple curettage.

Observation

A right handed, 28-year-old patient who was a manual laborer, with no particular past medical history consulted our unit in November 2004 for painful swelling of the ulnar side of the left wrist. The patient had begun having occasional wrist pain 15 months before, which was not relieved by aspirin. In April 2004, 7 months after the pain had begun and following a minor accident, the patient's condition worsened noticeably, and swelling gradually developed on the ulnar side of the wrist.

Clinical examination revealed a firm, painful, poorly differentiated swelling of the ulnar side of the wrist. The fingers and wrist were stiff and tender and the skin was bluish and shiny, suggesting a complex regional pain syndrome.

The patient, who had been in pain for more than a year, provided the first X-rays from 15 months before which did not show any obvious anomalies in the hamate bone, although

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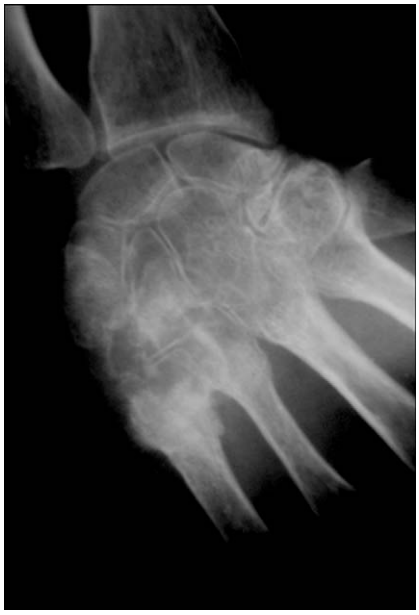

Figure 1 Marked osteolysis of the base of the hamate bone and the adjacent bases of the fourth and fifth metacarpals. Consolidation of the fracture of the base of the fifth metacarpal.

the scaphoid was tilted horizontally (with a ring sign) without scapholunar diastasis and a scapholunar angle of less than 70°. Another X-ray performed 7 months later following a trauma showed a fracture at the base of the fifth metacarapal bone, with the scaphoid still tilted horizontally. On the same X-ray, a faint geode (bone cyst) could be seen in the uncinate process of the hamate bone. In addition, results of another X-ray in November 2004 showed a fracture callus of the base of the fifth metacarpal bone with diffuse spotty demineralisation with significant osteolysis in the base of the hamate bone and the adjacent bases of the fourth and fifth metacarpal bones (Fig. 1). MRI was also performed showing a lesion in the base of the hamate bone with probable extension into the hypothenar muscles (Fig. 2). The lesion was hypointense on T1-weighted images and hyperintense on T2-weighted images, with heterogenous contrast enhancement. The bases of the fourth and fifth metacarpal bones were also hypointense on T1 images, but this was less marked.

A curettage biopsy was performed by direct anterointernal approach, without dissection of the ulnar neurovascular bundle. The tumoral tissue was inscribed in a geode (bone cyst) without extension into the soft tissues. This bone cyst was 2 cm along its main axis, and it was surrounded by a thin bone shell internally. Careful curettage was performed without filling.

Anatomopathological results showed an osteoblastoma of the hamate bone (Fig. 3). The lesion was rich in osteoblasts, with osteogenesis of more or less calcified, irregular trabeculae and a highly vacularized stroma with hemorrhagic foci. This lesion largely infiltrated the adjacent bone. Cytologically, the osteoblasts were of various sizes with a relatively abundant cytoplasm and eosinophils. Nuclei were round with nucleoli, regularly distributed chromatin and rare mitoses.

After curettage biopsy, the course of the disease rapidly improved with markedly less pain, swelling, stiffness and vasomotor difficulties; A two month post-operative X-ray showed significant bone reconstruction although a few geodes could still be seen at the base of the hamate bone. A CT scan clearly showed the geodes that had been identified during curettage.

During the final clinical follow-up in November 2006 (2 years of follow-up), the patient described occasional mechanical pain. Range of motion was slightly reduced, and there was spontaneous slight radial inclination of the wrist. Hand function was normal. On X-ray the hamate bone appeared dense with no signs of tumoral recurrence (Fig. 4). The radial inclination was obvious on X-ray, the scaphoid was still tilted horizontally in the frontal image, but the schapolunar angle was less than 70° on profile and there was no scapholunar diastasis on dynamic wrist X-rays.

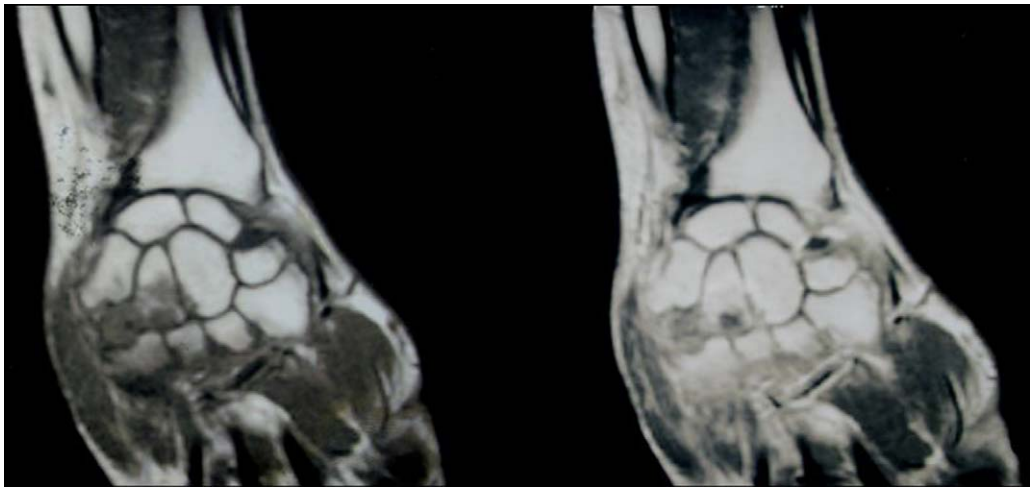

Figure 2 MRI: hypo-intense lesion on T1 images of the unciform process and the distal hamate bone. Heterogeneous uptake of contrast after injection of gadolinium.

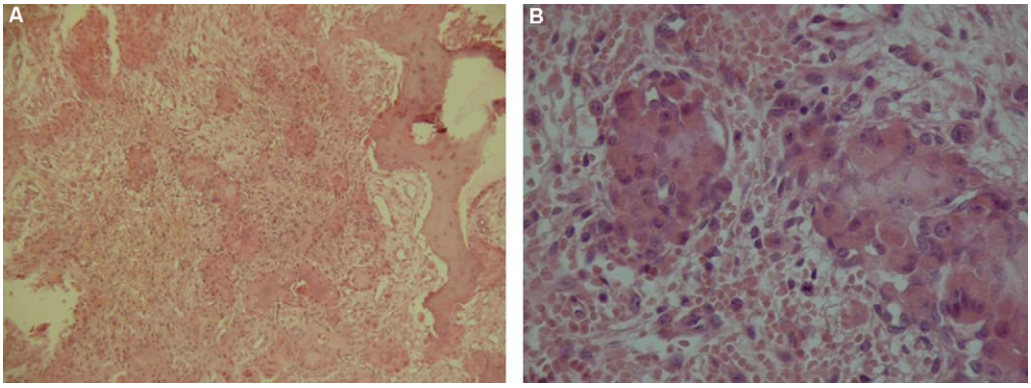

Figure 3 Histological results. A. Moderately cellular lesion dissociated by hemorrhages and which contains osteoid trabeculae of different sizes and shapes (HE × 150). B. Osteoblasts of different sizes, with abundant cytoplasm with regular nucleus and nucleoli (HE × 400).

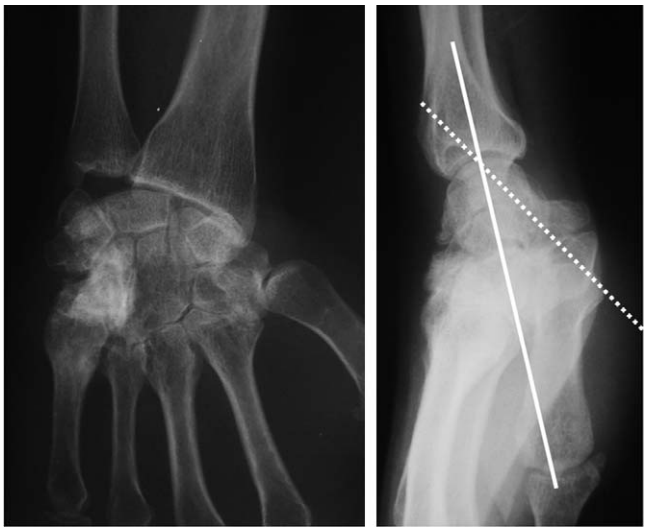

Figure 4 At 2 years of follow-up, no tumoral recurrence. Scapholunar angle less than 70°.

Discussion

Ostoeblastoma is a benign but locally aggressive tumor, which represents 1% of all primary bone tumors (Fanning and Lucas [6], Kroon and Schurmans [7], Lucas et al. [8]). Its tendency to develop in the spine is well known (40%) followed by the pelvis, the long bones, and the bones of the foot; (Bonnevialle and Railhac [9]). It is a particularly rare tumor in the carpal bones, and we only found nine cases studies in the literature. There were five scaphoid tumors (Castello et al. [10], Fanning and Lucas [6], Meade et al. [11], Ragois et al. [12], Xarchas and Leviet [13]), one in the triquetral bone (Marshall et al. [14]) and in three cases the tumor was located in the hamate bone (Apergis et al. [15], Menon et al. [16], Van Dijk et al. [17]). This tumor usually occurs in patients between 10 and 25 years old (Argenson [18]) and is twice as frequent in men (Chagnon et al. [19]). There is no characteristic pain and it is not influenced by circadian rhythms or drugs (Argenson [18]), which was also confirmed in our case. In our patient, the fracture of the base of the fifth metacarpal bone occurred after a minor trauma, and the X-ray for this trauma showed regional bone demineralisation. This fracture could therefore be considered pathological. Bone union was normal because the fracture appeared consolidated on X-ray, when the patient consulted our unit. The complex regional pain syndrome reported in our observation was obvious from clinical and radiological results, no scintigraphy was performed. We believe that this syndrome was secondary to the tumor rather than to the previously mentioned fracture. Indeed, spotty osteoporosis, which is characteristic of a complex regional pain syndrome, was already present when the fracture occurred. Moreover, this syndrome rapidly and clearly improved after curettage biopsy. Although the association of osteoblastoma and a complex regional pain syndrome is not common, it is important to note that the only triquetral tumor reported in the literature (Marshall et al. [14]), was revealed by a complex regional pain syndrome and that Schmidt [20] also reported a case of talar osteoblastoma revealed by the same syndrome. But these are anecdotal case reports and no causal relationship has been established between these two entities. The radial inclination of the wrist in our patient was associated with his wrist pain from the first, and could be seen on X-ray. The development of this antalgic posture explains the horizontal position of the scaphoid and the lack of scapholunar dissociation.

For Dahlin and Johnson [3] and Jaffe [2], an osteoblastoma is a circumscribed osteogenic tumor. For Lichtenstein and Sawyer [21], it is a predominantly osteolytic lesion but osteogenesis is possible. If the cortex is ruptured, the lesion is separated from neighboring tissue by a thin, reactional, peripheral bony shell. This was the case in our patient: Fig. 1 shows this bone shell and no invasion of neighboring structures was identified during curettage biopsy.

In a tumor of this type that does not have specific clinical or radiological signs, histological examination is necessary to determine the therapeutic strategy. Indeed, before biopsy, several differential diagnoses were possible: giant cell tumor, aneurysmal bone cyst, chondrosarcoma, or even osteosarcoma (Ragois et al. [12]). The criteria to distinguish osteoblastoma from osteoid osteoma are mainly microscopic (larger than 2 cm) and depend upon disease progression. Microscopically, the lesion may appear markedly similar to

Table 1 Course after treatment of carpal osteoblastomas.

Author	Location	Delay before the appearance of radiographic lesions	Initial treatment	Recurrence	Revision surgery	Follow-up (years)
Meade et al.[11]	Scaphoïd	32 months	Curettage graft	No	—	1
Fanning and Lucas.[6]	Scaphoïd	28 months	Curettage graft	No	—	3
Castello et al.[10]	Scaphoïd	36 months	Curettage	No	—	10
Marshall et al. [14]	Triquetrum	More than 24 months	Curettage graft	No	—	1
Apergis et al. [15]	Hamatum	14 months	Curettage graft	No	—	1
Our case	Hamatum	15 months	Curettage	No	—	2
Ragois et al. [12]	Scaphoïd	2 months	Curettage graft	4 months	Scaphoïdectomy arthrodesis 4 internal bones	3
Menon et al. [16]	Hamatum	Immediate	Curettage graft	11 months	Excision of the hamatum Carpo-metacarpal arthrodesis	4
van Dijk et al. [17]	Hamatum	NP	Curettage	6 months	Wide resection Wrist arthrodesis	3
Xarchas and Leviet [13]	Scaphoïd	More than 24 months	Primary resection Proximal carpal row	No	—	3 months

osteoid osteoma. Osteoblastoma is different from osteoid osteoma by its more abundant, vascularized loose connective tissue.

Although osteoblastomas are benign tumors, early recurrence and even progression to malignancy have been reported (Dorfmann and Weiss [22]). Certains authors, who consider the rate of recurrence of (10—19%) after resection of the lesion to be significant, systematically propose radical resection even for carpal locations (Lucas et al. [8], Van Dijk et al. [17], Jackson [23]). However this type of wide en bloc carpal resection may have fairly important functional consequences. Thus we analyzed the course of the disease after treatment in all carpal osteoblastomas reported in the literature including our case. (Table 1). Immediate radical resection of the proximal carpal row was only performed in one case in the literature (Xarchas and Leviet [13]). In the other cases, recurrence occurred in one third of the cases after treatment by curettage, with or without a graft. It is important to note that these frequent recurrences (more than 30% of the cases) complicated osteoblastomas with a potential for markedly rapid progression. The clinical history and the delay until the appearance of lesions on imaging results were markedly longer (more than 1 year) in cases without recurrence.

Dorfman and Weiss [22] have already described aggressive osteoblastomas that are more osteolytic with ''osteogenesis that sometimes occurs in irregular spicules, like osteosarcomas'' as the tumor develops. In these aggressive forms, the most important differential diagnosis is well-differentiated osteoblastic osteosarcoma. Normally, treatment of carpal osteoblastomas should be conservative, and radical resection should be limited to recurrent tumors but may also be considered in aggressive forms of the disease.

Conclusion

Carpal osteoblastomas are rare. The diagnosis is difficult because of the absence of specific clinical and radiological signs. A complex regional pain syndrome is a possible revealing sign. Resection of the tumor seems to be sufficient in lesions with a slow progression. It is therefore important to identify the aggressive forms of this benign tumor, which may then be an indication for immediate radical resection.

Conflict of interest

None.

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