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Case Report

Chondromyxoid Fibroma of The Lumbar Spine: Case Report and Review of The Literature

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Summary

Chondromyxoid fibroma originating from axial skeleton is a very rare benign bone neoplasm. There are a few reported lumbar cases in the literature and it can be pre-operatively misdiagnosed as other tumors of spine. A 20-year-old girl presented to our outpatient clinic with right hip and leg pain of 3 months' duration. Physical and neurological examinations were unremarkable and laboratory test results were within normal limits. Imaging studies revealed a cystic lesion in the right half of the L5 vertebra and extending to the posterior elements of the vertebra. CT-guided biopsy result was inconclusive. Surgery was planned. Digital subtraction angiography and embolization were carried out preoperatively. During surgery, L5 laminectomy and curettage of the lesion were performed, taking care to leave the cortex intact. Bilateral L4, S1, and left L5 transpedicular instrumentation and right L5 vertebroplasty were also carried out. The operation relieved the patient's pain. The paper discusses the clinical, histological, and radiological characteristics of chondromyxoid fibroma, as well as differential diagnosis and treatment modalities.

Key words: Chondromyxoid fibroma, Lumbar spine, Bone tumor, Curettage, Instrumentation

Lomber Omurgada Kondromiksoid Fibrom: Olgu Sunumu ve Literatürün Gözden Geçirilmesi

Özet

Kondromiksoid fibrom aksiyel iskelet sisteminde nadir görülen benign kemik tümörüdür. Literatürde bildirilmiş lomber olgular azdır ve omurganın diğer tümörleri ile karıştırılabilirler. Kliniğimize; 20 yaşında kadın hasta, 3 aydır olan sağ kalça ve bacak ağrısı ile başvurdu. Fizik ve nörolojik muayenesinde anormal bir bulgu saptanmadı ve laboratuvar test sonuçları normaldi. Görüntüleme çalışmalarında, sağ L5 vertebra korpusundan posterior elemanlarına kadar uzanan kistik bir lezyon tespit edildi. CT eşliğinde yapılan biyopsi sonuç vermedi. Bunun üzerine cerrahi planlandı. Ameliyat öncesinde dijital subtraksiyon anjiyografi ve lezyona yönelik embolizasyon işlemleri yapıldı. Operasyonda; L5 laminektomi, lezyonun küretlenerek çıkartılması ve kemik korteksin korunması amaçlandı. Bilateral L4,S1 ve sol L5 transpediküler enstrümantasyon ve sağ L5 vertebroplasti işlemleri gerçekleştirildi. Ameliyat sonrası hastanın şikayetleri geçti. Yazıda, spinal kondromiksoid fibrom klinik, histolojik, radyolojik karakterleri ve ayrıcı tanı, tedavi protokolleri eşliğinde incelendi.

Anahtar Kelimeler: Kondromiksoid fibrom, Lomber omurga, Kemik tümörü, Küretaj, Enstrümantasyon

INTRODUCTION

Benign cartilaginous tumors that originate in the spine can be categorized in four groups: chondromyxoid fibroma (CMF), benign chondroblastoma, osteochondroma, and enchondroma[2]. Chondromyxoid fibromas are the least common group, and are benign tumors of alleged cartilaginous derivation that account for 0.5% to 1% of all bone tumors[1,6-8,14,16,20,25,27-28,32]. There is a predilection for males and the male:female ratio for affected patients is 1.5:1[14]. Chondromyxoid fibromas are most frequently diagnosed in the second to third decades of life[6,14,18,20,25,32-34] but they also occur in older age[18]. Although CMFs are benign, local recurrence has been documented[5-6,14-15,20,34] and reports have also described malignant transformation[3,5-6,11,32]. The treatment of choice for these benign tumors is en bloc resection[6-7,16,20,25,27-28].

Vertebral CMFs constitute 8% of all CMF cases[34]. There are 46 spinal CMF cases in the English literature and the thoracic spine is the region most frequently affected[4,13,18,32]. Twenty-nine of these 46 total reported cases have demographic data available, and these patients were 13 males and 16 females (male:female ratio 0.8:1). Chondromyxoid fibroma of the lumbar spine is rare, and here we report only the tenth documented case[5,10,17,23,25,31-32] (Table I).

Table I Features of the reported cases of lumbar chondromyxoid fibroma in the literature

Authors	Patient age/sex	Tumor location	Presentation	Treatment	Follow-up time	Recurrence
Wu et al. [32]	n/a	Three lumbar cases	n/a	Curettage or excision	n/a	n/a
Saldua et al. [25]	8 years / M	L3 posterior elements	Back pain	Computed tomography-guided biopsy initially, then resection of L3 pedicle, lamina	13 months	No
Cabral et al. [5]	19 years / F	L1-L2 pedicles, transverse processes	Leg pain	Resection of left L1 and L2 laminae, pedicles, transverse processes. Re-resection at recurrence	60 months	Yes
Ramani [23]	44 years / M	T12-L1 pedicles, spinous processes, 12th rib	Cauda equina syndrome	Two-stage: T11-L2 laminectomy and excision, 12th rib resection	2 months	No
Mayer [17]	23 years / M	L2 spinous process	Incidental	Excisional biopsy	n/a	n/a
Gudscha [10]	23 years / F	L3 body	Back pain, leg weakness	Excision	36 months	No
Tsuji et al. [31]	9 years / M	L4 posterior elements	Back pain	Excision and bone grafting	44 months	No
Present case	20 years / F	L5 right pedicle and right half of vertebral body	Right hip and leg pain	Computed tomography-guided biopsy initially, then L5 laminectomy, resection of right pedicle and right half of vertebral body, then instrumentation and cement augmentation	n/a	n/a

abbreviation: n/a: not available

CASE PRESENTATION

A 20-year-old girl was admitted to our outpatient clinic with right hip and leg pain. The pain had begun 3 months earlier and had progressed slowly. Physical and neurological examinations were unremarkable and laboratory test results were within normal limits. Plain lumbar radiographs revealed a hypodense area in the right half of the L5 vertebral body (Figure 1a). Lumbar computed tomography (CT) demonstrated that the lesion was cystic, had a sclerotic well-demarcated wall, and extended from the right L5 vertebral body into the right pedicle (Figure 1b, 1c). Lumbar magnetic resonance imaging (MRI) showed a lesion that was hyperintense on T2-weighted images and that enhanced diffusely on T1-weighted images with contrast (Figure 1g, h). Spinal angiography identified the tumor as hypervascular, and embolization was carried out during the angiography procedure (Figure 2). Computed tomography-guided biopsy was done but the findings were nonspecific and surgery was planned. During surgery, L5 laminectomy and lesion curettage were performed and care was taken to leave the cortex intact. Bilateral L4, S1, and left L5 transpedicular instrumentation (Figure 1d, 1e) and right L5 vertebroplasty (Figure, 1f-white arrow) were also carried out. En bloc resection had been planned but this was not achieved because the lesion bled massively during the operation despite embolization. The histopathological diagnosis was CMF (Figure 3). After the operation, the patient was pain free and neurologically intact.

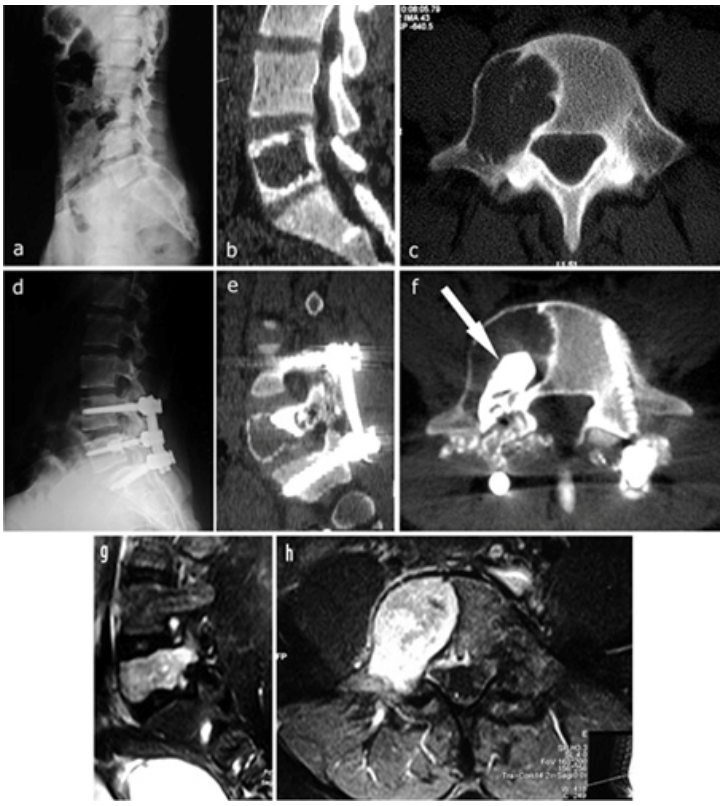

Figure 1: *The patient's preoperative plain lumbar (L) radiograph and lumbar computed tomography scan with sagital reconstruction show a hypodense area in the posterior portion of the L5 vertebral body, and the cystic lesion with sclerotic well-demarcated wall in the right half of L5 extending to the right pedicle (a, b, c). Postoperative plain radiograph and computed tomography scan of lumbar vertebrae with sagital reconstruction show the region after L5 laminectomy, careful curettage leaving the cortex intact, and bilateral L4, S1, and left L5 transpedicular instrumentation (d, e) with right L5 vertebroplasty (f- white arrow). Preoperative T1-weighted magnetic resonance imaging after intravenous gadolinium injection (g, h) revealed diffuse homogeneous enhancement of the lesion*

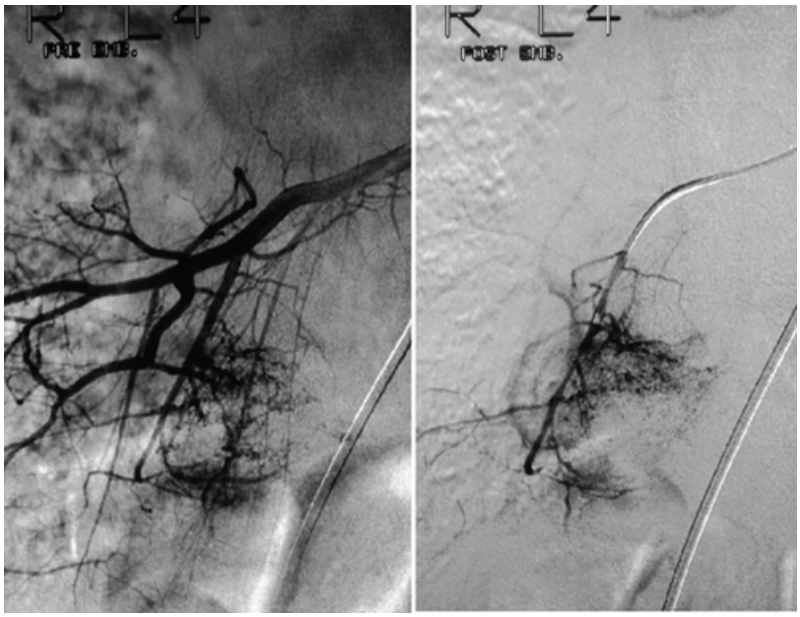

Figure 2: *A preoperative spinal angiogram showed that the lesion was hypervascular (left image: pre-embolization). Embolization was performed during the angiography session (right image: post-embolization)*

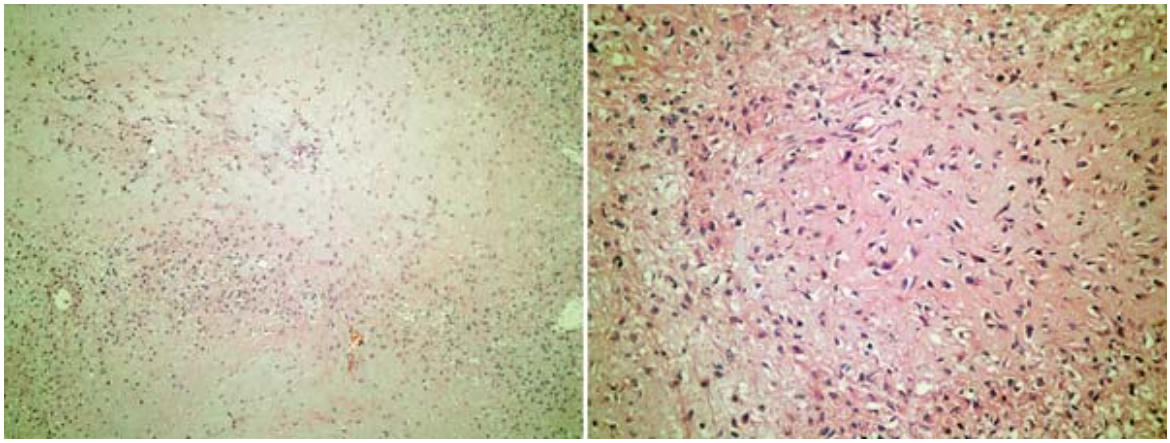

Figure 3: *The histological appearance of the chondromyxoid fibroma: Spindle cells in a characteristic lobular pattern with a central hypocellular area and hypercellular periphery (left image: 40x, HE) and spindle cells in a chondromyxoid matrix (right image: 200x, HE)*

DISCUSSION

Jaffe and Lichtenstein[12] reported the first case of CMF in 1948 and described the tumor as differentiated connective tissue containing chondroid and myxoid elements. Since that description, authors have documented numerous cases and discussed the histopathology, radiology, and general behavior of CMFs[6-7,12,14,20-22,27,32]. Despite the name fibroma, this tumor is clearly a neoplasm of cartilage tissue[5]. The first vertebral CMF case was reported by Benson and Bass[3] in 1955.

The World Health Organization defines CMF as ‘a benign tumor characterized by lobulated areas of spindle-shaped or stellate cells with abundant myxoid or chondroid intercellular material separated by zones of more cellular tissue that is rich in spindle-shaped or round cells with a

varying number of multinucleated giant cells of different sizes"[32]. These neoplasms usually arise in the second or third decades of life[6,14,18,20,25,32-34] and they rarely affect patients younger than 5 years or older than 60 years[28]. They can occur in any part of the skeletal system but mainly affect the long tubular bones of the lower extremities[1,3-6,11-14,16,18,20-21,25,27-29,32]. Most CMFs involve the mataphysis[1,4-5,13-14,25,29,33]. Other rare sites of CMF occurrence are short tubular and flat bones, such as the ribs, scapulae, sternum, mandible, skull, and vertebrae[5-6,14,20] Vertebral involvement is very unusual[3-5,14,16,18-21,24-25,27-28,30,32,34], and thoracic vertebrae are the most frequently affected[4,13,18,32].

Pain, swelling, restricted movement, and tenderness on palpation are the most common presenting signs and symptoms of CMFs[6,20,25]. In most cases, pain is the only clinical manifestation[7,21]. Symptoms may be present many years before CMF is diagnosed[1,5,12] but, occasionally, in vertebral cases aggressive tumor behavior can lead to compression fracture and rapid neurological deterioration[1,5,18-20,30].

While the radiologic appearance of CMFs has been well documented for most body sites, this is not the case for vertebral CMFs[7,16,28]. Most CMFs (any body site) are expansive and have a lobulated, lytic appearance and a sclerotic rim, and these features characterized our case of lumbar vertebra CMF as well[1,5,7,16,20,25,29,33]. Smaller CMFs are round and typically have a thin sclerotic margin, whereas larger ones tend to have cortical bone remnants at their periphery. On plain radiographs, most CMFs exhibit a trabecular appearance and osseous ridges[13]. The sclerosis and trabeculation are even better visualized on CT than on plain radiographs[13]. Contrast enhancement of CMFs is variable and depends on the fibromatoid and vascular components of each tumor[13]. As mentioned, these neoplasms have three tissue components (chondroid, myxoid, and fibrous) that result in heterogeneous intensities on MRI. The chondroid and myxoid components have intermediate to high signal intensity on T2- and proton-weighted images, and low signal intensity on T1-weighted images. The fibrous tissue component of CMFs can have different levels of intensity on MRI[13]. The coarse trabecular appearance on CT corresponds the low signal zones on T2-weighted MRI images[4].

Various case reports have described the gross appearance of CMFs as 'soft, pale gelatinous material"[22,25] or ' mass of grayish-white or bluish-grey"[27]. These neoplasms tend to appear more malignant in the vertebral column other than at other body sites[28]. Some authors have described CMFs causing erosion of the vertebral body and producing a sclerotic rim at the eroded edge[24], and we observed these features in our case as well. Vertebral CMFs can cause cortical expansion and focal destruction[1,28,34], and in many cases they destroy cortical bone[5,7,21,25]. In some patients, the primary bone tumor or metastatic lesion is misdiagnosed as a consequence of this infiltrative behavior of vertebral CMFs[1,20]. In most spinal CMF cases, the posterior elements of the vertebral column are primarily affected, and these tumors typically extend to local vertebral bodies in varying degrees[3-5,13,19-20,24,27-30,32,34]. Patients with vertebral CMF who present late in the course of disease often have extensive local invasion and cord compression, and such patients need extensive surgery[1,29].

Histologically, CMFs contain lobules composed of spindle or stellate cells in a myxoid background, and neoplastic cell aggregates at the periphery of lobules[1,13,16,32-33]. Spindle and stellate cells are more common in the hypocellular zones of CMF lobules, and cellularity increases towards the periphery of the lobules[1,13]. Osteoclast-like giant cells are also present at the periphery[13]. Electron

microscopy reveals that the stellate cells have both chondroblastic and fibroblastic cell characteristics[27]. There may be atypia in CMFs as well, but this has no clinical significance with respect to malignancy or recurrence[8,27,34]. Mitoses are exceptionally rare in CMFs[34]. In a study of 278 cases, Wu et al.[32] reported that 86.7% of the tumors had a lobulated appearance and the remainder had an atypical appearance.

Despite being a cartilaginous tumor, CMFs occasionally calcify[5,7-8,21,33-34]. This change is visible microscopically and is sometimes detected radiographically. Yamaguchi and colleagues[33] identified two rough categories of CMF calcification: i) that which occurs in a paucicellular chondroid matrix and ii) that which occurs between tumor cells. The former produces coarse granular, circumscribed, or trabecular patterns on radiographic images and the latter produces a 'chicken-wire' pattern[33]. On CT, the trabecular pattern is more specific to CMFs[33]. Although hypervascularity is not a common feature of this tumor, our patient's lesion was hypervascular. Despite embolization, her tumor bled massively during surgery.

The differential diagnosis for CMF includes chondrosarcoma, chondroblastoma, and fibrous dysplasia[18]. Chondrosarcomas are also lobulated, but they differ from CMFs in that the centers of the lobules are hypercellular, and CMFs do not have hyaline cartilage, which is a feature of the stroma of chondrosarcomas[18,20]. Like CMFs, fibrous dysplasia also features a myxoid background but these lesions are not lobulated. Based on these characteristics, chondroblastomas can be more easily confused with CMFs; however, CMFs tend to occur in the metaphysis, whereas chondroblastomas typically affect epiphysis[18]. In some cases of CMF, there may be a secondary aneurysmal bone cyst within the tumor[29,34]. In spinal cases of CMF, the part of the lesion that affects the spinous process is more suggestive of CMF than of an aneurysmal bone cyst[5].

Research has revealed associations between aggressive behavior of cartilage tumors and chromosome aberrations at 6q13-21[26]. Investigators have found that clonal rearrangements enclosing band 6q13, such as inv(6)(p25q13), are specifically linked with CMF[9]. This inversion has been proposed as a possible marker of CMF in cases where the diagnosis is difficult to confirm[9].

There are no specific treatment guidelines for vertebral CMFs but the suggested options are based on experience with long bone lesions[1,13]. In the past, curettage was thought to be adequate[12] but numerous cases of recurrence have been documented and the reported rates of recurrence after curettage with or without bone grafting are in the 10% to 25% range[1,6,20,27]. Moreover, curettage alone can leave tumor lobules behind[1,34]. Thus, as with other CMFs, the treatment of choice for spinal CMFs is en bloc resection[6-7,16,20,25,27-28]. When this is not feasible, as in situations where the tumor is less accessible, curettage with bone grafting is advised[6,8,13,16,27-28]. In our case, en bloc resection was planned preoperatively but the tumor bled excessively during the operation so we had to perform curettage with cement augmentation.

Five cases of recurrent spinal CMF have been described in the literature (Table II)[5,14-15,20,34] and 3 of these were in thoracic vertebrae[14]. Laminectomy and decompression of the posterior elements were the reported treatment options[5,15,19] and 2 cases (Table II) recurred[5,15]. Vertebrectomy has also been performed in 4 cases of spinal CMF and none of these tumors has recurred[4,16,24,30]. For CMFs in any location, recurrence is usually diagnosed within 1 year after surgery[32]. However, for the 5 recurrent spinal CMF cases in the literature (C7, T5-6, L1-2) (Table II), the time to recurrence ranged from 2 to 30 years[14]. The risk factors for

recurrence that have been identified for CMFs (all locations) include tumor fragility, which can result in incomplete removal, and separated tumor nests in the bone, which may be overlooked by the surgeon[7,27]. Age is also an important factor in CMF recurrence, with a higher rate observed in younger patients[20-22,32,34]. In the 278 CMF cases (all locations) reported by Wu et al., the average age of the 32 patients with recurrence was 22.6 years[32]. Authors have theorized that this may be explained by the fact that bones of younger patients have a thinner cortex and more spongiosa, and thus offer less resistance to tumor growth[22]. The higher rates of recurrence at younger age may also be due, in part, to the presence of large atypical nuclei and the larger quantity of mucinous material in CMFs of younger-aged patients[21]. Conversely, some authors have proposed that there is no relationship between histology and CMF recurrence[8,34].

Metastatic CMF has not been reported[13,18,25]; however, rare cases of malignant transformation have been documented[34]. Some of these patients received radiation therapy before transformation occurred, but the role of radiation therapy in this process has not been evaluated in depth[28].

Table II: Reported cases of recurrent spinal chondromyxoid fibroma.

Authors	Patient age/sex	Tumor location	Treatment	Time of recurrence post-surgery
Nunez et al.[20]	38 years / F	T5	Curettage and bone grafting	2 years
Kikuchi et al.[14]	11 years / M	T6	Curettage	30 years
Zillmer and Dorfman[34]	20 years / F	C7	Intralesional excision	7 years
Leal Filho et al.[15]	32 years / F	T5	Laminectomy	2 years
Cabral et al.[5]	19 years / F	L1, L2	Resection of left L1 and L2 laminae, pedicles, transverse processes.	5 years

CONCLUSION

Chondromyxoid fibroma is a rare bone tumor of cartilaginous origin that most frequently occurs in the long bones of the lower extremities. These neoplasms typically take a non-aggressive clinical course and rarely involve the spine. Lumbar vertebra CMF is particularly rare. This is the tenth lumbar CMF case reported to date in the English literature. The treatment of choice for CMF in any location is en bloc resection; however, when this is not feasible, curettage with bone grafting is successful. Although CMFs are benign, they can recur locally and can, in rare cases, transform to malignant status. As such, long-term follow-up is very important for CMFs of the spine or any other body site.

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