

# World Journal of *Clinical Cases*

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## Giant monostotic osteofibrous dysplasia of the ilium: A case report and review of literature

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### Abstract

#### BACKGROUND

Osteofibrous dysplasia (OFD) is a developmental skeletal disorder, and cases with a giant affected area in the pelvis are rare.

#### CASE SUMMARY

In this case report, a 48-year-old man presented with a large tumor in the right iliac region that turned out to be OFD. The patient had rebound tenderness in his right hip. After radiography examination, magnetic resonance imaging examinations and some physical examination, extensive bone destruction in the right ilium was confirmed. Moreover, changes in bone mineral density and peripheral cortical bone sclerosis with surrounding soft tissue swelling were observed. Thus, this patient was considered to have giant monostotic OFD of the ilium. The tumor-related area was removed completely by surgery, and the remaining cavity was filled by artificial bones from the opposite ilium. According to the results of follow-up, the patient had normal flexion and extension activities of the right hip joint, and there was no evidence of recurrence of the tumor.

#### CONCLUSION

Suture of iliopsoas and gluteus medius muscle following focus curettage and bone grafting is a promising and effective method to treat giant OFD of the ilium. It is a feasible way to fill a large cavity after removing a lesion like the one in this case.

**Key words:** Osteofibrous dysplasia; Monostotic type; Giant tumor; Ilium; Case report

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**Core tip:** Osteofibrous dysplasia is a developmental skeletal disorder, and cases involving the pelvis with a large affected area are rare. This report is the first case, to our knowledge, of a 48-year-old man with a huge tumor in the right iliac that turned out to be osteofibrous dysplasia. With the assistance of computed tomography and magnetic resonance imaging, the tumor was completely removed, and the left empty cavity was reasonably filled by pulling and suturing nearby muscles and using some artificial bone.

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## INTRODUCTION

Osteofibrous dysplasia (OFD) is a developmental skeletal disorder and is characterized by substitution of normal bone with benign cellular fibrous connective tissue<sup>[1]</sup>. Currently, OFD can be classified as monostotic, polyostotic and McCune Albright syndrome<sup>[2]</sup>. Most cases of monostotic lesions present no significant symptoms and are often found incidentally on radiography taken for other symptoms<sup>[3]</sup>. Monostotic fibrous dysplasia mainly affects patients in their third decade of life<sup>[4]</sup>. Generally, monostotic OFD involves the ribs, the proximal femur, and craniofacial bones<sup>[5]</sup>. Cases involving the pelvis are uncommon, and cases with a large affected area are even rarer in clinics. Here, we report one patient with giant monostotic OFD of the ilium who was treated by tumor curettage and bone grafting. In the surgical procedure, we sutured the iliopsoas and gluteus medius muscle together and filled the remaining empty cavity to reduce the dead cavity volume and achieve a good outcome. We report here the diagnosis and treatment of giant OFD of the ilium.

## CASE PRESENTATION

### Chief complaints

A 48-year-old man who was diagnosed with a right iliac tumor more than 2 mo ago during a general physical examination.

### History of present illness

The use of painkillers, like aspirin, did not alleviate his discomfort in the right hip. Radiography examination showed giant bone destruction in the right ilium (Figure 1A).

### History of past illness

Past and family medical history was unremarkable.

### Physical examination

On physical examination, the patient had deep tenderness in the right hip, and the flexion and extension activities of the bilateral hip joints were normal.

### Laboratory testing

The results of chest radiography examination and other laboratory examinations were all normal.

### Imaging examination

Subsequent computed tomography (CT) and magnetic resonance imaging (MRI) examinations indicated extensive bone destruction of the right ilium, with a size of about 10 cm × 5 cm × 5 cm. Bone mineral density was changed in this area, and peripheral cortical bone sclerosis with surrounding soft tissue swelling was noted (Figures 1 and 2).

## FINAL DIAGNOSIS

Postoperative pathological examination confirmed the diagnosis of OFD (Figure 3).

## TREATMENT

Intraoperative findings showed that the bone cortex in front of the focus in the right iliac bone was thin. A bone knife was used to chisel open the medial thin bone cortex, and the gray-looking tumor-like tissue was visualized. The texture was tenacious; its boundary was clear, without invasion of surrounding soft tissues. After the tumor was completely curetted, there remained a giant empty cavity. There was an even larger empty cavity after the giant empty cavity was filled with bone harvested from the opposite ilium and 20 g artificial bone. During the operation, a bone knife was used to chisel open a part of the external iliac plate in the thin bone cortex at the posterior-lateral side of the empty cavity. After the window was opened, parts of muscle bellies of the anterior iliopsoas and posterior gluteus medius muscle were pulled together and sutured to fill up the empty cavity.

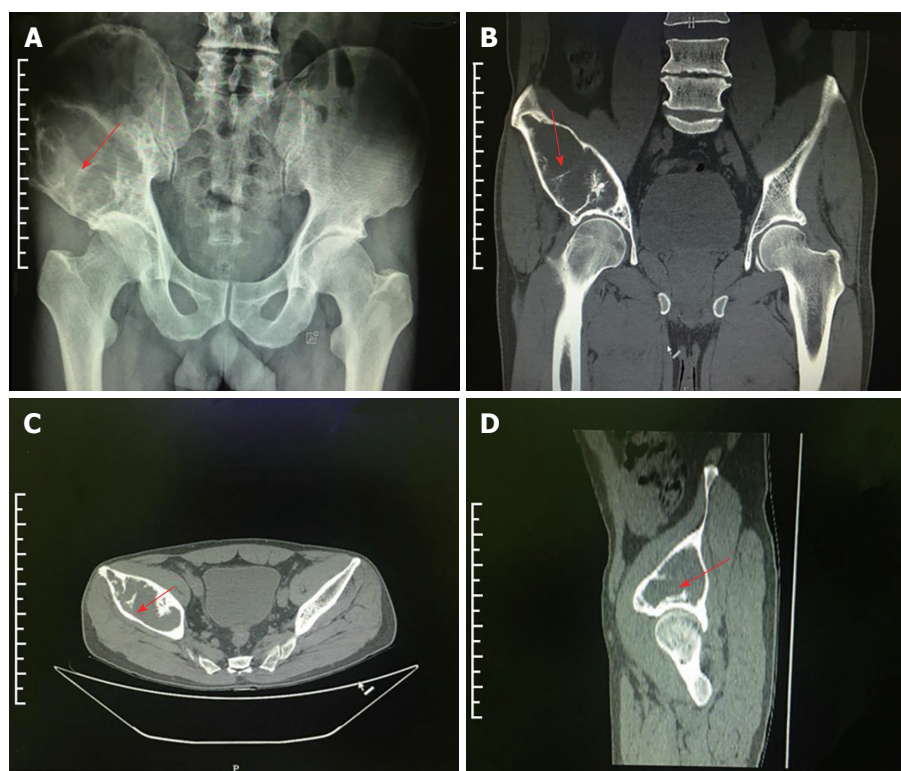
## OUTCOME AND FOLLOW-UP

Post-operative radiography reexamination (Figure 4) showed that the artificial bone and autogenous bone in the weight-bearing area of the acetabulum top were in place, and the coverage was satisfactory.

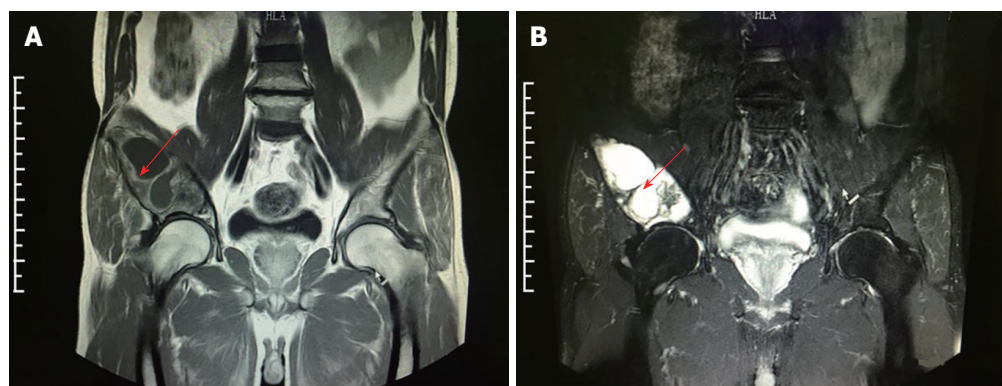
A follow-up at 6 mo showed that the visual analogous scale pain score was 0, and the patient had normal flexion and extension activities of the right hip joint. There was no evidence of recurrence of the tumor.

## DISCUSSION

In 1966, the lesion described in this case was named



**Figure 1 Radiography examination results.** A: Extensive bone destruction of the right ilium on radiography examination, with a size of about 10 cm × 5 cm × 5 cm, and bone density also changed; B: Computed tomography shows osteosclerosis in the surrounding area of the ilium tumor without significant periosteal reaction in the coronary site; C: Trans-sectional site; D: Sagittal position (arrow indicates the area of lesion).



**Figure 2 Magnetic resonance imaging examination results.** A: Extensive bone destruction in right ilium. T1w suggests low-density change; B: T2w suggests high-density change, with surrounding soft issue swelling (arrow indicates the area of lesion).

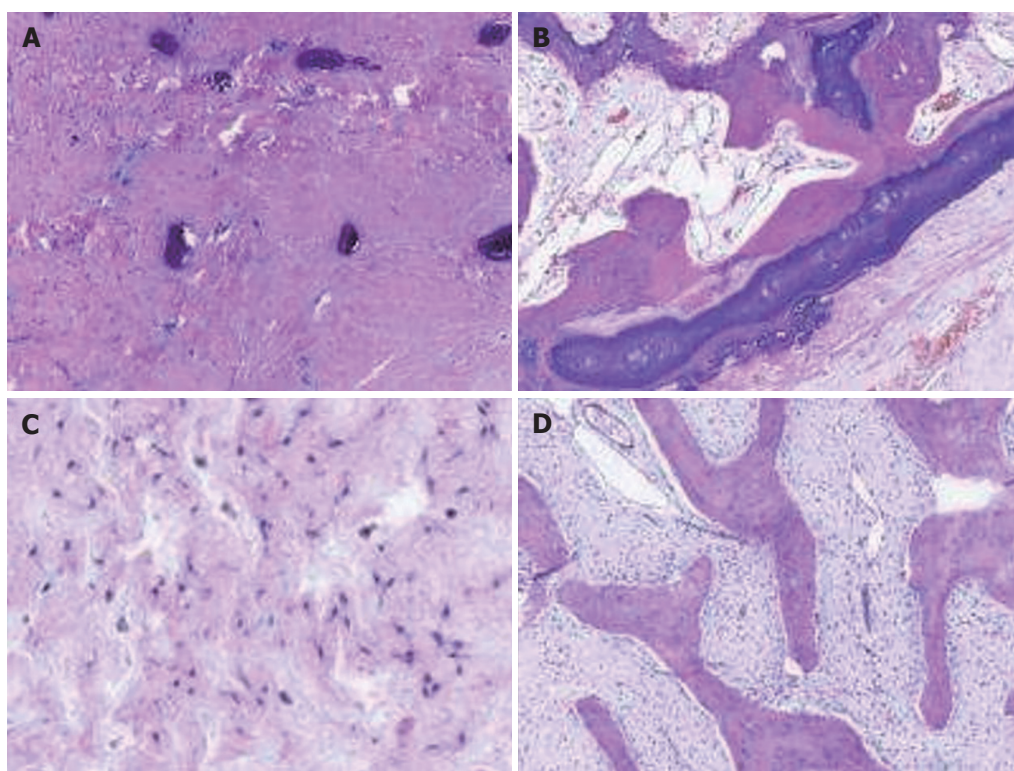
“ossifying fibroma” by Kempson<sup>[6]</sup>. In 1976, OFD was commendably categorized by Campanacci<sup>[7]</sup>. Subsequently, Campanacci and Laus<sup>[8]</sup> reported 35 cases from their facility; and 22 additional cases were also reviewed in the literature. OFD is a kind of fibro-osseous process that is commonly found in the diaphysis of the tibia. According to a series of 80 OFD cases reported by Park *et al.*<sup>[9]</sup>, 77 were tibia-related and only three appeared in the fibula. Moreover, nine involved of both the tibia and fibula on the ipsilateral side. In other case reports, the ulna and the radius were also common sites of involvement.

OFD is a kind of benign deformity-inducing fibro-

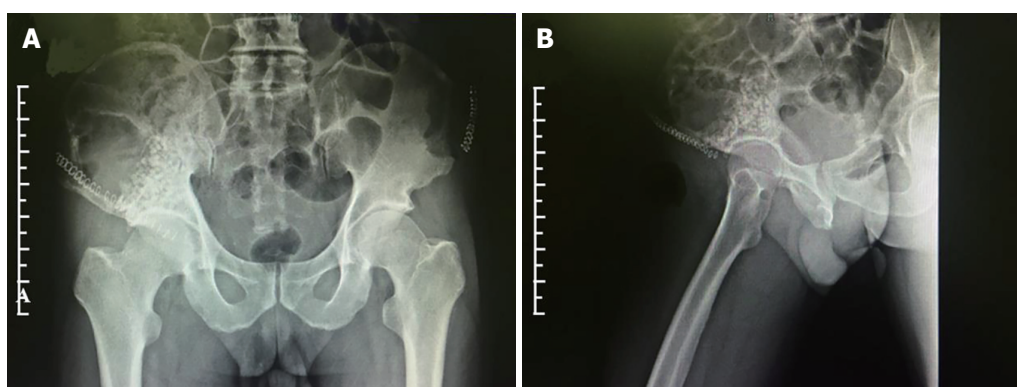
osseous lesion that usually occurs during childhood and often involves the mid-shaft of the tibia with or without involvement of the fibula. OFD is regarded as a common and benign skeletal disorder<sup>[10]</sup>. OFD appears generally as a localized mass, with the potential of pseudoarthrosis caused by bowed tibia, with or without pain. As the patient feels no special discomfort, misdiagnosis and occasional pathological fracture may occur<sup>[11]</sup>. The case reported here is extremely rare, not only because the large tumor was found in the right iliac of a middle-aged male but also because this tumor turned out to be OFD.

In immunohistochemical studies, single or strands





**Figure 3 Pathological reports.** A: Calcified woven bone can be seen (HE; 40 ×); B: No osteoblast and osteoclast hyperplasia can be seen (HE; 40 ×); C: It shows Fibroblast and Mucoid matrix (HE; 40 ×); D: It shows fibro-osseous lesions, with discontinuity of the trabecular bone and calcification (HE; 40 ×). HE: Hematoxylin-eosin.



**Figure 4 Postoperative radiography examinations.** A and B: Artificial bone and autogenous bone in the weight-bearing zone of top acetabular were in place with satisfactory coverage.

of keratin-positive cells are found in most of cases of OFD. In a series of immunohistochemical studies, cytokeratin-positive cells were identified in 80% of 85 OFD patients<sup>[12-15]</sup>. According to a case report by Kahn<sup>[16]</sup>, isolated cytokeratin-positive cells were distinguishable as mast cells and not epithelial cells in corresponding hematoxylin and eosin and Giemsa stained preparations. This discovery aided an issue concerning the histogenesis of all isolated cytokeratin-positive cells in previous studies. The OFD in long bones should not be confused with the entity gnathic ossifying fibroma with well-circumscribed mass that occurs in jaw. Additionally, the expression of cytokeratin (CK)

19 was demonstrated in OFD, whereas the expression of CK8 and CK18 was negative<sup>[15]</sup>. Similar findings, including a high incidence of CK1 positivity and a basal cell phenotype, were reported for both entities<sup>[17]</sup>. Furthermore, Bovée *et al.*<sup>[18]</sup> demonstrated that epidermal and fibroblast growth factor type 2 and its receptor were expressed in OFD. In this study, related laboratory diagnosis was performed: complete blood count and electrolytes were normal, and C-reactive protein and alkaline phosphatase were slightly elevated.

OFD is often identified with radiography examination, where it is typically manifested as glass-like change. The bone cortex grows expansively to become

thinner, and the boundary is clear without periosteal reaction<sup>[19]</sup>. In clinic, radiography examination combined with CT and MRI examinations is conducive to determine the surgical procedure<sup>[20]</sup>. In this case, the imaging examination combined with the medical history of the patient indicated that there was extensive bone destruction in the right ilium, the focus had a clear boundary, and the bone cortex was extensive and thin and showed a ground glass-like change, without significant periosteal reaction. The affected area was considered to be a benign tumor-like lesion, with a high possibility of it being OFD. The involving range of tumor was large, and a part of the focus was located in the non-weight-bearing area of the acetabulum top. Thus, it was necessary to carry out bone grafting to reduce damage to the functional structures in the weight-bearing area. Complete curettage of the tumor led to a large residual cavity, which could easily form a dead cavity and increase the risk of infection. Therefore, a part of the external iliac plate in the thin cortical bone was chiseled open, and the anterior iliopsoas and posterior gluteus medius muscle were pulled together and sutured. This method not only effectively reduces the dead cavity, but it also minimizes the surgical trauma and does not affect the overall stability of the pelvic ring, making it conducive to postoperative recovery.

At present, the major treatment strategies for OFD are conservative treatment and surgical treatment<sup>[21]</sup>. Although it is a benign lesion with no symptoms, it is progressive and may lead to severe defects and lesions in bone and skin<sup>[2]</sup>. The surgical methods for OFD mainly include focus curettage and bone grafting combined with or without external fixation<sup>[22-25]</sup>. Curettage is a common treatment method for benign lesions, aggressive lesions, some cartilaginous malignant lesions and bone metastases<sup>[26]</sup>. However, the bone cavity created after curettage often needs to be filled with filling substances, such as acrylic cement or bone grafts, to guarantee its mechanical stability<sup>[27-29]</sup>. In this case, the allogeneic bone mixed with autogeneous bone was implanted in the weight-bearing area of the acetabulum. There is a large amount of cancellous substance in the ilium, and it has a good osteogenic activity. Compared to other materials, it has a faster creeping replacement. In addition, under the condition that the stability of the pelvic ring is unaffected, simple windowing in the external iliac plate and suturing between muscles can reduce the dead cavity and avoid internal fixation. Therefore, this method not only reduces the operation time and the operating difficulty but also lowers the cost for the patients.

In conclusion, the case recorded in this report was rare because most OFD occur in the mid-shaft of tibia with or without involvement of the fibula. Furthermore, the surgical methods utilized were effective without influencing the stability of pelvic ring. Moreover, the cavity generated by curettage was treated by filling

up with mixture of autograft and allograft. Finally, the patient had noticeable recovery, specifically as normal flexion and extension activities of the right hip joint were observed in the patient without any evidence of tumor recurrence. Therefore, it is reasonable to believe that performing focus curettage and bone grafting with suture of iliopsoas and gluteus medius muscle will provide a promising and effective method in treating giant OFD of the ilium. This method not only reduces operation time and the operating difficulty but also lowers the cost for the patients. To our limited knowledge, it is the first case report to describe a large OFD tumor in the right iliac, its removal, and the filling of the empty cavity created after tumor removal by surrounding muscles and artificial bone. A limitation in this case is that the long-term efficacy needs further observation in the following period.

## EXPERIENCES AND LESSONS

Some experiences and lessons were shared in this case, specifically: (1) combination of MRI and CT examinations to make precise diagnosis; (2) the allogeneic bone mixed with autogeneous bone was implanted in the weight-bearing area of the acetabulum to fill the hole caused by surgery; and (3) parts of muscle bellies of the anterior iliopsoas and posterior gluteus medius muscle were pulled together and sutured to fill up the empty cavity. Above-mentioned processes in this case not only reduce the operation time and the operating difficulty but also avoid the utilization of internal fixation and lower the expense.

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