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Research Paper

The first total vertebral involvement of benign fibrous histiocytoma: A case report and literature review

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ABSTRACT

Background: Benign fibrous histiocytoma (BFH) is a rare bone tumor, extremely seldom in the spine.
Methods: We present a 52-year-old patient diagnosed with a BFH in the thoracic spine treated with total en bloc spondylectomy. A review of the published literature was also conducted.
Results: Non-ossifying fibroma (NOF) and BFH are named as one tumor called NOF/BFH. A total of 20 spinal BFHs have been previously reported, mainly involving the posterior elements. We present a BFH with total vertebral involvement. Curettage and excision are the main treatment options with limited recurrence.
Conclusions: This is the first total vertebral BFH up to now. Spinal BFH occupies rather low aggressiveness. With rather limited recurrence and malignant degeneration, surgical interventions seem enough for its management.

1. Introduction

Benign fibrous histiocytoma (BFH) is a rare bone tumor accounting approximately 1% of all benign bone tumors, commonly located in the meta-epiphysis and diaphysis of long bones, mostly in male and adult patients [1]. Spinal involvement is rather rare: to the best of our knowledge, only 19 cases have been previously described since 1979 [1–15]. Among the previous reports, almost all were involved in the posterior elements such as spinous process, lamina, transverse process, while total vertebral involvement was not found. Here, we report the first BFH of total vertebral involvement managed with total en bloc spondylectomy.

2. Case report

A 52-year-old female, complaining thoracic back pain and slight weakness of the lower extremities for 1 month presented to another local hospital. Her preoperative X-rays of the thoracic spine showed a lytic lesion of the T6 vertebral body. Computed tomography of the thoracic spine also indicated osteolytic bone destruction of the T6 vertebra, without involvement of the paravertebra and posterior elements. Furthermore, the magnetic resonance (MR) showed destruction of the posterior vertebral wall and slight compression of the dura. Hypointense was seen on both T1 and T2 sequence, and homogeneous enhancement was observed after the use of Gd-DTPA (Fig. 1). The local hospital carried out a needle biopsy and a bone marrow aspiration, while exact diagnosis was not acquired.

After admitting to our spine tumor center, the positron emission tomography/computed tomography (PET/CT) showed increased concentrations of radioactive material in the single T6 vertebra, which was suspected as a primary malignant tumor of the bone. No other abnormality was found in the clinical blood tests including the blood routine test, tumor markers and M-spike. With careful physical examinations, nearly all her motor functions were normal. Neurological evaluation did not confirm abnormal reflexes. $5^{-}/5$ strength was detected from her lower extremities. She had an unremarkable past medical history.

Based on the information above, total removal of the vertebral lesion with total en bloc spondylectomy was planned to verify the pathological diagnosis, acquire a complete resection and reconstruct the spine. According to the processes of vertebral total en bloc spondylectomy, the posterior elements of T6 were removed and the dura was decompressed first. Then the bilateral dissection of T6 vertebra and excision of the bipolar discs, followed by the total removal of T6 vertebra as a whole. After necessary disposition, the thoracic spine was reconstructed by a mesh cage filled with allogeneic bone and the screws-rods system. Combined with the preoperative images, postoperative pathology indicated a BFH (Fig. 2). the patient recovered uneventfully and was able to walk with the help of a walker 3 weeks after the operation. Until the present follow up of 40 months, the

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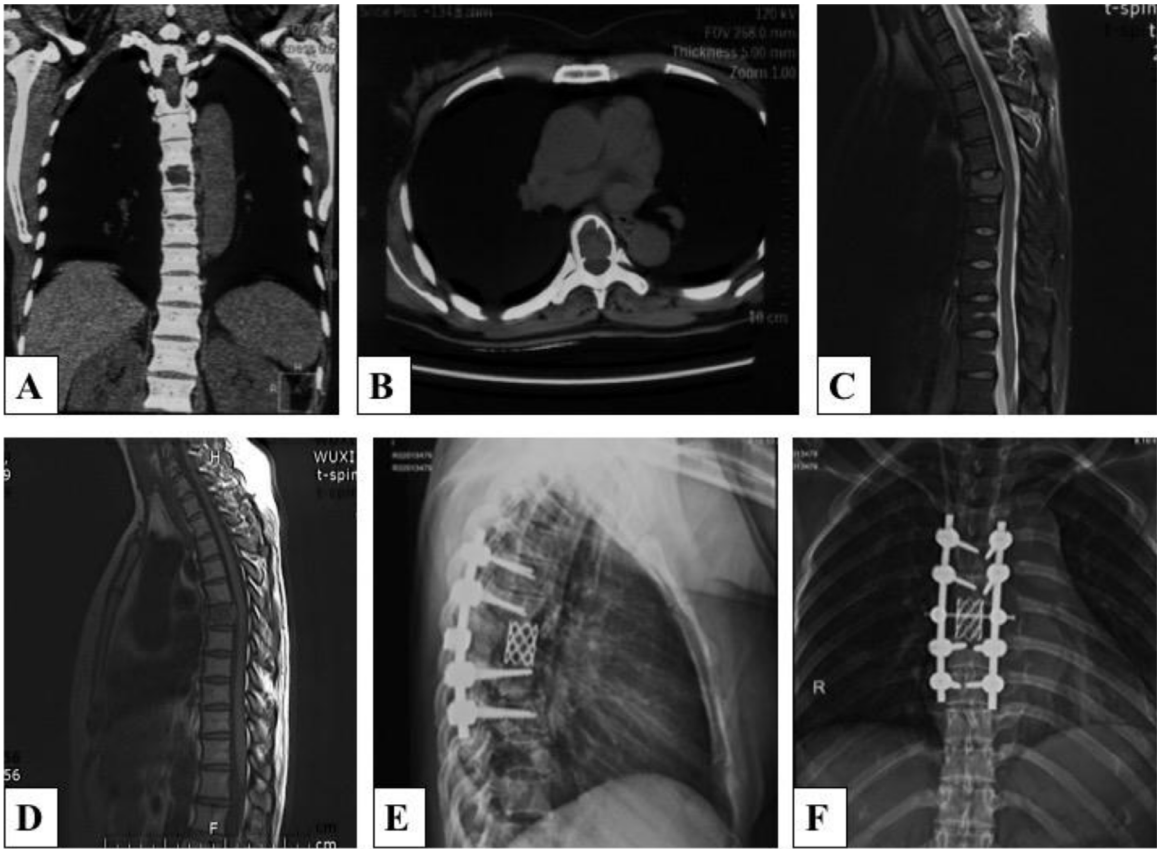

Fig. 1. A, B: Preoperative CT revealed obvious bone destruction of the vertebra with intact posterior elements. **C, D**: Preoperative MRI indicated a compressed fracture of the vertebra and slight compression of the dura. **E, F:** The postoperative X-ray after TES**.**

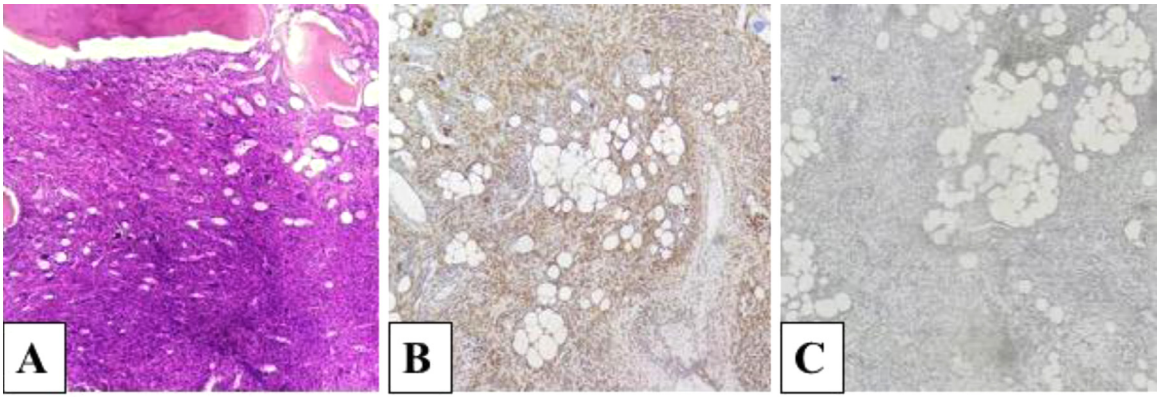

Fig. 2. A: variable amount of characteristic spindle-shaped fibrohistiocytic cells showed a storiform arrangement; multinucleated giant cells also could be seen (HE x200). **B:** plenty of fibroblasts were showed, accompanied by foam cells (HE x200). **C:** actin was negative in immunohistochemistry (IHC x200), indicating no muscular origination.

patient was rather well with no evidence of disease.

3. Discussion

Benign fibrous histiocytoma (BFH) was first described by Dahlin in 1978 [16]. It was controversial all the time in terms of the difference between non-ossifying fibroma (NOF) and BFH [5,17]. According to previous literatures, BFH was thought to afflict adults between the third and sixth decade, affecting the metaphysis of long bones and in some cases also sacrum, ileum, ribs, and spine. NOF is found only in children and young adults up to 20 years of age [3,7]. In all, the differential diagnosis of them was almost microscopically impossible, mainly different in ages of disease onset [9,10]. In 2003, the 3th edition of WHO classification of soft tissue tumors deleted the name of NOF. However, the 4th edition in 2013 classified NOF and BFH as the same tumor named as NOF/BFH since they occupied almost the same histological features [18].

As reported, BFH usually occurs in the lower limbs, pelvis, and humerus but rarely involves the spine. Spinal BFH usually produces non-specific pain or discomfort as a primary presenting symptom, similar to other benign tumors, and may cause neurologic symptoms secondary to nerve roots and cord compression, just as the situation in our case. Skeletal BFH appears as an osteolytic lesion with sharply defined and frequently sclerotic borders without matrix mineralization [1,16]. Histologically, BFH consists of a variable amount of spindle-shaped fibrohistiocytic cells, multinucleated giant cells and foam cells,

Table 1
Summary of the published case reports of spinal BFH

Case	Age/Sex	Symptoms	Imaging examinations	Location	Approach and Treatment	FU (mon)
Guamaschelli et al[2]	14/M	BP	X ray, myelography	T5 extradural	Posterior, piecemeal	6, NOD
Destouet et al[3]	24/M	NP	NA	C-2 posterior elements	Posterior, curettage, graft	39, NOD
Roessner et al[5]	41/M	NP, stiffness	X ray	Part of C-3, 4 vertebra	Combined, piecemeal, graft	3, NOD
Mirra et al[4]	18/M	NP	NA	C-2 spinous process	Posterior, excision	60, NOD
Mirra et al[4]	24/M	NP	NA	C-2 spinous process	Posterior, excision	NA
Mirra et al[4]	28/M	NP	NA	C-6 spinous process	Posterior, excision	59, NOD
Hoeffel et al[6]	13/M	Scoliosis, stiffness	X ray, CT	T-12 left vertebra, pedicle, lamina	Combined, excision, graft	24, NOD
Peicha et al[7]	44/F	NP	X ray, CT, MR, ECT	C-2 odontoid fracture	Anterior, excision, graft	60, possible recurrence
Grohs et al[1]	33/F	Abdominal, NP, LP	X ray	L-3 posterior elements	Excision, graft	65, NOD
Van et al[9]	6/M	NP, restricted rotation	CT, MR, biopsy	C-1 posterior arch	Posterior, marginal resection, hemilaminectomy	12, NOD
Balasubramanian et al[10]	18/F	BP, LP	X ray, MR, biopsy	S1-4 intraspinal and presacral	Posterior, subtotal resection, lumbopelvic stabilization	5, residual tumor
Kuruvath et al[11]	24/M	BP	MR	T-3 posterior elements	Posterior, T-3 laminectomy, vertebrectomy, cage + screw-rod fixation	30, NOD
Morales et al[14]	16/F	BP, leg weakness	CT, MR	L4, 5 posterior elements and paravertebral	Posterior, complete resection, cage+screw-rod fixation	NA
Demiralp et al[4]	21/M	BP, LP	X ray, CT, MR, biopsy	L-2 posterior elements	Posterior, curettage, graft, screw-rod fixation	72, NOD
Kim et al[12]	66/F	Severe claudication	MR	L-5 intraspinal, extradural	Posterior, total tumor excision, screw-rod fixation	6, NOD
Khor et al[13]	65/M	Fever, myalgia	PET/CT, MR, biopsy	T-8 left pedicle	Posterior, curettage	NA
Skunda et al[8]	14/M	NP after trauma	CT, MR, MRA	C-2 left transverse foramen	Anterior, curettage	NA
Donati et al[15]	13/M	BP, LP	X ray, CT, MR, ECT, biopsy	L4-S2 intraspinal,extradural	Posterior, complete resection, screw-rod fixation	12, NOD
Present case	52/F	BP, leg weakness	X ray, CT, MR, PET/CT, biopsy	Total T6 vertebra	Posterior, TES, mesh cage + screw-rod fixation	40, NOD

Note: BP: back pain, LP: leg pain, NP: neck pain, CT: computed tomography, MR: magnetic resonance, MRA: magnetic resonance angiography, ECT: emission computed tomography, PET/CT: positron emission tomography/computed tomography, NOD: no evidence of disease.

showing a storiform arrangement [16]. BFH shares clinical, radiological, and histological features with other benign lesions such as NOF, giant cell tumor (GCT), fibrous dysplasia, aneurysmal bone cyst (ABC), osteoblastoma (OB), and eosinophilic granuloma (EG), making its diagnosis challenging [10]. Usually, though imaging examinations such as X ray, CT, MR, ECT, and PET/CT were commonly employed, biopsy is often inevitable for preoperative diagnosis.

According to the English literature, only one case of spinal NOF [17] and 19 cases of spinal BFH were reported up to now. To learn about the clinical features of spinal BFHs, we reviewed the spinal BFHs in English literatures. The epidemiologic features, locations, treatment methods and prognosis were showed in Table 1. Age of the reported spinal BFHs ranged between 6 and 66 years, with a mean age of 28.11 years. A female predominance existed in spinal BFHs (F/M: 13/6), quite different from that in the total BFHs (F/M: 1/2) [12]. Learning from the reported cases, local pain was the cardinal symptom when admission. Usually, duration of pain ranged from weeks to several years. Cervical spine was the most frequent place of spinal BFHs (n = 8), followed by the thoracic spine and lumbar spine (n = 5 respectively). Another obvious feature was that nearly all the lesions involved the posterior elements of vertebra, while total vertebral involvement without involvement of posterior elements was not reported up to now. We believe that this is the first case of that.

Due to the characteristics above, the main treatment mode was curettage or complete resection through the posterior approach, followed by bone graft or feasible stabilizations. In our present case, the tumor resection and spine reconstruction mode were greatly different from previous cases. The total en bloc spondylectomy, which was recommended as the best option for primary malignant tumor, solitary metastasis and benign aggressive tumor in spine [19], was adopted to acquire the total removal of this suspected malignant bone tumor. For reliable reconstruction after tumor resection, a mesh cage and the screws-rods internal fixation system were employed, providing immediate segmental stability. Postoperative follow up was carried out routinely and no recurrence was discovered with complete neurological recovery at the last follow up of 40 months. As the name of BFH, it is a benign bone tumor occupying an excellent prognosis after tumor resection. Recurrence and metastasis were observed only in rare cases [1,3]. In the present 19 BFHs, only one case (case 8) of possible recurrence was reported during the mean follow up of 32.87 months (4 were not available). And there is no evidence in the literature to support the role of radiotherapy in the treatment of BFH.

4. Conclusion

NOF and BFH are now classified as the same tumor named as NOF/BFH, a very rare benign bone tumor. Surgical interventions seem enough for the management of spinal BFHs with rather limited recurrence and malignant degeneration. To the best of our knowledge, our report is the first BFH with total vertebral involvement and treated with TES. Treatment with en bloc resection and reconstruction of double columns showed good clinical and radiological results.

Ethics approval

The study was approved by the Ethics Committee of Changzheng Hospital.

Consent for publication

Written informed consent was obtained from the patient for publication of his clinical details and/or clinical images. A copy of the consent form is available for review.

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Authors' contributions

JRX and XHY conceived and designed the study. JY and JJ drafted the manuscript. JBH reviewed and edited the manuscript. All authors provided intellectual input to the study and approved the final version of the manuscript.

Availability of data and materials

All data generated or analyzed in this study are included in the article.

Declaration of competing interest

The authors declare that they have no competing interests.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.jbo.2019.100274.

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