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Pathologic odontoid fracture and benign fibrous histiocytoma of bone

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Abstract We present the case of a 44-year-old female patient, who sustained an odontoid fracture after a minor trauma (uncomplicated fall). The radiologic evaluation revealed a skeletal tumor of the second cervical vertebra together with a fracture line at the base of the odontoid process of the axis. The patient underwent surgery, the tumor was resected and the odontoid was stabilised using an autologous cortico-cancellous bone

graft and a halo fixator. Histologic examination revealed benign fibrous histiocytoma, which is reported to be a very rare skeletal tumor.

Key words Pathologic fracture · Odontoid process · Benign fibrous histiocytoma of bone

Introduction

Benign fibrous histiocytoma of bone (BFH) is a very rare skeletal tumor, which has also been described as “xanthoma”, “xanthofibroma” and “xanthogranuloma.” Dahlin and co-workers could find only eight cases of BFH in a total of more than 8000 skeletal tumors [7]. Although it presents as a biologically non-malignant tumor, it may show local aggressive and infiltrative growth parameters.

Case report

A previously healthy 44-year-old female patient was admitted to our institution after a minor trauma and showed clinical signs of a mild head injury (headache and nausea, no loss of consciousness, no amnesia). Due to mild neck pain additional X-rays of the cervical spine were taken. The conventional radiographs in two planes showed an area of osteolysis within the body of the second cervical vertebra extending to the odontoid process and afflicting almost half of it. Additionally there was a fracture line through the base of the odontoid without any displacement. For further evaluation conventional tomography (Fig. 1) and MRI were performed, confirming both the expansive process and the pathologic fracture through the base of the odontoid. An additional isotope bone scan did not reveal any further skeletal foci.

We therefore decided to perform surgical stabilisation. The axis was exposed via a left anterior approach, the whole tumor was

excochleated and the defect was filled up by an autologous cortico-cancellous bone graft harvested from the ipsilateral iliac crest. Due to the extension of the tumor internal fixation was impossible, so we fixed the cervical spine by use of a halo fixator (Fig. 2). The postoperative course was uneventful, the patient was discharged 3 weeks after the procedure, the fixator was removed 12 weeks post-operatively.

Histologic evaluation revealed BFH (benign fibrous histiocytoma of bone) without malignant aspects like atypical cells, mitosis etc. (Fig. 3).

At follow-up 5 years postoperatively, the patient did not report any significant complaints. There was only mild weather-dependent pain and intermittent mild dysphagia. Clinical examination showed a mild to moderate restriction of motion (15° rotatory deficit, 20° extension deficit), there were no neurological symptoms. Radiologic follow-up showed the bone graft to be completely consolidated and stable (Fig. 4A). However, conventional tomography revealed a suspicious area within the axis – possibly a recurrence of the primary tumor (Fig. 4B,C). Due to the lack of clinical symptoms the patient refused further evaluation such as MRI and also further follow-up.

Discussion

The microhistologic appearance of BFH is similar to that of non-ossifying fibroma (NOF) [5, 6, 15, 17]. Both tumors present with a predominantly fibroblastic structure with focal histiocytologic differentiation. The cells are

Fig. 1A, B Conventional tomography of the upper cervical spine after admission of the patient. There is an area of osteolysis in the body of the second cervical vertebra, extending to the odontoid process. A fracture line runs through the base of the odontoid; there is no displacement. The *arrows* mark the fracture and the osteolysis

Fig. 2A, B Postoperative anteroposterior (AP) and lateral views of the odontoid (the *arrows* mark the borders of the bone graft)

Fig. 3A, B Histologic evaluation of the resected tumor: benign fibrous histiocytoma (BFH). **A** Photomicrograph shows a predominantly fibroplastic structure with focal histiocytologic differentiation. Fusiform and oval cells form a storiform pattern. There are no signs of malignancy. **B** Section through the fracture zone. The same histologic pattern, with the fracture line in the center of the section

Fig. 4 **A** Lateral view of the upper cervical spine at follow-up 5 years postoperatively. The bone graft is stable and completely consolidated. **B** Conventional tomograph, AP view. The fracture is solidly healed; however, there is a suspicious area of osteolysis (signs of recurrence?) in the body of the second cervical vertebra (*arrows*). **C** Conventional tomograph, lateral view. The borders of the cortico-cancellous bone graft are marked by *arrows*, there are reliable signs of bony healing

fusiform or oval and form a storiform pattern. There are singular groups of foam cells and multinuclear giant cells. Neither atypical cells and nuclei nor increased mitotic activity can be found [4, 12, 16].

Despite this uniform histologic pattern, BFH and NOF can be differentiated by clinical evaluation:

BFH afflicts adults between the third and sixth decade, affecting the metaphysis of long bones and in some cases also sacrum, ileum, ribs, and spine [1, 4, 5, 7, 8, 15]. On the other hand, NOF is found only in children and young

adults up to 20 years of age. The tumors are eccentrically located in the metaphyseal area, 96% of all lesions are found in the lower limb [6, 13]. Usually, the main clinical sign of BFH is pain, whereas NOF does not cause any complaint unless a pathologic fracture has occurred [5].

On X-ray BFH presents as a lytic lesion with sclerotic margin without periosteal reaction or soft tissue involvement. There may be also signs of endotumoral ossifications shaped like woven bone. The most important differential diagnosis in adults is giant-cell tumor [1, 2, 4–6, 8, 14].

Although BFH is a biologically non-malignant skeletal tumor, it may show local aggressive growth pattern. In most cases, the lesion heals after surgical treatment such as curettage or excision without recurrence. Only in rare cases have late recurrences been observed [5, 6, 15, 16], which was likely in our own case. On the other hand NOF usually shows spontaneous healing tendency.

There are no reports in the literature of pathologic fractures in cases of BFH. Neither Dahlin and Unni [7] nor Freyschmidt and Ostertag [10] mentioned the occurrence of pathologic fractures in their few cases of BFH. So to our knowledge our case is the first in the literature.

There is no reliable report in the literature of malignant degeneration of BFH. The only paper, written by Bhagwande in 1966, dealt with the case of a 21-year-old male patient and was not able to withstand a detailed examination. This was due to the fact that in all probability there were cells of a malignant fibrous histiocytoma already within the primary tumor [3, 4].

Hastrup and Jensen report a case of osteosarcoma formations within a BFH, which is to be considered as an absolute rarity [11].

Conclusion

BFH presents as a non-malignant skeletal tumor of the adult, the location of which is very variable. Usually surgical intervention is the treatment of choice. Prognosis after resection or curettage usually is good, recurrences are observed only in very rare cases. Usually there is no danger of malignant degeneration. Formations of osteosarcoma within a BFH have to be considered as an absolute rarity.

Besides radiography and conventional tomography, CT and MRI are mandatory for precise evaluation. In cases of a pathologic fracture, such as the one presented in our paper, surgical stabilisation has to be performed anyway.

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