BRIEFLY NOTED

Monostotic Fibrous Dysplasia of the Thoracic Spine

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FIBROUS DYSPLASIA accounts for 2.5% of all bone masses and 7% of benign bone tumors. It is a condition characterized by the metaplastic replacement of the medullary component of predominantly long bones with fibrous tissue.[2,3,10] This change is responsible for the characteristic clinical features, including bony deformities, pathologic fractures, and pain.[5,10] The monostotic form of fibrous dysplasia occurs more frequently than the polyostotic disease.[3] The former occurs with equal frequency between the sexes.[2,3,5,10] It is considered to be a disease of children and young adults which neither regresses nor enlarges after the cessation of bone growth.[2,3,5,10,12,14,15]

The monostotic form does not exhibit the laboratory abnormalities, extraskeletal manifestations, or endocrinopathies associated with the polyostotic disease.[2,4,5,12,14,15] Monostotic fibrous dysplasia occurs most frequently in the metaphyses of long bones and the ribs.[3,5,14,15] The clinical presentation of painful swelling or pathologic fractures[2,3,5,14,15] and the radiologic picture of an expansile, trabeculated lesion with preserved cortical margins[3,10,11,14,15] fail to differentiate the monostotic form from many other isolated lesions of bone.[15] Excisional biopsy and recognition of the histologic picture of immature trabeculae of woven bone without surrounding osteoblasts in a predominantly fibrous network is required for diagnosis[3,5] and prevention of occasional malignant transformation.[2,4,5,12]

The occurrence of vertebral lesions in polyostotic fibrous dysplasia has been estimated at 7% (cervical lesions) to 14% (lumbar lesions).[4] The true incidence is probably much lower.[2,6] Only six previous cases of vertebral involvement (three cervical, three lumbar) in monostotic fibrous dysplasia have been reported.[1,4,7,11-13]

The first case of monostotic fibrous dysplasia of the thoracic spine is presented here, as are its plain film, computerized axial tomographic, and magnetic resonance characteristics. The features of these seven cases of monostotic fibrous dysplasia of the spine are reviewed.

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CASE HISTORY

A 20-year-old male presented with the acute onset of right sided neck pain after suffering blunt trauma to the head. There was no history of fractures or painful swelling. No history of precocious puberty could be elicited. Physical examination revealed normal cervical lordosis without limited range of motion or tenderness. The right triceps reflex was diminished, as was the strength of the right extensor digitorum. There were no cutaneous pigmented lesions, physical stigmata of endocrinopathy, or osseous deformities. Thyroid function studies were normal, as were serum calcium, phosphorus, and alkaline phosphatase.

Plain roentgenograms of the spine showed a trabeculated, expansile 4 cm irregular lesion involving the right side of the T1 vertebral body and the right pedicle, superior articular facet, and the lamina. The mass extended superiorly to involve the right inferior facet of C7. The cortex was thinned and expanded but intact (Figure 1). Computerized axial tomography clearly demonstrated the mixed density of the tumor which deformed the right side of the spinal canal. The interface with normal vertebral body demonstrated the sclerosis of compressed bone (Figure 2). Magnetic resonance imaging exhibited a mottled increased intensity signal of the lesion in a T2 weighted image (Figure 3). Bone survey demonstrated no additional lesions.

The patient underwent laminectomy of C7 and T1 with costotransversectomy and pediculectomy of T1. With the patient in Gardner-Wells traction to maintain spinal alignment, computerized axial tomography showed the sclerotic border of residual lesion within the vertebral body (Figure 4). A second procedure via an anterior paratracheal approach permitted complete removal of the fibrous, expansile mass from the T1 vertebral body. Although this exposure provided the option of median sternotomy if necessary, it was possible to perform the entire operation through a suprasternal incision. The defect was filled with a corticocancellous strut taken from the iliac crest. Postoperatively, spinal stabilization was assured with a halo vest until fusion occurred at 3 months.

Postoperative computerized axial tomography confirmed complete resection of abnormal tissue and good placement of the bone graft (Figure 5). Neurological examination of the patient was normal. Pathologic examination of the specimen indicated fibrous dysplasia (Figure 6).

DISCUSSION

The present case is characteristic of monostotic fibrous dysplasia with regard to clinical presentation, radiologic appearance, and pathologic findings. Its unique feature is the occurrence of the monostotic lesion in the thoracic spine. The features of this case and the six previous cases of monostotic fibrous dysplasia of the spine are presented in Table 1.

Three of the previous lesions were located in the cervical spine while the remaining three were confined to the lumbar region. There was a slight female preponderence which accounted for 57% of the cases. Age at presentation ranged from 20 to 58 (mean 33 years old). The most common complaint was pain, occurring in four of the seven patients.

The characteristic radiographic appearance of "ground-glass" was observed in this case. The typical ballooned, expanded appearance of a bone with preserved cortical margins suggested the diagnosis of fibrous dysplasia. The present report is the first demonstration of the magnetic resonance imaging characteristics of this

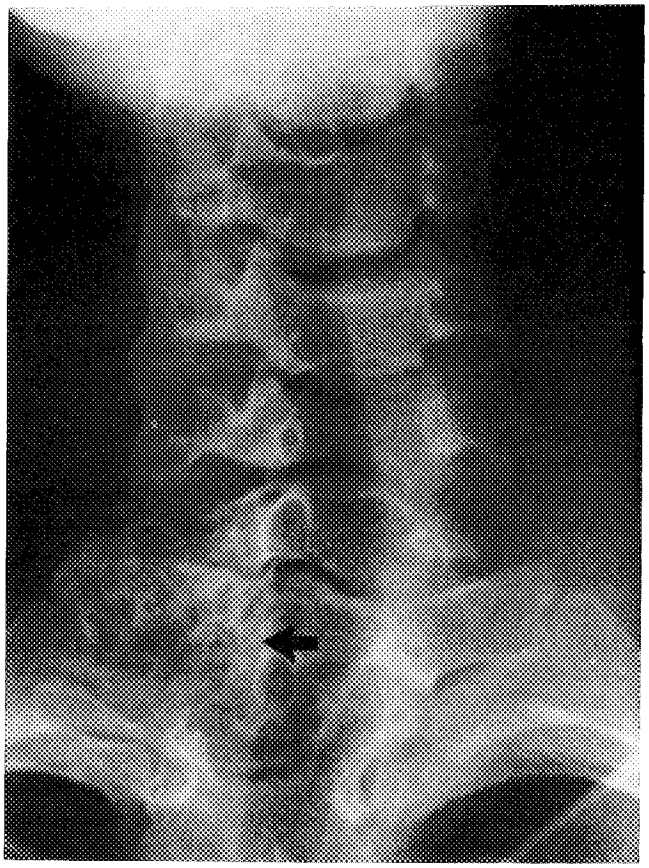

Fig 1. Roentgenogram of the cervical spine demonstrating a 4 × 4 cm irregular, sclerotic and lytic mass (arrow) extending lateral and superior from the right side of the T1 vertebral body.

lesion. Magnetic resonance imaging is the modality of choice to demonstrate the characteristics of fibrous lesions.[9] Normally, the cortical bone of the spine is a signal voided region. The pathologic bone of fibrous dysplasia has a higher signal intensity than the normal medullary bone of the vertebra due to replacement of mature bone with irregular, woven bone and fibrous tissue and com-

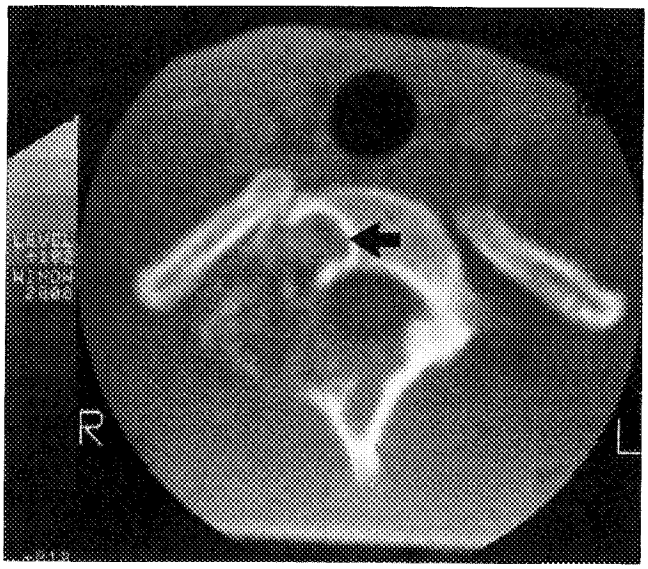

Fig 2. Computerized axial tomogram (bone window) showing the dense rim of compressed bone between the normal T1 vertebral body and the lesion (arrow) involving the body, pedicle, and lamina.

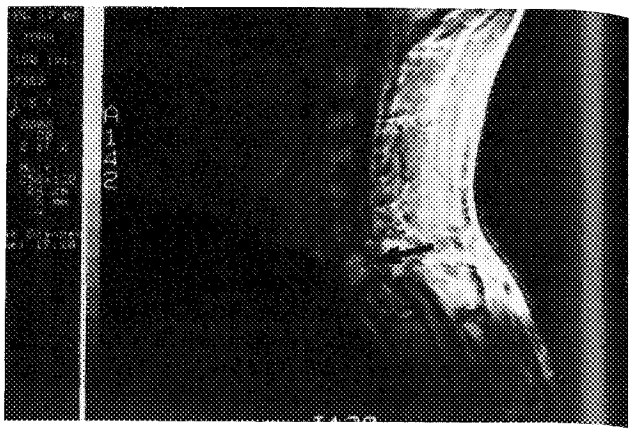

Fig 3. Magnetic resonance scan (T2 weighted image) revealing the heterogeneous higher signal intensity of the T1 vertebral body (arrow) compared to the normal cervical spine.

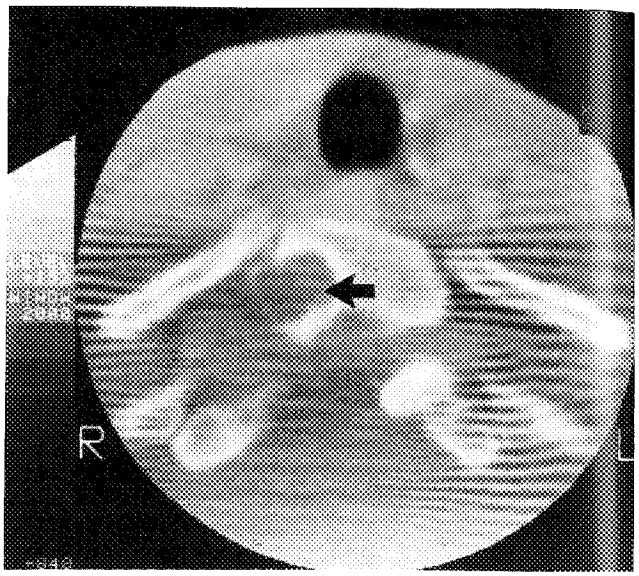

Fig 4. Computerized axial tomogram (bone window) demonstrating resection of the posterior component of the mass. The anterior portion protrudes from the right side of the T1 vertebral body (arrow).

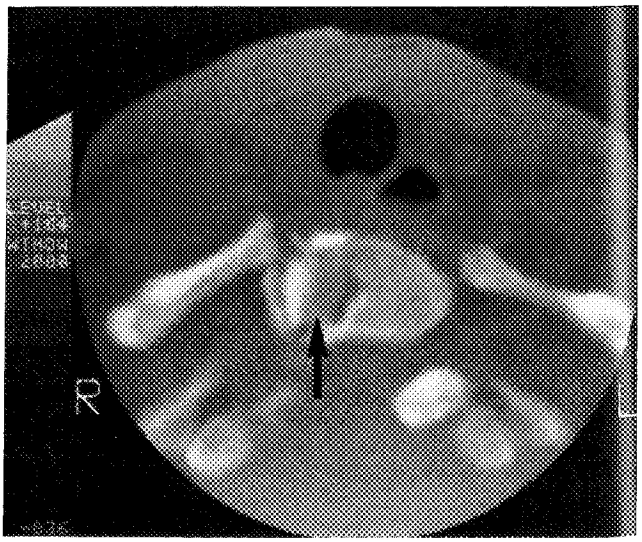

Fig 5. Postoperative computerized axial tomogram (bone window) confirms adequate positioning of the bone graft (arrow) and no residual pathologic tissue.

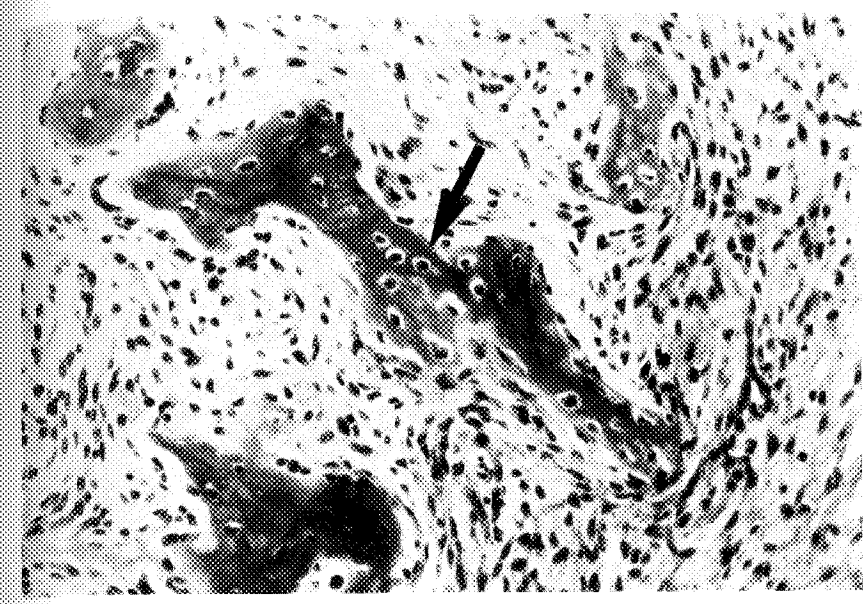

Fig 6. Photomicrograph reveals immature woven bone (arrow) dispersed in a field of predominantly disorganized fibrous tissue (Hematoxylin and eosin; original magnification ×100).

Table 1. Characteristics of the Reported Cases of Monostotic Fibrous Dysplasia of the Spine

	Sex	*Age*	*Level*	*Presentation*	*Location*
Schlumberger[13]	m	20	C4	post-traumatic	body
Rosendahl-Jensen[12]	f	35	C4	post-traumatic	body, superior process, transverse and spinous processes
Resnik and Lininger[11]	f	27	C6	pain	body, pedicle, lamina
Rosenblum et al	m	20	T1	pain	body, superior articular process, pedicle, lamina
Ledoux-Lebard and Soulquin[7]	f	58	L1	myelopahy	body, pedicle
Daniluk and Witwick[1]	f	28	L4	pain	transverse process, articular processes
Harris et al[4]	m	42	L4	pain	transverse process

pression of normal medullary bone. The heterogeneity of the high intensity signal is secondary to the nature of the mixture of interwoven elements which occur in this process.[9] Other tumors of the vertebral column more typically present a homogeneous appearance, frequently demonstrating a lower intensity signal than surrounding medullary bone.[8]

The paratracheal approach through Burn's fascia has been used in surgery of the lower cervical spine, particularly for anterior cervical discectomy. Combined anterior and posterior surgery and fusion has been most frequently used in the management of neoplastic disease of the spine, especially for metastases. This initial description of its use in fibrous dysplasia of the spine demonstrates that all metaplastic tissue can be removed with maintenance of spinal stability.

Fibrous dysplasia is a benign bone lesion without destructive capability. Traditionally, once monostotic fibrous dysplasia of the spine has been confirmed by biopsy, no further therapy has been recommended. Only one of the previously reported patients presented with neurologic deficit.[7] Persistent pain and focal neurologic findings in the current case prompted radical surgery to eliminate all pathologic tissue. Although the initial procedure was sufficient to decompress the C8 nerve root, a second operation was deemed necessary. The unusual appearance of pain after the cessation of bone growth in this patient, and the persistence of pain after the initial operation, suggested a more aggressive variant of the disease. Complete removal of dysplastic tissue, usually not necessary, was carried out to forestall the possibility of malignant degeneration in some portion of the lesion. Histologic evidence of malignancy could not be demonstrated after either operation.

This report and review of the literature serves to demonstrate the occurrence of fibrous dysplasia as an isolated lesion at any location along the spinal column. The clinical characteristics, radiographic features, and management of this mass have been discussed.

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Spinal Tuberculosis with Spontaneous Ventral Extrusion of Two Vertebral Bodies

A Case Report

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THE CLASSICAL PICTURE of spinal tuberculosis includes involvement of two vertebrae with rarefaction, destruction of the intervening intervertebral disc, and paravertebral abscess formation. A rare total displacement of two sequestrated vertebral bodies, anatomically intact, is reported. A similar case has not been found in the literature.

CASE REPORT

An 11-year-old black boy complained of thoracolumbar back pain for 7 months. There was no history of trauma or previous tuberculous involvement. Examination revealed marked cachexia, and a gibbus at the thoracolumbar level. Spinal movements were painful and restricted, with marked spasm of the erector spinae. Tenderness was present posteriorly in the midline, from T9–L2 vertebral levels. A fluctuating mass, measuring 20 × 10 cm, extended obliquely downwards from the 12th rib to the iliac crest on the left side. Neurologic examination was normal. Roentgenograms of the spine showed complete sequestration of the bodies of T11 and T12 vertebrae, both of which were displaced to a position immediately in front of the L1 body. The pedicles of the displaced vertebrae were eroded close to the bodies (Figure 1). A large paravertebral abscess was present. Laboratory tests indicated leukocytosis and microcytic anemia. The erythrocyte sedimentation rate (ESR) was 120 mm/1st hour, Westergren. Radioisotope scanning showed no additional spinal or extraspinal tuberculous lesions. Tubercle bacilli were positively identified in three successive early morning sputum specimens, confirming the diagnosis.

An anterior decompression, debridement, and rib vascular pedicle graft was performed as a first-stage surgical procedure via a left-sided thoracoabdominal approach. The ninth rib, together with a collar of soft tissue containing the intercostal artery, was mobilized as a vascular pedicle graft.[1] A large paravertebral abscess was incised and 500 ml of yellow pus drained. The vertebral bodies of T11 and T12 were lying free in the abscess cavity. A debridement procedure which included soft tissue and bone was completed, with decompression of the spinal canal. A bed for the reception of the bone grafts was prepared in the bodies of the T9 and L1 vertebrae. A free fibular graft and a vascularized rib graft were locked into position in the prepared bed. The diaphragm and chest wall were repaired in the routine manner, and an intercostal underwater drain was inserted. The postoperative course was uneventful. Histologic examination confirmed the diagnosis of tuberculous spondylitis. Postoperative management included a triple-drug regimen of isoniazid, pyrazinamide, and ethambutol.

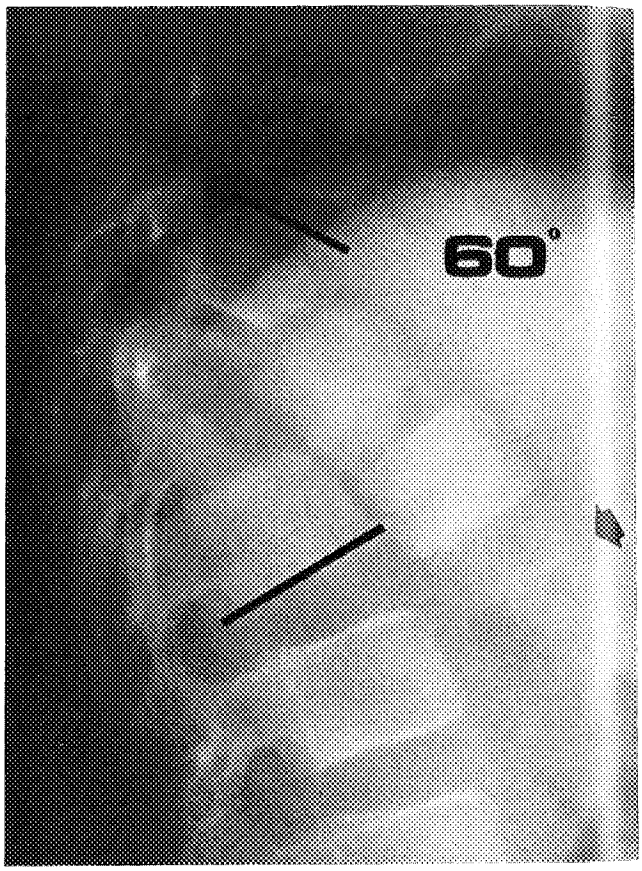

Fig 1. Lateral radiograph of the thoracolumbar spine. The vertebral bodies of T11 and T12 can be identified in front of L1. The radiopaque line inferior to T12 vertebral body (arrow) marks the superior end-plate of L1 vertebra. The kyphotic angle is 60°, measured from T10 to L1 levels.

At a second-stage procedure after 2 weeks, a posterior fusion with Harrington compression instrumentation from T9 to L1 was completed. The patient was mobilized in a spinal brace after the wounds were healed. Tomograms showed bony fusion of the vascular pedicle graft after 5 weeks and hypertrophy after 5 months (Figure 2). The kyphotic deformity was reduced from 60 to 30°. The brace was continued for 9 months, and the antituberculosis medication for a total period of 12 months. At a 22-month follow-up examination, the patient was symptom-free, the kyphus was unchanged at 30°, and there were no signs of recurrence of infection.

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