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Traumatic Vertebral Fracture with Coexisting Enchondroma: a Case-Report and Review of the Literature

Valsecchi D^{1*}, Ackfeld T¹, Gondar R¹, El Rahal A¹, Stauffer E², Otten P^{1,3} and Maestretti G¹

¹Spine Unit, Department of Orthopedic Surgery, Hôpital Fribourgeois - HFR, University of Fribourg, Fribourg, Switzerland ²Units of Hysto-Cytopathology, Promed Laboratoire Medical, Fribourg, Switzerland

³Department of Neurosurgery, "Ste-Anne" Clinic, Fribourg, Switzerland

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*Corresponding author: Daniele Valsecchi, Spine Unit, Department of Orthopaedic Surgery, Hôpital Fribourgeois -HFR, Chemin des Pensionnats 2-6, 1700 Fribourg, Switzerland. Tel: +41-26-306-28-04; E-mail: daniele.valsecchi@h-fr.ch

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Abstract

Enchondroma of the vertebral spine is a rare clinical entity. For symptomatic enchondroma, surgical resection is the treatment of choice. Here we present the atypical case of a patient with non-symptomatic spinal enchondroma (D9), discovered because of a dorsal trauma that led to a D9 fracture. The pre-operative diagnostic workup gave a suspicion of a co-existing neoplastic lesion, leading to the consideration of a more likely pathologic fracture than a traumatic lesion, with no segmental instability. Therefore, kyphoplasty with Vertebral Body Stenting System and intraoperative biopsy were performed, with evidence of the unusual pathologic lesion. The postoperative evolution showed a complete recovery. After multidisciplinary oncologic discussion, we decided to perform a strict follow-up with MRI at 3 and 12 months, instead of performing a vertebrectomy. At 1-year followup, the patient presented no clinical symptoms, and no evidence of lesion recurrence was assessed at the imaging. Though not the recommended treatment choice, kyphoplasty seems to be a potential treatment option for a traumatic fracture superposing on non-previously detected and asymptomatic enchondroma.

Abbreviations

VBS: Vertebral Body Stenting; MRI: Magnetic Resonance Imaging; BKP: Balloon Kyphoplasty.

Introduction

The vertebral column's primary tumors are rare, counting only for 10% of primary bone tumors [1,2]. Enchondroma of the vertebral spine is even rarer, with only a few cases reported in the literature. Enchondroma is a benign bone tumor that belongs to the overarching group of chondromas, a slow-growing tumor producing hyaline cartilage [3]. Enchondroma are tumors of the medullary cavity that are the result of the continued growth of residual hyaline cartilage rests, displaced from the growth plate [4,5]. These tumors count for about 5%–10% of the bone tumors and are most commonly located in hands and feet and only rarely origin from the vertebral column [5,6].

Enchondroma are usually asymptomatic lesions discovered as incidental findings on routine radiological studies, and simple follow-up is indicated [7]. For spinal enchondroma and when symptomatic, patients usually present with pathologic fracture or radiating pain and require surgical treatment. Treatment of choice, in this case, is complete surgical resection [3].

Although enchondroma is benign cartilaginous lesions, sarcomatous degeneration rarely occurs—approximately 4%

of enchondroma change into secondary chondrosarcoma [8]. Histopathological and radiological differentiation between enchondroma and chondrosarcoma is challenging, and close follow-up is necessary [9].

When treating patients with spinal enchondroma, little literature is available. Entering an even bigger dilemma, when treating a patient with a traumatic fracture on an asymptomatic enchondroma, no literature is available. Here we report an asymptomatic patient who sustained a D9 vertebral fracture with adequate trauma and had the incidental finding of a co-existing enchondroma.

Materials and Methods

An electronic search in the "Pubmed" database from 1966 to 2020 was performed. The following term was used in the search strategies: "enchondroma" and "spine" in title and abstract. All articles in the English language were scanned. Inclusion criteria were clear histopathological differentiation between periosteal chondroma and medullary enchondroma. Exclusion criteria were articles containing the overarching group chondroma without

differentiating between periosteal chondroma and medullary enchondroma, articles written in any other language than English, and literature where no original article could be found. Moreover, tables in other publications were scanned for enchondrome in vertebral spine and were included if the primary source was available. They were not included in the original article was not available on "Pubmed." The result of our literature research is listed in (Table 1). Case-series, cohort studies, case-control studies, and case reports were included in our research, but only case reports were found.

Literature review: Five cases of enchondroma in the cervical spine, [17,18,19,20,21] two cases of enchondroma in the thoracic spine, [6,21] and one enchondroma case in the lumbar spine [6,21] were described. All symptomatic patients, either with pain, neurological deficit, or pathological fracture, were treated with surgical resection. Neither cases of traumatic fracture on enchondroma, nor cases of traumatic fracture on chondroma were described. The outcome in all patients was satisfying.

Table 1: Pubmed was searched for "enchondroma" and "spine" in title and abstract. All articles in the English language were articles were scanned for cases where histopathological examinations could clearly reveal enchondroma.

Literature	Localization	Clinical features	Treatment	Outcome
Jeong & Paeng, 2015	C4	24-year-old woman	C4 right hemilaminectomy and	At the 6-month: no recurrence,
[17]		Pain, muscle weakness, sensory	facetectomy.	no pain, and her numbness and
		deficit.		motor weakness were improved.
				no neurological symptoms.
				At 3-year no recurrence.
Willis & Heilbrun, 1986	C4	24-year-old woman	Block excision of C4 and partial	At 1-year: no recurrence complete
[18]		upper extremity paresthesia and	excision of C3 and C5 via ant.	resolution of neurological
		weakness.	approach + iliac crest bone graft	symptoms and pain.
		C4 fracture after minor trauma	+ halo 3-month.	
		Bony destruction of C4 with		
		extension to C3 and C5.		
Shurland et al. 1999 [19]	C5	11-year-old girl Pain in neck ROM,	C5 left hemilaminectomy.	Follow-up at x weeks: returned to
		no neurological deficit.		full activity.
Wani et al. 2011 [20]	C1	25-year-old male progressive	Complete tumor resection via	At 9-month: significant
		onset of quadriparesis for the	posterior midline approach.	improvement of symptoms, but
		past 8-month.		spasticity still present.
Guo et al. 2017 [6]	Т3	Rapidly progressive and severe	Complete surgical resection with	At 1-year: no recurrence.
		pain on upper back	via a posterior approach: T3 + T4	
		no motor deficit, mild sensory	lamina and spinous process +	
		loss distal to the right ankle.	posterior pedicle screw fixation.	
McCarthy et al. 2015	C5, C6, Th5,	39-year-old man	Anterior cervical decompression	At 8-month: amelioration upper
[21]	Th6	Ataxic gate M5/5 upper motor	and fusion with C5-C6 partial	and lower extremity.
		testing, diminished lower motor	corpectomy.	
		testing.	Decompression and partial	
			vertebrectomy via posterior and	
			lateral approaches.	
Nag & Falconer, 1966	L4	50-year-old man	Laminectomy L3,L4,L5 + partial	At 2-year: symptom free.
[22]		Bilaterale sciatica.	removal L4.	

Case Presentation

History and Examination: A 39-year-old healthy male patient presented at our emergency department with acute thoracic back pain after a skiing accident. The patient sustained a fall at a moderate to fast speed with an unknown mechanism. At the hospital admission time, the patient presented a progressive pain over the lower thoracic spine. The neurological examination was normal. A CT study revealed a burst fracture of D9, classified as A4, N0 and M0, according to the AOSpine classification for thoracolumbar traumatic fracture (Figure 1A). We decided for hospitalization and further investigation with MRI, in order to check for a tension band's lesion or collateral vertebral fractures, not detected at the CT-scan. Surprisingly, the MRI showed, only on D9, an atypical very homogenous hyperintensity of the vertebral body and pedicles, in STIR rated images (Figure 1B). This was more suspicious for a neoplastic than for a traumatic lesion. Furthermore, no signs of anterior and posterior tension band lesions were found. Not looking for a neoplastic lesion, the imaging was performed without contrast-enhancing. Primary radiological differential diagnosis included a traumatic fracture with an atypical STIR hyperintensity pattern and a traumatic fracture on a co-existing bone tumor. A thoracoabdominal CT-scan did not find a primary lesion, and a metastatic spinal lesion was unlikely.

Due to the traumatic fracture pattern, without the tension band's involvement, but with the co-existing suspicious primary lesion, surgical treatment was discussed. The aim was to stabilize the anterior column and give anterior support, as in pathologic pure bony fractures, without pedicle screws fixation. Therefore, a BKP with VBS and cementation of D9 was performed. Due to suspicious pre-operative imaging, and intraoperative biopsy of the trabecular bone was associated. Free mobilization was allowed post-operatively. Post-operative standing thoracic spine x-rays were obtained and showed a satisfying result (Figure 2A). Postoperative pain was managed with first step analgesics, and the patient could be sent home on the second post-operative day.

Pathological findings: The histopathological results suggested a fracture zone in the context of a chondrogenic tumor (enchondroma). The microscopic examination of the specimen revealed a cartilaginous tumor between bone spans without any sign of the destruction of the underlying tissue (Figure 3). No atypical cells or malignant nuclei were found, and immunohistochemically examinations showed no malignancy (Figure 5).

Post-operative course: Clinical follow-up took place at three weeks, six weeks, three months, six months, and one year. Follow-up did not show recurrence or complication. The patient has gotten back to manual labor, and his normal physical activity level without pain or restriction (Figure 4).

The radiological workup (MRI + contrast agent and X-ray), done at 3 and 12 months, did not show any cartilaginous lesions or operation-related complications (Figure 2B, Figure 4).



Figure 1: A: Preoperative CT-scan (bone window) showing an A4 burst fracture of D9; B: Preoperative MRI, with STIR sequences showing a traumatic D9 fracture with a very homogeneous pattern of hyperintensity, without anterior or posterior tension band lesion.



Figure 2: A: Postoperative X-ray, standing position, coronal and lateral projections. The procedure of Vertebral Body Stenting (VBS) was performed through percutaneous approach. Intraoperative biopsy was preliminary obtained, via intra-vertebral carottage over the right pedicle, and then 22 mm stenting cannulas were introduced bilaterally via wires. Two balloons were inflated until 22 bar, without bone fragment's displacement. We cemented each balloon with 3.5 cc of polymethylmethacrylate. B: standing X-ray, lateral projection, after 12 months, without morphological modification.



Figure 3: MRI at 1-year follow-up. A: sagittal STIR sequences, showing no lesion recurrence and B: axial T1-weighted contrast-enhanced sequences, with postoperative but non-pathological intervertebral enhancement.



Figure 4: A and B: After 12 months follow-up, the patient showed a good ROM of the spine. VAS was assessed as 0/10.



Figure 5: Photomicrographs showing histopathological findings. Predominant cartilaginous pattern between bone spans in the tumor specimen from the primary surgery. A: scale bar 500 µm, B: scale bar 200 µm, C: scale bar 50 µm, D: scale bar 20 µm.

Discussion

Dealing with vertebral fractures is challenging, especially when the fracture's origin is not well defined, and an underlying malignancy is suspected. Here the dilemma was to distinguish between a pathological and a traumatic fracture in a patient with the first hypothesis of a vertebral tumor of unknown origin at the time of operation.

Pathologic fractures are defined as fractures in a pre-existent pathological bone that occur without adequate trauma. Frequent causes are osteoporosis, bone destruction due to tumorous growths, osteomalacia, osteonecrosis, augmented bone resorption, insufficient bone production, or pathological bone remodeling [10].

According to the patient's age and expected bone quality, our patient sustained a vertebral fracture in the context of a kinematicadequate accident. Respectively, our first and main aim was to treat the traumatic lesion. According to the radiological findings, we have also considered the lesion's co-existing likely pathologic nature, and anterior column reinforcement was necessary. The fracture was treated with VBS, leaving the opportunity to followup the lesion by MRI and eventually perform radiotherapy [11].

At the time of operation, the origin of the tumor was still unknown. According to the poor data in the literature, we discussed the need to perform a vertebrectomy when receiving the histopathological results. In the aim to avoid an eventual overtreatment, in a patient with complete recovery and no residual symptoms, the case was discussed during a multidisciplinary oncological board, and Oncologists gave an independent clinical and radiological assessment. At the end of this workup, a regular follow-up was decided.

Treatment of choice for symptomatic vertebral enchondroma is complete surgical resection, by vertebrectomy and pedicle screws spine fixation, for the amelioration of symptoms and minimizing the risk of sarcomatous degeneration [3,6]. Radiotherapy is contraindicated while causing radiation myelitis and being ineffective [12].

Minimally invasive procedures such as Balloon Kyphoplasty (BKP) and Percutaneous Vertebroplasty (PV) were not described for enchondroma spinal lesions but are an accepted treatment option for traumatic fractures and pathologic spine fractures [11]. Several studies have shown advantages in treating pathologic vertebral spine fractures, e.g., in Multiple Myeloma, when spinal fixation contains a higher risk for material infection due to immunosuppression [13,14].

The VBS is a minimally invasive technique with a low complication rate and an adequate improvement in function, pain, and deformation of osteoporotic and traumatic fractures [14,15]. Additionally, the bone cement filling may have a beneficial effect on recurrence rates due to heat destruction, as shown for intramedullary chondrosarcoma of long bones [16].

Conclusion

Traumatic fracture on pre-damaged pathologic bone, in this case, seems to behave like any other traumatic fracture, and treatment of a traumatic fracture accordingly is possible. At 1-year follow-up, our patient is active, has no symptoms, and no radiological tumor progression. Moreover, when seeing a young patient with back pain and suspicious MRI findings, with or without any given trauma, a vertebral biopsy is highly recommended and, although rare, an enchondroma should always be considered differential diagnosis.

Though not the recommended treatment choice, BKP with VBS seems to be an option for a traumatic fracture on co-existing spinal enchondroma, when incidentally discovered in a non-symptomatic patient; nevertheless, a treatment by vertebrectomy should be considered and discussed case by case in a multidisciplinary oncologic team.

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