Research Paper

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

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A R T I C L E   I N F O

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A B S T R A C T

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 paravertebral 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...
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 3rd 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

| Case          | Age | Sex | Symptoms          | Imaging examinations | Location                          | Approach and Treatment                                                                 |
|---------------|-----|-----|-------------------|----------------------|-----------------------------------|----------------------------------------------------------------------------------------|
| Guarnaschelli et al | 14  | M   | BP, neck pain     | X-ray, myelography   | T5 extradural                      | Posterior, piecemeal                                                                    |
| Destouet et al  | 24  | M   | NP                | NA                   | C-2 posterior elements             | Posterior, curettage, graft                                                            |
| Roessner et al  | 41  | M   | NP, stiffness     | X-ray                | Part of C-3, 4 vertebra            | Combined, piecemeal, graft                                                             |
| Mirra et al    | 24  | M   | NP                | NA                   | C-2 spinous process                | Posterior, excision, graft                                                             |
| Mirra et al    | 28  | M   | NP                | NA                   | C-6 spinous process                | Posterior, excision, graft                                                             |
| Hoeff         |     |     | BP, LP            | X-ray, CT            | T-12 left vertebra, pedicle, lamina| Combined, excision, graft, biopsy                                                     |
| Peicha et al   | 44  | F   | NP                | X-ray, CT            | S1-4 infraspinal and presacral     | Anterior, excision, graft, screw-rod fixation                                           |
| Van et al      | 6   | M   | NP, restricted rotation | CT, MR, biopsy      | C-1 posterior arch                  | Posterior, marginal resection, hemilaminectomy                                         |
| Balasubramanian et al | 18  | F   | BP, LP            | N/A                  | S1-4 vertebra, pedicle             | Posterior, subtotal resection, lumbopelvic stabilization                                |
| Morales et al  | 16  | F   | BP, leg weakness  | CT, MR, ECT, biopsy  | L4, 5 posterior elements and paravertebral | Posterior, curettage, complete resection, screw-rod fixation                           |
| Khor et al     | 65  | M   | Fever, myalgia    | PET/CT, MR, biopsy   | T-8 left pedicle                   | Anterior, curettage, complete resection, screw-rod fixation                           |
| Leng et al     |     |     | BP, leg weakness  | X-ray, CT            | L-1 left transverse fracture        | Anterior, TES, mesh cage + screw-rod fixation                                           |
| Kim et al      | 14  | M   | Fever, neck pain  | CT, MR, biopsy       | T-5 left pedicle                   | Anterior, TES, mesh cage + screw-rod fixation                                           |
| Khan et al     |     |     | BP, leg weakness  | X-ray, CT            | T-5 left pedicle                   | Anterior, TES, mesh cage + screw-rod fixation                                           |
| Kim et al      | 12  | M   | BP, LP            | X-ray, CT            | L-6 left transverse fracture        | Anterior, TES, mesh cage + screw-rod fixation                                           |
| Present case   | 52  | F   | BP, leg weakness  | X-ray, CT            | T5 vertebra                        | Anterior, TES, mesh cage + screw-rod fixation                                           |

Note: BP: back pain, LP: leg pain, NP: neck pain, CT: computed tomography, MR: magnetic resonance, ECT: emission computed tomography, PET/CT: positron emission tomography/computed tomography, TES: total en bloc spondylectomy, NOF: no evidence of disease.

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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