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# Chondromyxoid fibroma of the seventh cervical vertebra

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Chondromyxoid fibroma is a rare benign bone tumor representing less than 0.5% of all bone tumors. It commonly involves the long tubular bones. Involvement of the spine is rare. A 35-year-old man presented with history of neck pain, restriction of neck movements, pain and numbness along the medial aspect of the left forearm and weakness with wasting of the left hand. A presumptive diagnosis of a bony tumor such as an aneurysmal bone cyst or a giant cell tumor involving the seventh vertebral body was made on plain X-rays, MRI and bone scan. He underwent C7 central corpectomy, incomplete intralesional curettage with iliac bone grafting and C6 to T1 interspinous wiring. The histological diagnosis was chondromyxoid fibroma. On eight years' follow-up, CT scan showed no progression of the tumor with good alignment and fusion of the graft at the site of the corpectomy. The authors conclude that corpectomy and iliac bone grafting for chondromyxoid fibroma has a good outcome on long-term follow-up.

**Key words:** Chondromyxoid fibroma, cervical spine, corpectomy

Chondromyxoid fibroma (CMF), an uncommon benign tumor of bone, was first described by Jaffe and Lichtenstein<sup>[1]</sup> in 1948. Chondromyxoid fibroma occurs predominantly in patients in the second or third decade of life with a slight male predominance.<sup>[2]</sup> It usually affects the long tubular bones, especially the tibia and femur. Involvement of the spine, especially the cervical spine is rare.<sup>[3]</sup> This report reviews the available literature and discusses the surgical management of this rare tumor.

## Case Report

A 35-year-old man presented with history of neck pain, restriction of neck movements, pain and numbness along the medial aspect of the left forearm and weakness with wasting of the left hand for three months.

On examination he had wasting of the left triceps, forearm muscles and the hypothenar eminence of the left hand. There was Grade 4/5 power in the left triceps

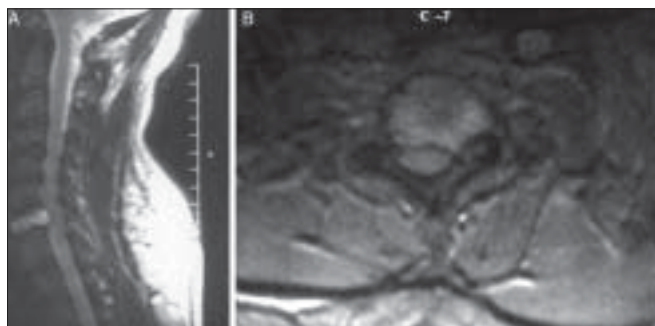
and weakness of the intrinsic hand muscles. There was subjective impairment of all modalities of sensation in left C7, 8, T1, 2 dermatomes. The left triceps reflex was diminished. The rest of the neurological examination was normal. There was tenderness over the lower cervical spine with restriction of the neck movements.

Bone scintigraphy revealed increased uptake in the C7 body. Plain radiographs of the cervical spine showed sclerosis and minimal loss of height of the C7 vertebral body with posterior bulge. The MRI of the cervical spine showed altered signal intensity of C7 body and the left-sided pedicle, hypointense on T1W and hyperintense on T2W sequences [Figure 1A,B]. The disc spaces were normal. The subarachnoid space anterior to the cord at C7 was effaced.

The possibility of a benign bone tumor such as an aneurysmal bone cyst or a giant cell tumor of the seventh cervical vertebra was considered.

Corpectomy of C7 with partial excision of the tumor was performed. Thickening of the pre-vertebral tissue at C7 to T1 level was seen during anterior approach. A grayish vascular tumor in the C7 vertebral body covered the PLL and dura. The tumor was not friable or hemorrhagic and could be removed piece-meal with curettes and disc forceps. An autologous iliac bone graft was placed in the defect after excision and a C6 to T1 interspinous wiring was done.

Microscopic examination showed hypocellular weakly eosinophilic chondromyxoid-like matrix with palisading



**Figure 1:** (A) Preoperative T2W sagittal MR of the cervico-thoracic spine showing an expansile bony lesion involving the C7 vertebral body and causing cord compression. (B) Preoperative T2W axial MR of the cervico-thoracic spine showing the bony lesion involving the C7 vertebral body

spindle cells that imparted a lobular pattern. Psammoma bodies were not seen [Figure 2]. There was no cellular atypia or increased mitoses. The final diagnosis was chondromyxoid fibroma.

The postoperative course was uneventful. Postoperative plain radiographs and CT scans showed satisfactory alignment of the cervical spine. The neck was immobilized in a Philadelphia collar for six months.

On eight years' follow-up, the patient had no neurological symptoms. Plain radiographs and CT scans showed no progression of the tumor with good alignment and union of the graft at the site of the corpectomy [Figures 3A, B and 4A, B].

## Discussion

Benson and Bass<sup>[4]</sup> were the first authors to report CMF of the spine. Only 31 cases of chondromyxoid fibroma of the spine have been reported in the English language literature. Chondromyxoid tumors of the cervical spine are rare and to date only eight cases have been reported.<sup>[5]</sup>

The etiology of the development of CMF is unknown. Recent studies suggest that pericentromeric inversion; inv (6)(p25q13) is a diagnostic marker for this tumor.<sup>[6]</sup>

Histologically this benign cartilaginous neoplasm consists of lobulated areas of spindle or stellate cells

with abundant myxoid or chondroid intercellular material. The lobules are more cellular at the periphery. Osteoclast-like giant cells are often seen at the periphery of the lobule. Calcification is found microscopically in approximately one-fourth of the patients. The stellate cells have features of both chondroblastic and fibroblastic differentiation on electron microscopy.<sup>[7]</sup> On conventional radiographs, a CMF usually appears as a well-margined, expansile, lucent lesion.<sup>[8]</sup> Smaller tumors are usually round with a thin sclerotic margin and uncommonly contain visible calcification or trabeculation. In larger lesions, remnants of cortical bone reinforcing the tumor at the periphery can appear on radiographs as trabeculation. These osseous ridges are responsible for the bubbly cystic radiographic appearance of CMF. CT scans display the sclerotic margin calcifications and trabeculation better than conventional radiographs. They are useful for examining the cortical integrity of the lesion. Contrast enhancement is variable as it depends on the fibromatoid component and vascularity of the tumor. On MRI, CMFs have a heterogeneous appearance due to their different tissue components. The chondroid and myxoid tissues have an intermediate to high signal on proton-density and T2-weighted images and a low signal on T1-weighted images. The fibrous tissue components have a variable appearance. Chondromyxoid fibromas show a high accumulation of fluorodeoxyglucose in PET studies.

In CMF involving the spine the most common site of involvement is the thoracic vertebra.<sup>[2]</sup> Involvement of the cervical vertebrae is rare. The posterior elements and the posterior part of the vertebral bodies are most commonly involved. Radiological differential diagnosis includes chondrosarcoma, osteoblastoma, aneurysmal bone cysts, giant cell tumor of bone, metastasis, multiple myeloma and collapsed hemangioma of the vertebral body.<sup>[3]</sup>

Due to the rarity of the tumor in this location there is no well-established management protocol.

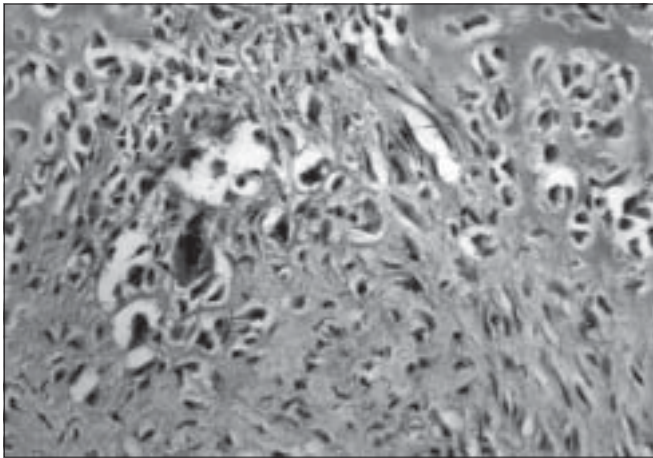


Figure 2: Photomicrograph showing tumor composed of spindle and stellate cells with abundant chondroid intercellular material (H and E, x90)]

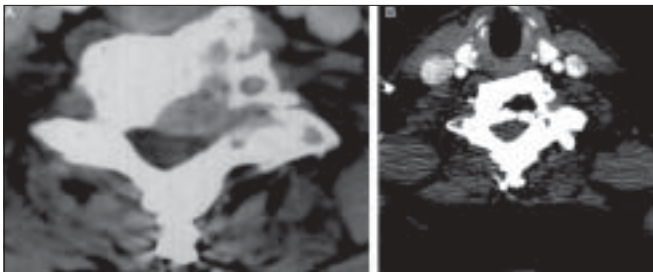


Figure 3: (A) Serial postoperative axial CT scan of the cervical spine done one year after the surgery. (B) Serial postoperative axial CT scan of the cervical spine done eight years after the surgery showing graft in situ and good fusion at the graft site

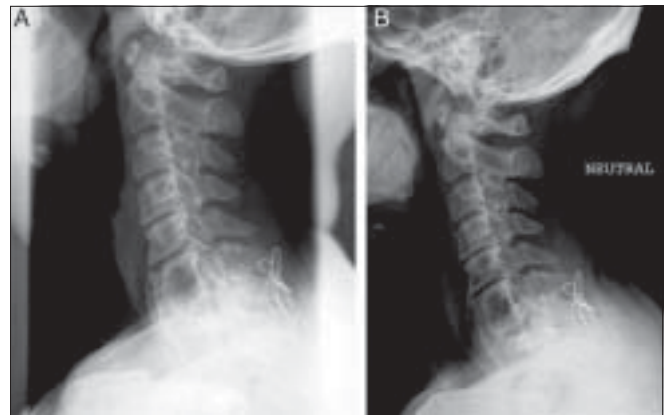


Figure 4: (A) Postoperative X-ray of the cervical spine done three years after surgery. (B) Postoperative X-ray of the cervical spine done eight years after surgery showing the graft in situ, good alignment and no evidence of instability

The possibility of recurrence after surgery, 11% in a series with 278 patients,<sup>[2]</sup> dictates that the tumor should be removed en bloc. This is relevant for CMF in the long bones but such strategies in the cervical spine may produce instability. Hence less radical procedures such as curettage with or without bone grafting are employed to preserve mechanical stability. This, however, significantly increases the chance of recurrence to 80% in the case of 'curettage alone' and 7% in the case of 'curettage with bone grafting'.<sup>[9]</sup> In the management of this case we decided in favor of 'curettage with bone grafting' and no local recurrence is seen until now.

Lopez-Ben *et al.*<sup>[5]</sup> report the case of a man with a CMF involving the C2 vertebra - complete removal of tumor was achieved via a trans oral approach followed by a C1-C4 fusion. There was no evidence of neck pain, instability or local recurrence on a two-year follow-up.

Such a 'complete resection' is not always possible. Several authors have reported favorable results [Table 1] even in the absence of a complete excision. In this case the tumor was involving the entire body of the seventh cervical vertebra. Extension into the left C7 pedicle and into the paraspinal muscles on the left side made complete excision in this case difficult.

Adjuvant radiation is not recommended because

of the potential of malignant transformation. Zillmer and Dorfman<sup>[3]</sup> report a case of a woman with a CMF involving C7 that was wrongly reported as a giant cell tumor and was treated with radiation therapy following intralesional excision. She presented seven years later with recurrent CMF and malignant fibrous histiocytoma. Dahlin and Unni<sup>[14]</sup> also have reported a case with development of a fibrosarcoma at the site of a tibial CMF that had received radiation therapy.

In our patient only a partial excision was achieved. No radiation therapy was given. On an eight-year follow-up, there was no recurrence of tumor. In addition the iliac bone graft had fused well and there was no evidence of instability at the follow-up. The patient had resolution of his neurological deficits postoperatively and had maintained this complete neurological recovery over a significantly long period.

## Conclusion

Chondromyxoid fibroma involving the cervical spine is rare. Complete resection is the treatment of choice. When complete resection is not possible, partial resection with bone grafting yields good results. A close long-term follow-up is mandatory, more so in young patients in whom only a partial excision has been achieved as this tumor is known to recur.

**Table 1: Chondromyxoid fibroma of the cervical spine - presentation, management and outcome**

Authors, year [ref]	Age/sex	Site	Clinical presentation	Radiographic findings	Treatment	Results and follow-up
Schajowicz and Gallardo 1971 <sup>[7]</sup>	6/F	C3	No clinical details	Expanded spinous process	Resection - details unavailable	Well at 4 years
Standefor <i>et al.</i> 1982 <sup>[10]</sup>	20/F	C7	Neck pain radiating to the left arm	Bulky mass in the neck, destruction of C5 body and posterior elements with mottled calcification	1500 rad to neck, fusion C7-T1, soft tissue resection	No recurrence - 15 months
Provelegios <i>et al.</i> 1988 <sup>[11]</sup>	32/M	C4	Pain in the left shoulder	Lytic lesion of posterior body, sclerotic margin, spinal canal not involved (myelogram)	Curettage bone graft	No evidence of disease - 2 years
Zillmer and Dorfman 1989 <sup>[3]</sup>	20/F	C7	No clinical data	Body, neural arch eroded. Spinal canal involved	Intralesional excision and radiation therapy	Presented seven years later with malignant transformation
Rivierez <i>et al.</i> 1991 <sup>[12]</sup>	41/F	C5	Torticollis, upper limb pain	Destruction of part of body, posterior elements, peripheral scoliosis	Resection in 2 stages	No evidence of recurrence 10 months
Lopez-Ben <i>et al.</i> 2002 <sup>[5]</sup>	20/M	C2	Neck pain	Lytic lesion C2 body, instability at C1-C2	Trans-oral resection followed by occiput to C4 posterior fusion	Resolution of pain, instability and no recurrence 2 years
Bala <i>et al.</i> 2006 <sup>[13]</sup>	36/M	C2	Incidental	Expansile, septated, enhancing cystic lesion of the C2 body, destruction of the lateral mass and posterior elements on the right	Trans-oral approach, bony tumor curetted - cavity packed with bone. Partial excision achieved	18 months follow-up no pain. Repeat imaging - consolidating bone graft and a stable volume of residual tumor

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Accepted on 10-04-2007

**Source of Support:** Nil, **Conflict of Interest:** None declared.