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ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Dr. Antonio Corvino is a PhD in the Motor Science and Wellness Department at University of Naples "Parthenope". In 2008, he obtained his MD degree from the School of Medicine, Second University of Naples. Then, he completed a residency in Radiology in 2014 at University Federico II of Naples. In 2015, he undertook post-graduate training at Catholic University of Rome, obtaining the 2nd level Master's degree in "Internal Ultrasound Diagnostic and Echo-Guided Therapies". In 2016-2018, he served on the directive board of Young Directive of Italian Society of Ultrasound in Medicine and Biology. His ongoing research interests involve ultrasound and ultrasound contrast media in abdominal and non-abdominal applications, etc. (L-Editor: Filipodia)

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WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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Imaging characteristics of a rare case of monostotic fibrous dysplasia of the sacrum: A case report

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Abstract

BACKGROUND

Fibrous dysplasia (FD) is a common benign intramedullary fibro-osseous lesion. Involvement of the spine is rare, with the literature including only case reports, and cases of monostotic FD (MFD) in the sacrum are extremely rare. A correct preoperative diagnosis of spinal MFD is important for clinicians to select proper treatment.

CASE SUMMARY

We retrospectively assessed a case report of MFD in the sacrum. This patient was examined by computed tomography (CT) and magnetic resonance imaging (MRI), and the diagnosis was confirmed by pathology. A review of the literature was performed to analyze the imaging characteristics and differential diagnoses of spinal MFD. For our patient, the CT scan showed the lesion to be expansile, with ground glass opacity and a sclerotic rim. On MRI, the lesion showed iso-low signal intensity on T1WI and iso-high signal intensity on T2WI. A low signal rim was found on T1WI and T2WI. Our patient was treated by posterior focal excision, decompression, bone grafting, fusion and pedicle screw fixation. A satisfactory result was achieved, with pain disappearance. No complications had occurred at the 1-year follow up.

CONCLUSION

MFD is an expansile osteolytic change. Ground glass opacity and a sclerotic margin are obvious characteristics. The lesion often involves the vertebral body and posterior element. Knowledge of these imaging characteristics of spinal FD could be helpful for diagnosis and prevent unnecessary procedures.

Key Words: Monostotic; Sacrum; Fibrous dysplasia; Spine; Computed tomographic; Case

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Core Tip: This report presents a rare case of monostotic fibrous dysplasia (MFD) involving the sacrum. The imaging manifestations of MFD include expansile lesions, ground glass opacity, and sclerotic rims. Most lesions show iso-low signal intensity on T1WI and iso-high signal intensity on T2WI. These features can provide a suggestive diagnosis to distinguish MFD from giant cell tumour, aneurysmal bone cyst, and vertebral haemangioma. Accurate diagnosis of MFD in the spine is of great value for clinicians to choose an appropriate treatment.

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INTRODUCTION

Fibrous dysplasia (FD) is a benign intramedullary fibro-osseous lesion originally described by Lichtenstein in 1938^[1]. FD is most commonly found in the long bones^[2], and it is rare in the spine and extremely rare in the sacrum^[3]. In the spine, FD is found either in the monostotic or polyostotic form, but monostotic FD (MFD) is rare^[4]. According to our literature search, this is the seventh case reported in the sacrum. Kinnunen *et al*^[5] reviewed the literature and collected 136 cases of FD confirmed by pathology, including all parts of the body, in which the incidence in a vertebral body was 24/136 (18%). The proportions of vertebral body FD were 17% in the cervical region, 29% in the thoracic region, 29% in the lumbar region, and only 8% in the sacral region (2/24). Firat *et al*^[6] reviewed a single case of MFD in the sacrum, and Schoenfeld *et al*^[7] described three patients with monostotic disease of the sacrum.

Treatment of spinal MFD depends on the clinical symptoms. Asymptomatic patients with stable lesions can undergo clinical observation. Patients with severe pain can undergo surgery. A correct diagnosis of spinal MFD is important for the clinician to select an appropriate treatment. Therefore, in this rare case report of MFD in the sacrum, we reviewed the literature to analyze the clinical, radiographic, and pathologic features of spinal MFD.

CASE PRESENTATION

Chief complaints

A 60-year-old woman suffered from low back pain for more than 2 years and aggravating radiating pain and numbness in both lower limbs for more than 2 months. The study was approved by the institutional review board of the hospital, and the patient signed an informed consent form.

History of present illness

Two years prior, the patient had low back pain and discomfort with no obvious cause that could be relieved after rest. The patient paid no attention to the pain, and no further examination or treatment was performed. Then, the symptoms recurred, which were sometimes mild and sometimes severe. In the previous two months, the patient had experienced radiating pain of both lower limbs and numbness on the posterolateral side of the leg, especially on the right side. After rest, the symptoms were partially relieved, and then the pain gradually aggravated and numbness occurred.

History of past illness

The patient denied any history of hepatitis, tuberculosis, malaria, hypertension, heart

disease, diabetes, cerebrovascular disease, mental illness, surgery, trauma, blood transfusion, or food or drug allergy.

Personal and family history

The patient had no history of contact with an epidemic area, situation or contaminated water; no residential history in a pastoral, mining, high fluoride or low iodine area; no contact history of chemical, radioactive or toxic substances; and no history of drug abuse, smoking, drinking, or recreational tourism.

Physical examination

Tenderness and percussion pain were observed in the L4-S1 spinal space, and the pain radiated to the right lower limb, buttocks, posterolateral areas of the upper and lower legs, and right foot including the toes. The lumbar range of motion was limited. The straight leg raising test was positive at 40° in the right lower limb and negative in the left lower limb. The temperature and tactile sensation of lateral pain were decreased in the right leg. No skin lesion or soft-tissue mass was noted.

Laboratory examinations

Laboratory tests included examinations of liver and kidney function, electrolytes, tumour markers and coagulation and routine blood tests. Among them, the glutamic oxaloacetic transaminase level was 11.6 U/L (reference range 13-35 U/L), the total protein level was 57.5 g/L (reference range 65-85 g/L), the albumin level was 36.4 g/L (reference range 40-55 g/L), the urea level was 7.7 mmol/L (reference range 2.6-7.5 mmol/L), and the chlorine level was 108.4 mmol/L (reference range 96-108 mmol/L). The tumour markers were all in the normal range. Routine blood tests were normal.

Imaging examinations

The patient did not undergo X-ray examination before the operation. Imaging studies included computed tomography (CT) and magnetic resonance imaging (MRI), which demonstrated an expansile lesion involving both the anterior and the posterior elements of the vertebral body and no involvement of the surrounding structures. The lesion exhibited ground glass opacity with a high-density sclerotic rim on CT and presented a mixed signal intensity on MRI (Figure 1A-F).

FINAL DIAGNOSIS

The patient underwent surgery, and the lesion in the sacral vertebra was fully removed along the S1 pedicle channel and sent for pathological examination. Pathological haematoxylin and eosin (HE) staining results supported the diagnosis of FD (Figure 1G).

TREATMENT

Posterior debridement, iliac bone grafting and internal fixation were adopted. The patient underwent an X-ray examination at three days after the operation (Figure 1H and I).

OUTCOME AND FOLLOW-UP

Currently, neither clinical symptoms nor signs of tumour recurrence have been detected during a follow-up period of more than 18 months.

DISCUSSION

Overview

According the World Health Organization's (WHO) 2020 version of the bone tumour classification, FD is classified as a benign tumour in the category of other mesenchymal tumours of bone. It is a common non-malignant fibro-osseous lesion,

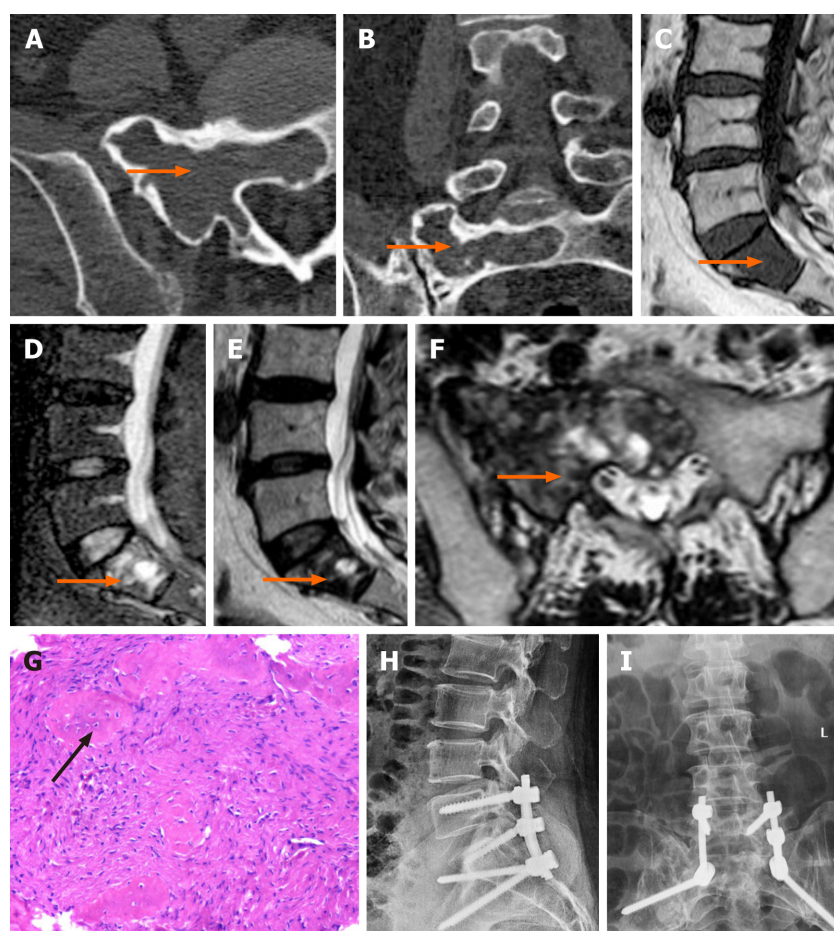


Figure 1 A 60-year-old female with monostotic fibrous dysplasia in the sacrum. A and B: Computed tomography images showing an expansile lesion with a sclerotic rim and ground glass opacity; C: Sagittal T1WI showing low signal intensity; D-F: Sagittal T2WI, FS T2WI and axial T2WI showing iso-high signal intensity; G: Pathological haematoxylin-eosin (HE) staining ($\times 10$) showing tumour-like hyperplasia of fibrous tissue with surrounding fibrotic bone formation (arrow); H and I: After the operation, the patient underwent an X-ray examination.

accounting for 7% of benign bone tumours^[2]. MFD occurs more frequently (70%-80%) than polyostotic FD (20%-30%) or McCune-Albright syndrome, which is a variable condition with endocrine and cutaneous abnormalities (3%)^[8]. The occurrence of FD in the spine is rare, but among these cases, occurrence in the cervical and thoracic regions is relatively more common. Reports that describe FD in the sacrum are rare. Based on a literature review^[9-11], the demographic details of spinal MFD patients are summarized in [Table 1](#).

Aetiology and clinical manifestations

At present, the exact aetiology of MFD is unknown. A sporadic activating mutation of Gs alpha on chromosome 20q13.2-13.3 has been reported to be associated with the occurrence of FD^[12], which leads to increased utilization of cyclic adenosine monophosphate^[13].

The peak incidence of spinal MFD is in the third to fifth decade of life, and there is no significant gender difference^[7,14]. MFD develops during skeletal development. However, clinical manifestations depend on the site of the lesion^[13,15]. Pain is usually proportional to the degree of vertebral involvement^[16].

Histopathological and imaging characteristics

MFD is characterized by the replacement of bone marrow with poorly organized spicules of immature bone^[10,15]. Insufficient bone mineralization results in loss of mechanical strength, which may result in skeletal deformity and vertebral collapse.

A typical CT characteristic is “ground glass opacity”^[16]. The ground glass opacity appearance is due to the presence of many irregular spicules of bone within the fibrous stroma^[17]. Furthermore, expansile and lytic changes are common features. A sclerotic rim is observed around the lesion, and the cortical bone is thin with no disruption.

Table 1 Demographic details of patients with monostotic fibrous dysplasia in the spine

	Cases	Percentage (%)
Sex		
Male	24	60
Female	16	40
Age (y)		
Range (mean \pm SD)	11-77 (37.85 \pm 15.84)	
Presentation		
Posttraumatic	3	7.5
Pain	19	47.5
Pain and radiculopathy	7	17.5
Incidental finding	5	12.5
Other	6	15

MRI is useful for evaluating the entire extent of the lesion and the soft-tissue components. The MRI characteristics of FD are variable; typically, imaging results demonstrate hypo-intensity with several iso-intense regions on T1WI and iso-intensity with several hyperintense regions on T2WI. The heterogeneous signal may be related to the trabecular bone, collagen fibres, cystic changes, and haemorrhage in the lesion. MFD may be surrounded by a thick, sclerotic rim, called a rind^[18]. The rind can be seen as hypo-intense on T1WI and T2WI. No obvious soft-tissue involvement is observed.

Differential diagnosis

The differential diagnosis of spinal MFD should include benign and intermediate (locally aggressive) tumours, such as giant cell tumour (GCT), vertebral haemangioma (VH), and aneurysmal bone cyst (ABC)^[4,8,17]. GCTs commonly occur in the first and second sacral vertebrae in patients 20-40 years of age. A GCT is an invasive intermediate tumour with swelling and irregular changes. Multiple vacuoles are visible, the surrounding sclerotic rim is not obvious, and the cortical bone is partially interrupted (Figure 2)^[19,20]. An ABC is classified as a tumour with an intermediate, undefined, neoplastic nature^[8]. It is common in patients younger than 20 years old. It involves expansive bone destruction, thinning of the cortical bone, and partial cortical interruption. The presence of a “fluid-fluid” level on MRI is characteristic^[21], and the hardening margin is visible (Figure 3). A typical sign of haemangioma is the “corduroy cloth” manifestation^[7]. Large trabecular bone can be seen on CT and MRI, which is usually non-expandable and has a circular-like shape. It exhibits high or low signal intensity on T1WI and high signal intensity on T2WI.

In addition, chordoma should also be excluded because it occurs in the sacrum, often in the midline of the third to fifth sacral vertebrae. It is a malignant tumour with osteolytic bone destruction and a large soft tissue mass. Chordoma is relatively easy to differentiate from FD. It exhibits low and high signal intensity (related to haemorrhage) on T1WI, and it exerts high signal intensity (related to the intratumoural gel and mucinous tissue) on T2WI (Figure 4).

The treatment of MFD focuses on symptom relief and preventing lesion progression^[22]. Bisphosphonates are first-line drugs for pain relief^[7,10]. When conservative management fails, surgery should be considered to restore spinal stability.

CONCLUSION

This report presents a rare case of MFD involving the sacrum. The imaging manifestations of MFD include expansile lesions, ground glass opacity, and sclerotic rims. Most lesions show iso-low signal intensity on T1WI and iso-high signal intensity on T2WI. These features can provide a suggestive diagnosis to distinguish MFD from GCT, ABC, and VH. Accurate diagnosis of spinal MFD is of great value for the clinician to choose an appropriate treatment.

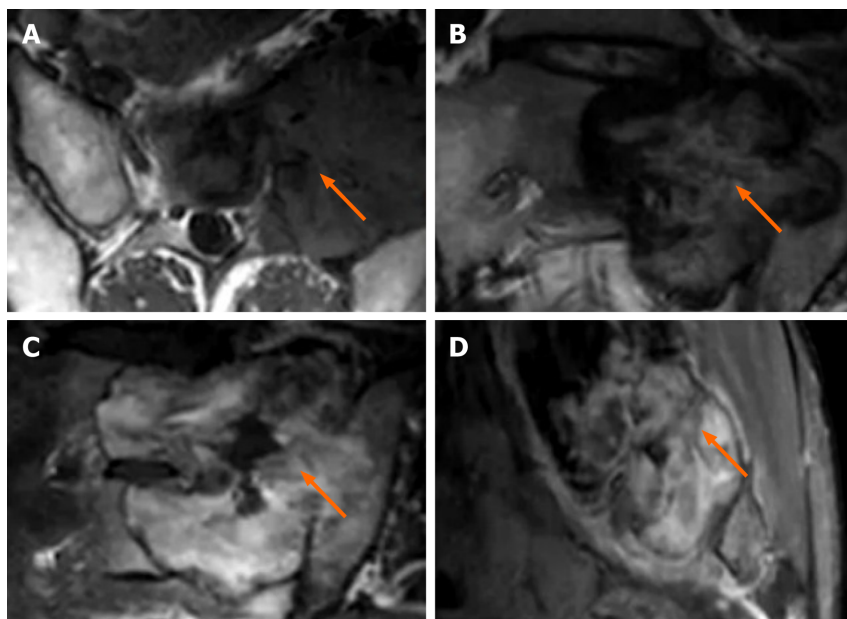


Figure 2 Male, 35 years old. The sacrum showed expansive bone destruction. The pathological result was a giant cell tumour of the bone. A: Axial T1WI shows iso-low signal intensity with a sclerotic rim; B: Axial T2WI shows iso-high signal intensity; C and D: The lesion showed obvious heterogeneous enhancement.

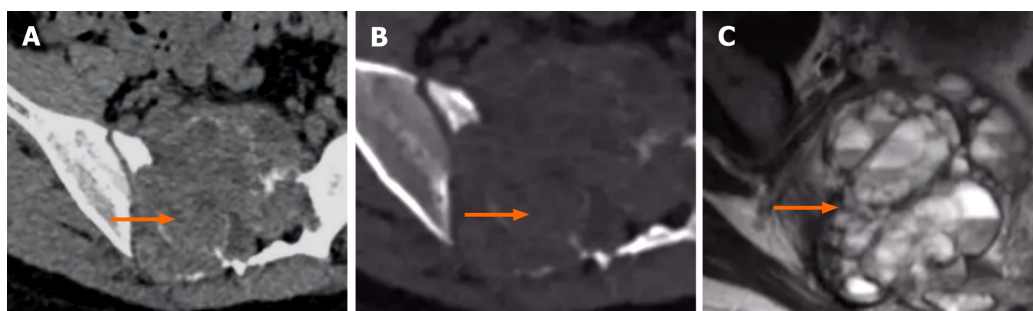


Figure 3 Female, 18 years old. The sacrum showed obvious expansive bone destruction. The pathological result was an aneurysmal bone cyst. A and B: Axial computed tomography showing multi-atrial osteolytic destruction. High density separations are visible in this lesion; C: Axial T2WI showing multiple fluid-fluid levels.

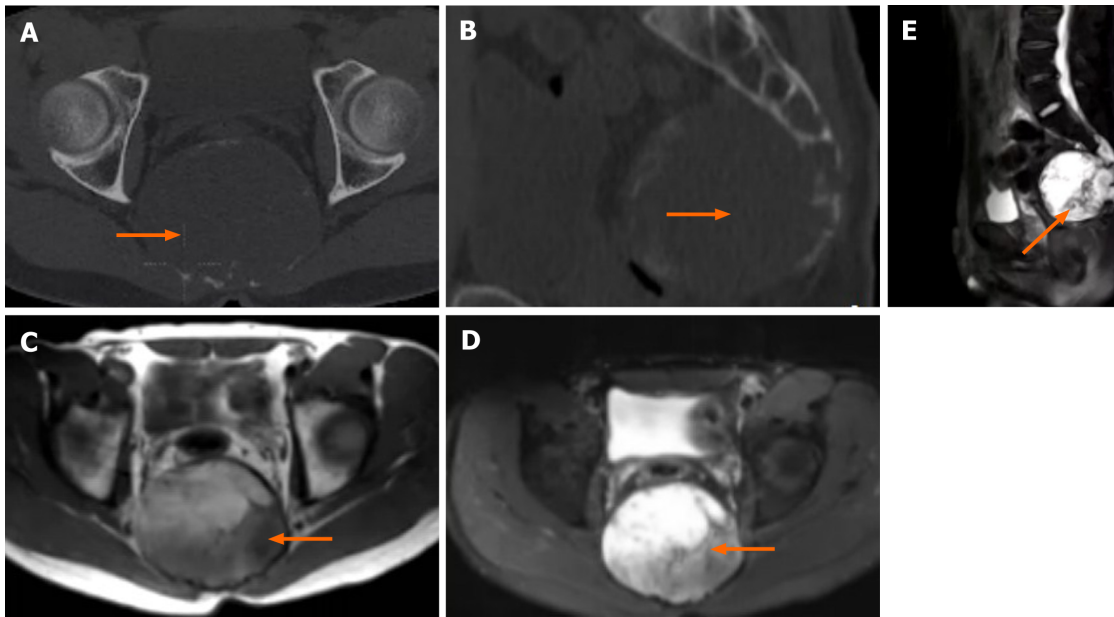


Figure 4 Male, 37 years old. The pathological result was chordoma. A and B: Axial and sagittal computed tomography showing osteolytic bone destruction of the sacrum with a large soft tissue mass. Destruction of the cortical bone is visible; C: Axial T1WI showing high and low signal intensity; D and E: FS T2WI showing iso-high mixed signal intensity.

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