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Case Report

Giant osteochondroma of ilium: a case report and literature review

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Abstract: Osteochondroma is one of the most common benign bone tumors. It usually grows on the metaphysis of long bones and rarely develops in bones of scapula, feet, hands, and pelvis. The management of this disease is en-bloc excision of the tumor. We present a 45-year-old female subject, who complained of having found a mass on the right hip for more than 20 years which was diagnosed to be osteochondroma on X-ray, computed tomography (CT) and 3-dimensional (3-D) reconstruction. We performed en-bloc excision for the patient. Pathologic examination of surgical specimen confirmed the diagnosis of osteochondroma. The patient made a complete recovery and there has been no recurrence after one year of follow-up. Osteochondroma usually represents an osteo-cartilaginous aberrant overgrowth of normal epiphyseal growth plates. The disease has a slow onset and a long history. X-rays and CT scans are sufficient for diagnosis before surgery and the final diagnosis should based on pathology. Differential diagnosis includes chondrosarcoma or other neoplasms. When osteochondroma causes pain, compression of peripheral nerves, or continuous growth and other clinical symptoms, en-bloc excision of the tumor is needed. Better recognition and more comprehensive evaluation of these rare cases should be highlighted to avoid misdiagnosis during our clinical practice.

Keywords: Osteochondroma, ilium, en-bloc excision

Background

Osteochondroma is one of the most common benign bone tumors [1]. This disease accounts for approximately 15% of all bone tumors according to Pongkripetch and Sirikulchayanonta's series reports and 45.3% of benign tumors according to Barbosa et al.'s reports [2, 3]. Osteochondroma usually occurs at the metaphysis of distal femur, proximal radius, proximal tibia, and proximal fibula, presenting as a firm and immovable mass arising near the end of the long bone [4]. It rarely develops in bones like scapula, feet, hands, and pelvis. Pelvic osteochondroma has been reported with an incidence of 5% of all osteochondromas [5]. The management of this disease is en-bloc excision of the tumor. Indications for excision are cosmetic deformity, pressure symptoms on adjacent tissues, or malignant transformation to chondrosarcoma although this is rare [6].

In the present study, we treated a middle-aged female subject with giant osteochondroma originating from the ilium. We also reviewed previous literature to make a comprehensive summary about osteochondromas originating from the ilium.

Case presentation

A 45-year-old female subject, who complained of having had a mass on the right hip for more than 20 years, came to our Trauma Center in May 2019. She told us that the mass had been first noted more than 20 years before, with a gradual increase in size during the subsequent period. She never felt pain from the mass during this time. No associated medical co-morbidities or family history with similar complaints were detected.

Physical examination showed a size about 18 cm times 15 cm times 10 cm mass in front of

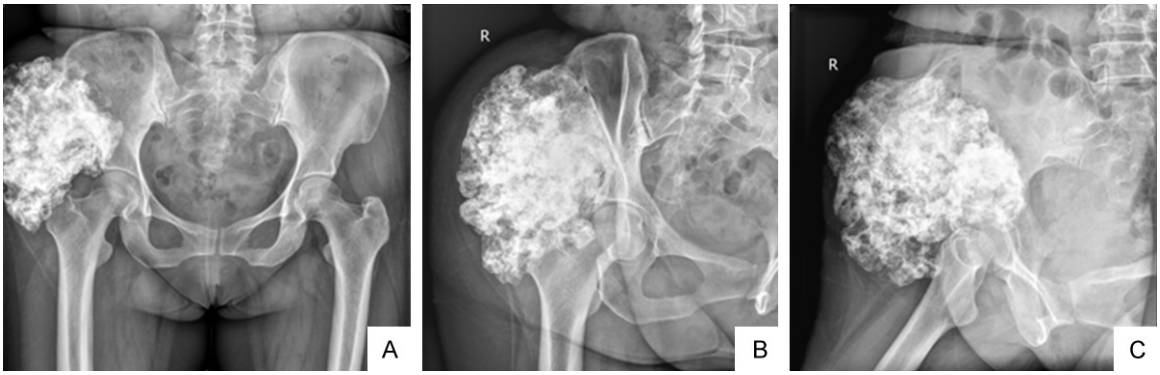

Figure 1. X-rays including the A-P view of pelvis (A), the oblique view of ilium (B) and the obturator view of pelvis (C) all showed a mass of popcorn-like high-density shadows with clear boundaries on the right side of the ilium.

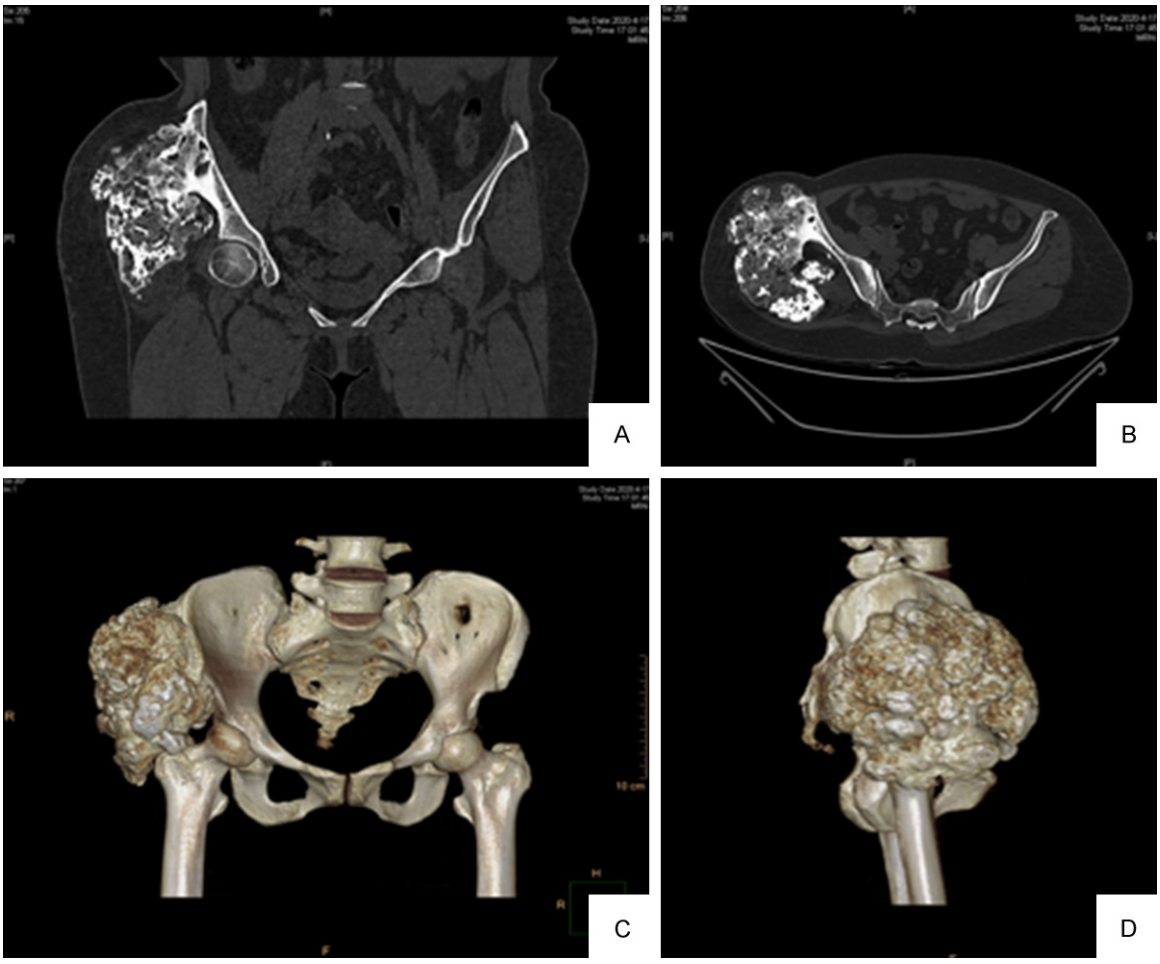

Figure 2. Transverse section (A) and Coronal section (B) of CT scan combined with 3D reconstruction (C, D) showed a pedunculated exostotic mass about 15 × 15 × 8 cm in size.

the right hip. The texture of the mass was hard and it was immobile but was not adherent to the overlying skin. The movement of bilateral hip joints was unrestricted. The blood circulation and sensation of the right lower limb were normal. The Anteroposterior (A-P) view pelvic X-ray showed, on the right side of the ilium, a popcorn-like high-density mass with clear

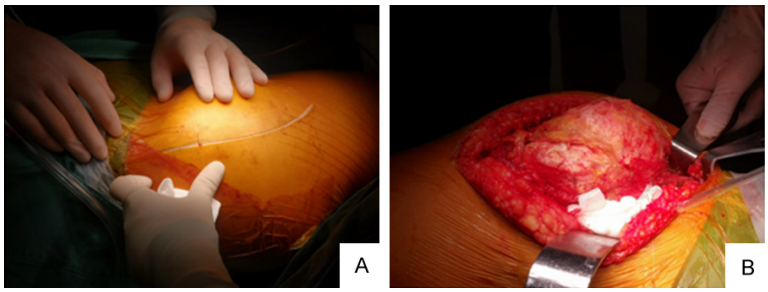

Figure 3. Incision over the surface of the mass (A) and exposure of the pedicle and basement of the mass (B).

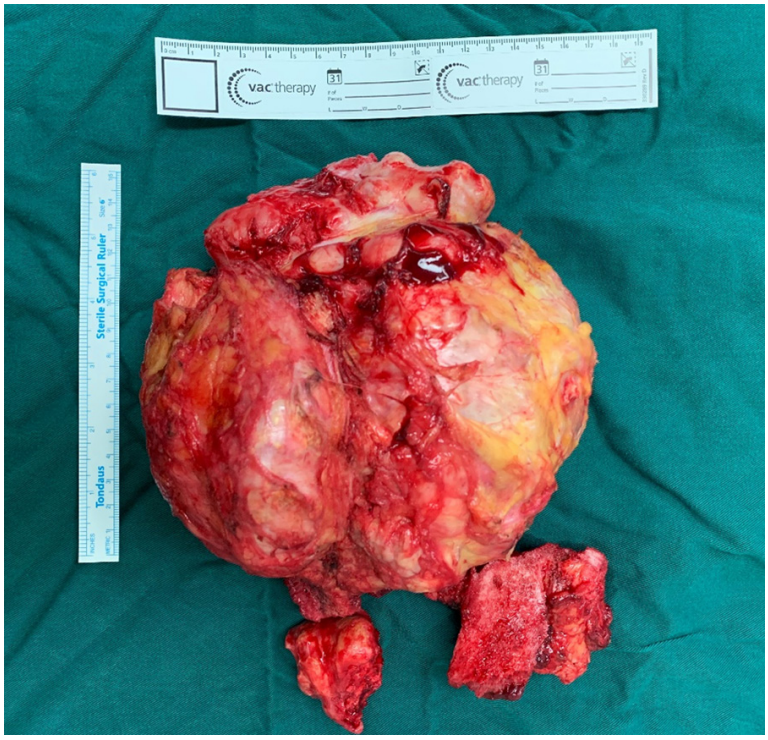

Figure 4. Gross view of the surgical specimen obtained by en-bloc resection showed a 16 × 15 × 10 cm cartilaginous bone mass.

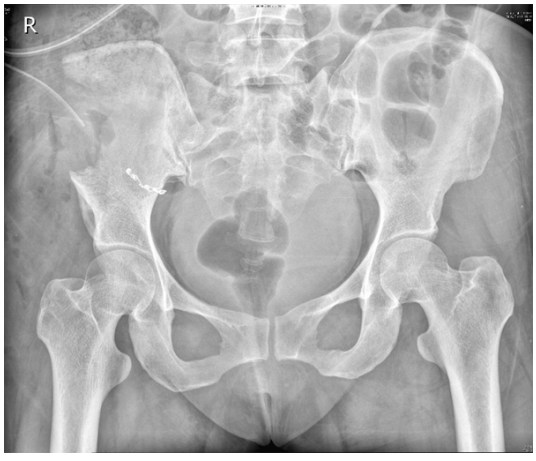

Figure 5. X-ray of the orthographic view of the pelvis after surgery showed the osteochondroma was completely removed with part of the ilium lacking.

boundaries (**Figure 1**). Computed tomography (CT) scan and 3-dimensional (3-D) reconstruction were performed to confirm its anatomic position. From CT scan and 3-D reconstruction, irregular masses of increased density could be seen attached to the right ilium, and popcorn-like high density shadows could be seen inside (**Figure 2**). Furthermore, we have also observed that the stem of the bone mass is larger in 3-D reconstruction than in CT transverse plane (**Figure 2C**, **2D**). According to these imaging findings, the patient was diagnosed with osteochondroma.

We performed en-bloc excision of the osteochondroma. The incision was made over the mass. Then the muscles were separated from the surface of the mass and the iliac fossa. Next, the pedicle and base of the mass were explored. Then, the whole mass including part of the iliac crest was resected. After hemostasis, a drainage tube was put in the incision and the muscle from the iliac crest was repaired. The incision was sutured finally (**Figure 3**). The real size of the osteochondroma was measured and documented as 16 cm × 15 cm × 10 cm (**Figure 4**).

An X-ray during the operation confirmed complete resection of the osteochondroma. The A-P view X-ray of the pelvis after operation showed that the osteochondroma was completely removed with part of the iliac crest (**Figure 5**).

Pathologic examination after the operation found a cartilage cap of variable thickness in the specimen, which confirmed the diagnosis of osteochondroma (**Figure 6**).

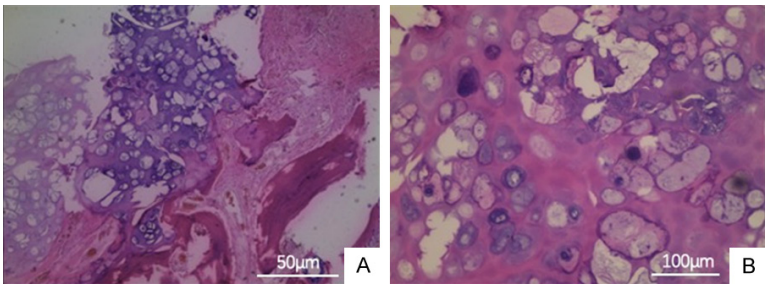

Figure 6. Low-power appearance (A) showed a cartilage cap of variable thickness, which was made up of abundant cellular hyaline cartilage and chondrocytes (original magnification 40 ×; hematoxylin-eosin stain). High-power view (B) showed highly active enchondral ossification (original magnification 100 ×; hematoxylin-eosin stain).

The drainage tube was removed on the third day after the operation. She was discharged six days after operation. Two weeks after discharge, she was evaluated in the outpatient department and had her sutures removed. Subsequently she was started on physiotherapy with passive and active exercises. The patient was regularly followed up, and there was no recurrence of the osteochondroma one year after operation.

Literature review

We made a review of cases of osteochondroma originating from the pelvis. This literature review summarized all cases which met the requirements from OVID (January 1990 to September, 2020) and PubMed. We screened the title and abstract first, and then read the full text to collect all useful information. The included cases had a comprehensive description about occurrence, symptoms, physical examination, radiologic characteristics, treatment, and follow-up results. Finally, 9 studies in English were included in the present review [6-14]. These are summarized in the **Table 1**.

All of these cases were osteochondroma originating from the pelvis in adults, with a average age of 31.33±8.90 (range 18 to 46) years. Among these 9 patients, 6 were male [6, 9, 11-14] and 3 were female [7, 8, 10]. The disease duration varied from 5 months to 10 years. Slowly progressive swelling over time was commonly the first noted symptom [6, 7]. Pain was present in most patients [8-10, 12, 14]. The limitation of range of motion of hip joint was another symptom, and was present in 2 patients [9, 10]. When doing a physical examination, the bony masses were all firm and immobile. The tumor size varied (maximum 8 cm × 8 cm × 5 cm; minimum 3.3 cm × 2.8 cm × 2.0 cm). All patients underwent successful compete tumor excision. Among these patients, only 1 case was followed up more than 2 years. No recurrence was noted in all cases.

Discussion and conclusion

Osteochondroma is a common benign bone tumor, but it is unusual that osteochondroma originates from the ilium region, let alone develops into such a huge tumor as in our case. When osteochondroma causes pain, compression of peripheral nerves or has continuous growth and other clinical symptoms, en-bloc excision of the tumor is needed.

Osteochondroma usually represents an osteocartilaginous aberrant growth of normal epiphyseal growth plates [1]. The pathologic structure includes substrate, bone, hood of transparent cartilage and integument of fiber. Frequent chondroblasts in the deep layer of integument, produce transparent cartilage. Cartilage can be converted into tumor-like spongy bone by continuing calcification and ossification [15]. The number of osteochondromas may be solitary or multiple [16]. Multiple osteochondromas are always associated with hereditary multiple exostoses (HME), an autosomal dominant disorder [17-19]. Complications occur more frequently with HME, which include deformity, fractures, vascular impairment, formation of pouches, malignant transformation, and neurological sequelae [8].

This disease has a slow onset and a long history similar to the case we reported. Early symptoms are not obvious, and they increase and protrude with the growth and development of the human body. X-rays and CT scans are diagnostic in most cases and allow anatomic delineation of the lesion [20]. Osteochondroma typically presents as a pedicle or sessile bone-like protrusion on imaging. The cancellous bone and cortical bone are connected to normal bone. The shadow of the cartilage can be seen at the top of the tumor, and irregular calcifica-

Table 1. Summary of included cases of pelvis osteochondroma

Case	Patient Characteristics	Symptoms	Physical Examination Findings	Size and Location	Pathologic	Treatment and Results
Herode P et al [6], 2015	24-y-old Male with a disease course of 2 y	Swelling in the right side inguinal region; irritation and pain on walking and squatting	Not tender nor mobile; fixed to underlying bone	8*4.6*5 cm; over the right inguinal region; on ventral surface of the right superior pubic ramus	Bony trabeculae along with cartilage with no evidence of dysplasia	Surgical excision; ilioinguinal approach
Ratra R et al [7], 2020	18-y-old Female with a disease course of 5 y	Swelling	The swelling was fixed to the underlying bone	3*5 cm; in the groin at the pubic tubercle of the right side	Non-malignant osteochondroma	Extended ilio-inguinal approach; no recurrence during 1-y follow-up period
Moraes FB et al [8], 2014	42-y-old Female with a disease course of 1 y	Sciatic pain in her left leg; worsening of the pain with hip movement; after using anti-inflammatory drugs and opioids, the pain was not improved	Paresthesia on the anterolateral face of the left leg and foot; diminished motor strength (grade 4) on dorsiflexion of the foot (L4), extension of the hallux (L5) and plantar flexion (S1)	4 cm; in bone of the left ischium, along the path of the sciatic nerve	Osteochondroma	Surgical excision; Kocher-Langerbeck route; no recurrence during 2 months follow-up period
Gokkus K et al [9], 2013	25-y-old Male with a disease course of 10 y	Limping on the right side. radiating pain and sciatalgia in the right extremity	Tenderness around the ischial tuberosity; Radiating pain and paresthesia at the ipsilateral lower extremity, and also weakness at the plantar flexion of the foot	The ischial ramus	Characteristic pattern of an osteochondroma which is covered by a thick cartilaginous cap formed with columnar arrangement of chondrocytes	Surgical excision; modified Gibson incision
Chun YS et al [10], 2016	32-y-old Female with a disease course of 10 y	Chronic right groin pain along with limited hip joint mobility	A mass with hard consistency was palpable in the right groin area; Snapping phenomenon with mild pain when her right hip was forced into a squatting position	3.3*2.8*2.0 cm; right ilium	Cartilaginous cap at the margin of the exostosis	Surgical excision
Zhu J et al [11], 2018	31-y-old Male with a disease course of 1 y	No pain and other discomfort	A single well-defined mass at the right ischium	8*8*5 cm; right ischium	Lobules of mature univaculated adipocytes and bony trabeculae, with scattered patchy islands of benign cartilage	Surgical excision
Zhang Y et al [12], 2017	36-y-old Male with a disease course of 5 months	Radiating pain and numbness in the lower left extremity; left-sided pelvic pain		Pelvis	An osteochondroma with a grossly irregular surface, actively proliferating chondrocytes	T-shape posterior-only approach
Wang WY et al [13], 2010	46-y-old Male with a disease course of 8 y	Decreased stream and dysuria		7*7.2*8.5 cm; symphysis pubis	Hyalinized fibrocollagenous tissue with evidence of cartilaginous metaplasia and calcification, consistent with an osteochondroma	Surgical excision; no recurrence during 4 y follow-up period
Julsrud ME et al [14], 1994	28-y-old Male with a disease course of several y	Right-buttock pain that was aggravated by running or sitting	His right pelvis was higher than his left when he stood	3.3*3.2 cm; right Ischium	Osteochondroma	Surgical excision

tion and/or ossification can be seen in the middle of the shadow. In our case, CT and 3-D reconstruction clearly showed the size and boundary of the osteochondroma, and also showed the degree of compression of the tumor to the surrounding ilium, which allows for the design of the operative plan.

According to the statistics, malignant transformation of osteochondroma is less than 1% [21]. Differential diagnosis from other neoplasms like chondrosarcoma is necessary. Chondrosarcoma can be primary or secondary to osteochondroma [22]. Osteochondroma with unparalleled continuous growth after skeletal maturity should raise the possibility of malignant chondrosarcoma [23, 24]. Chondrosarcoma on CT shows a predominantly soft-tissue mass with a heterogeneously mineralized center. There may be destruction of adjacent bone or abnormalities of adjacent soft tissues. A poorly defined outer cortical margin and bone destruction can also suggest malignancy [14]. In our case, we found the pedicle connecting the tumor to the ilium on CT, and there was no evidence of bone destruction and invasion of the surrounding tissue. Besides, the final results of the microscopic examination indicated that the chondrocytes grew actively and did not deteriorate.

In conclusion, osteochondroma rarely develops in bones like scapula, feet, hands and pelvis. It can also greatly impair the quality of life of the patients and result in some symptoms. Better recognition and more comprehensive evaluation for these rare cases should be highlighted to avoid misdiagnosis during our clinical practice.

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Disclosure of conflict of interest

None.

Abbreviations

CT, computed tomography; 3-D, 3-dimensional; HME, hereditary multiple exostoses; A-P, anteroposterior.

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