Chondromyxoid fibroma of the lumbar spine: case report and literature review

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Abstract

Purpose Chondromyxoid fibroma (CMF) is a benign tumour of the bone that typically occurs in long bone metaphysis. Spinal involvement is uncommon, but more frequent in the cervical and thoracic segments. Lumbar involvement is extremely rare. We report the ninth case of lumbar CMF and the first one involving the articular process of the vertebra. A review of the literature is also intended making special emphasis on the differential diagnosis with other benign spinal tumours of the bone.

Methods A 21-year-old Caucasian male suffering from low back pain that increased with sports and interrupted sleep was diagnosed with a tumoural lesion in the right inferior articular process of L5.

Results Complete surgical excision of the tumour was accomplished. Histological diagnosis confirmed a CMF. The patient remains asymptomatic at 1-year follow-up.

Conclusion Despite the low incidence of CMF in the lumbar spine, differential diagnosis must include this subtype of lesion among other benign tumours of the bone and cartilage. Histological diagnosis is essential in order to provide the patient with an accurate management of the pathology. Recurrence rate is to be considered even in the case of complete surgical excision. Radiotherapy

administration is controversial due to suspicion of malignant transformation of the tumour.

Keywords Chondromyxoid fibroma · Lumbar · Spine · Treatment

Introduction

Chondromyxoid fibroma (CMF) is a benign tumour of the bone that typically occurs in the metaphysis of long bones. Spinal involvement is uncommon, but more frequent in the cervical and thoracic segments. Lumbar involvement is extremely rare. To our knowledge, fewer than 50 cases of spinal CMF have been reported in the literature to date. Among them, only eight cases involve the lumbar spine [1–6].

In this article, we report the exceptional case of a patient diagnosed with a benign tumour of the right inferior articular process of L5 that was successfully treated and diagnosed as a CMF. This is the first CMF of a lumbar articular process described in the literature to date. Besides that, we review the literature and make special emphasis on the differential diagnosis with other benign spinal tumours of the bone.

Case report

A 21-year-old Caucasian male was attended at our centre referring low back pain when playing sports. The symptoms had become more intense after suffering a traffic accident and the pain started to interrupt the sleep. No neurological deficit was evidenced in the physical exam. Plain X-ray showed a lytic lesion in the right inferior

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articular process of L5. A magnetic resonance imaging of the lumbar spine revealed a bone lesion in the inferior right facet of the fifth lumbar vertebra associated with muscular oedema. A computed tomography of the lumbar spine confirmed the presence of a lytic lesion of the bone, 18 mm in size and with lobulated margins that contained a thin central calcification (Fig. 1).

Complete en bloc excision of the tumour was accomplished through a posterior lumbar approach under general anaesthesia and in the prone position. A wide bilateral exposure of the posterior elements of L5 and S1 was achieved. The right inferior articular process of L5 was completely removed in one piece. Surrounding soft tissue was also excised for histological analysis purpose. Vertebral fusion was not performed since the procedure was rejected by the patient. Histological diagnosis confirmed a CMF without malignancy features that did not invade surrounding soft tissues (Fig. 2). The patient remains asymptomatic and no evidence of recurrence or lumbar instability has been observed after 1-year follow-up.

Discussion

Primary bone tumours of the spine account for less than 5% of all bone tumours. Benign tumours comprise 23% of them [7]. Patients can be asymptomatic although most of them may present with local pain. Neurological deficit depends on the extension of the lesion. The radiological features that suggest a benign behaviour of the lesion include: well-circumscribed lesion with well-defined and sclerotic margins, homogeneous architecture, absence of periosteal reaction, homogeneous distribution of calcifications and minimal radiological change on follow-up imaging studies [8]. Differential diagnosis includes osteogenic tumours (osteoid osteoma, osteoblastoma), cartilaginous tumours (enchondroma, osteochondroma), vascular tumours (hemangioma, angioliopoma), non-neoplastic reactive lesions (aneurismatic

bone cyst) and other benign lesions (eosinophilic granulomas, giant cell tumour). Malignant tumours (multiple myeloma, metastasis, chondrosarcoma, osteoblastoma and chordoma) must also be considered in certain cases. Table 1 comprises differential diagnosis among primary benign tumours of the spine [8, 9].

Chondromyxoid fibromas account for less than 0.5% of all bone tumours. First described in 1948 [10], they are commonly found in long bone metaphysis (femur, tibia and fibula) and exceptionally located in the spine. Most of spinal CMF reported in the literature are located in the thoracic spine rather than in the cervical and lumbar segments, a distribution that can be compared with the classic one of other spinal neoplasms. So far, eight cases of CMF of the lumbar spine have been reported in the literature. However, three of them are not well-described since they are part of a larger series (Table 2) [1–6]. This fact confirms the exceptional case hereby described. The low incidence of this kind of tumours explains the low level of suspicion frequently raised among radiologists. Thus, histological diagnosis is essential in order to provide the patient with an accurate management of the pathology. The classic histopathological features of CMF include stellate and spindle-shaped cells arranged in a lobulated growth pattern matrix with mixed chondroid, myxoid and fibrous areas. The cellular areas contain plump mononuclear cells resembling chondroblasts and multinucleated giant cells may also be present. Stellate and spindle cells predominate in the less cellular areas. Foci of calcification are not usually a prominent feature unless patients are older or the lesions are on or near the surface of the bones. Cellularity increases towards the periphery of the lobules, and there may be striking atypia, which has no clinical significance with regard to recurrence or malignant transformation [11].

The management of spinal CMF is derived from the experience on long bones' lesions. Thus, complete en bloc excision must be achieved whenever feasible, whereas

Fig. 1 Plain lumbar X-ray, anteroposterior view showing a lytic lesion in the right inferior articular process of L5 (*left*). Computed tomography of the lumbar spine, coronal (*centre*) and axial (*right*)

views showing a lobulated and lytic lesion in the right inferior articular process of L5, with eroded but intact cortex

Table 1 Differential diagnosis of primary benign tumours of the spine

Tumour	% in spine	Age (year)/sex	Spine segment	Column	Symptoms	CT	MRI	Treatment	Results
Osteoid osteoma	10–25	10–20/M	Lumbar (59%) Cervical (27%) Thoracic (12%)	Posterior (93%) > anterior (7%)	Back pain; night; relief with salicylates	Radiolucent nidus, Ca^{2+} , sclerosis (<1 cm)	Heterogeneous in T1 and T2	Surgery (excision, curettage)	Cure
Osteoblastoma	40	10–20/M	Cervical = thoracic = lumbar	Posterior > anterior	Back pain, torticollis, scoliosis	Round, sclerosis (>1 cm)	Heterogeneous (Ca^{2+} , haemorrhage)	Surgery; RT resistance	Recurrence if incomplete resection
Enchondroma	1	10–30/M	–	Anterior > posterior	Asymptomatic; Ollier's syndrome, Maffucci's syndrome	Ca^{2+} , rings and arcs	Assess extension	Surgery if symptomatic	
Osteochondroma	3–7	20–30/M	Cervical and high thoracic	Posterior	Asymptomatic; back pain	Ligamentous insertion, Ca^{2+}	Hyperintensity (cartilage) hypointensity (Ca^{2+})	Surgery if symptoms or diagnosis needed	Cure. Low recurrence. Malignization 1–5%
Hemangioma	30–50	40–60/F	Thoracic	Anterior	Asymptomatic; back pain; myelopathy	Vertical trabeculae, polka-dot pattern	Hyperintense in T1 and T2 (fat)	Embolization, surgery, RT (30–40 Gy), vertebroplasty	
Angiolipoma	Rare	–	–	Anterior, epidural	Myelopathy, radicular pain and deficit	Fat and soft tissue	Fat and soft tissue	Surgery	Cure
Aneurysmal bone cyst	20	0–20/F	Cervical and thoracic	Posterior (60%) > anterior (40%)	Back pain, radicular pain, myelopathy	Bone lysis (2–9 cm)	Trabeculae, haemorrhage	Aggressive surgery; preoperative embolization	Recurrence 20–30% if incomplete resection
Eosinophilic granulomas	10–15	0–20/M	Thoracic and lumbar	Anterior	Myelopathy, cifosis, fever, weight loss	Bone lysis, no sclerosis	Hypointense in T1, hyperintense in T2	Conservative; surgery; chemotherapy if multifocal	Poor prognosis in >2-year old and hepatic or bone marrow lesions
Giant cell tumour	20	20–30/F	Sacral	Anterior	Radicular pain	Lytic lesion, bone expansion, internal septa	Bone and soft tissue components	Aggressive surgery	Invasive, high recurrence
Chondromyxoid fibroma	7	20–30/M ~ F	Thoracic	Posterior	Asymptomatic, pain	Sclerosis, Ca^{2+} , trabeculae	Heterogeneous	Surgery	Recurrence 11%; malignization after RT

CT computed tomography, MRI magnetic resonance imaging, Ca^{2+} calcification

Fig. 2 Histopathological photographs (*left* H&E $\times 20$; *centre* H&E $\times 40$; *right* H&E $\times 60$) showing a partially lobulated growth pattern matrix which contains mixed areas of chondroid, fibrous and connective differentiation. Strands of spindle and stellate cells are

present and mixed with strands of chondrocytes. Hypocellular myxoid matrix is bounded by hypercellular septae. Mitosis or atypia are non-existent

Table 2 Reported cases of chondromyxoid fibroma of the lumbar spine

References	Age (years)/sex	Location	Column	Symptoms	Treatment	Follow-up (months)
Gudscha [1]	23/F	L3	Anterior (body)	Back pain; leg weakness	Excision	36, no recurrence
Tsuji et al. [2]	9/M	L4	Posterior (lamina, pedicle)	Back pain	Excision + bone graft	44, no recurrence
Mayer [3]	23/M	L2	Posterior (spinous process)	Asymptomatic	Biopsy	–
Cabral et al. [4]	19/F	L1–L2	Posterior (pedicle, transverse process)	Leg pain	Extensive excision	60, recurrence
Wu et al. [5]	–	three cases, lumbar	–	–	Curettage or excision	–
Saldua et al. [6]	8/M	L3	Posterior (pedicle)	Back pain	Biopsy; excision + fusion	13, no recurrence
Present case	21/M	L5	Posterior (inferior articular process)	Back pain	Excision	12, no recurrence

vertebral fusion should be carried out in case tumour resection involves spinal instability. Curettage is recommended when complete excision is not possible [11]. In such cases, bone grafting has been proven to reduce recurrence rate, what makes this approach highly recommended [11, 12]. In the case hereby reported, vertebral fusion was rejected by the patient himself due to labour matters.

Despite the benign behaviour of CMF, recurrence rate has been reported to be as high as 11%, mainly during the first year following initial surgery [5]. Nevertheless, the rate increases up to 80% when surgical excision is incomplete [12]. The administration of radiotherapy is controversial, since it has been associated with an increased risk of malignant transformation in spite of the absence of confirmed cases involving the spine [13].

Conclusions

Despite the low incidence of CMF in the lumbar spine, differential diagnosis must include this subtype of lesions among other benign tumours of the bone and cartilage.

Histological diagnosis is essential in order to provide the patient with an accurate management of the pathology. Complete en bloc excision is the treatment of choice. Recurrence rate is to be considered even in case of complete surgical resection. Thus, close follow-up must be warranted. Radiotherapy administration is controversial due to suspicion of malignant transformation of the tumour.

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Conflict of interest None of the authors has any potential conflict of interest.

References

1. Gudscha A (1968) A case of chondromyxoid fibroma of the spine. *Ortop Traum Protez* 29:50–52
2. Tsuji H, Otsuka Y, Tamaki T et al (1975) Chondromyxoid fibroma of the vertebra. Report of a case. *J Jpn Orthop Assoc* 49:305–312
3. Mayer BS (1978) Chondromyxoid fibroma of lumbar spine. *J Can Assoc Radiol* 29:271–272

4. Cabral CE, Romano S, Guedes P et al (1997) Chondromyxoid fibroma of the lumbar spine. *Skeletal Radiol* 26:488–492
5. Wu CT, Inwards CY, O’Laughlin S et al (1998) Chondromyxoid fibroma of bone: a clinicopathologic review of 278 cases. *Hum Pathol* 29:438–446
6. Saldua NS, Riccio AI, Cassidy JA (2008) Chondromyxoid fibroma of the lumbar spine in a pediatric patient. *Orthopedics* 31:610
7. Kelley SP, Ashford RU, Rao AS et al (2007) Primary bone tumours of the spine: a 42-year survey from the Leeds Regional Bone Tumour Registry. *Eur Spine J* 16:405–409
8. Erlemann R (2006) Imaging and differential diagnosis of primary bone tumors and tumor-like lesions of the spine. *Eur J Radiol* 58:48–67
9. Boto GR (2007) Benign bony tumours of the spine. In: de Campos JM, Vaquero J (eds) *Tumors of the spine and spinal cord*. Editorial Universitaria Ramón Arces, Madrid, pp 117–130
10. Jaffe HL, Lichtenstein L (1948) Chondromyxoid fibroma of bone: a distinctive benign tumor likely to be mistaken especially for chondrosarcoma. *Arch Pathol* 45:541–551
11. Schajowicz F, Gallardo H (1971) Chondromyxoid fibroma of bone. A clinico-pathological study of thirty-two cases. *J Bone Joint Surg Br* 53:198–216
12. Gherlinzoni F, Rock M, Picci P (1983) Chondromyxoid fibroma. The experience at the Instituto Ortopedico Rizzoli. *J Bone Joint Surg Am* 65:198–204
13. Rahimi A, Beabout J, Ivins J et al (1972) Chondromyxoid fibroma: a clinicopathologic study of 76 cases. *Cancer* 30:726–736