

Unusually Giant Solitary Osteochondroma of the Ilium: A Case Report with Review of Literature

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Learning Point of the Article:

Giant iliac osteochondroma is a rare but cosmetically disabling tumor and its timely diagnosis followed by en bloc excision warrants an immediate improvement in the quality of life.

Abstract

Introduction: Osteochondroma is the most common benign bone tumor where a chondrogenic lesion is derived from aberrant cartilage from the perichondral ring. Although it commonly arises from the growing ends of long bones, less commonly, it may arise from the scapula, pelvis, or vertebra.

Case Report: We encountered a 16-year-old male patient with a painless left pelvic solid mass for 3 years, which was suggestive of osteochondroma on X-ray and magnetic resonance imaging findings. Besides cosmetic issues, the main indication for surgery was the constant discomfort in wearing pants/shorts/belts. He underwent en bloc excision followed by a biopsy of the surgical specimen by two independent histopathologists confirming the tumor to be osteochondroma. He was followed up for 2 years with no signs of post-operative complications or recurrence. This case represents one of the very few reported so affecting the iliac wing, where the excision was performed before skeletal maturation. We also performed a review of the current literature on iliac wing osteochondroma to understand the tumor better, identify gaps in current knowledge, and suggest areas for future research.

Conclusion: Since one of the differential diagnoses includes secondary chondrosarcoma, which could be a rare progression of osteochondroma, early recognition and comprehensive evaluation of such unusual cases needs to be dealt with a high index of suspicion to avoid misdiagnosis and to provide effective treatment.

Keywords: En bloc excision, ilium, osteochondroma.

Introduction

Osteochondroma is the most common benign bone tumor accounting for 20 to 50% of all benign osseous tumors [1, 2]. However, the true incidence is difficult to predict as most cases are asymptomatic and do not seek medical attention due to painless mass. Osteochondromas may be solitary (85%) or multiple (15%). The multiple forms are an autosomal dominant

syndrome referred to as hereditary multiple exostosis or familial osteochondromatosis. It usually grows on the metaphysis toward the rising end of the long bones (femur, tibia, humerus, etc.) but may develop in flat bones (scapula, feet, and pelvis) [3]. Patients typically present between the ages of 10 and 30 with a slight predilection for the male gender. Continuity of the cortex and medulla of the tumor, along with the underlying bone, is a

Author's Photo Gallery



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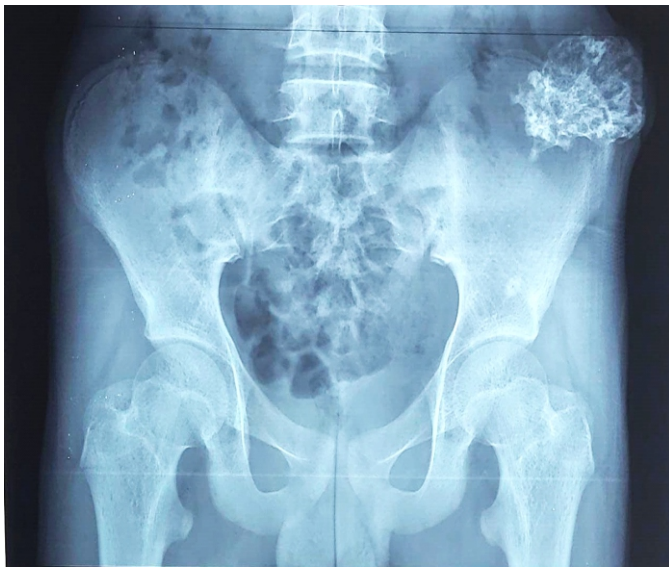


Figure 1: X-ray anteroposterior view of the pelvis with both hips showed a mass of popcorn-like high-density shadows with clear boundaries on the left side of the ilium.

pathognomonic feature in establishing the diagnosis [4]. Pelvic osteochondroma has been reported with an incidence of 5% of

all osteochondromas [1]. The treatment of this tumor is en bloc excision for cosmetic deformity or pressure symptoms [5].

Case Report

We encountered a 16-year-old male patient with a painless left pelvic solid mass for 3 years with a gradual increase in size during the subsequent period. It was suggestive of osteochondroma in radiology. We used magnetic resonance imaging (MRI) to characterize the lesion definitively. There were no associated medical comorbidities or positive family history for a similar problem. On physical examination, bony-hard, non-tender, and fixed but non-adherent mass from overlying skin measuring approximately 10 cm × 7 cm × 6 cm was noted over the anterolateral left iliac wing. Local examinations were negative for lymphadenopathy, dilated veins, or skin changes on or around the mass. Spine and left hip range of motions was full and free with normal distal neurovascularity of the lower extremity and normal gait.

Pelvis with both hips (PBH) anteroposterior (A-P) view radiogram demonstrated a popcorn-like high-density mass with well-defined, clear boundaries on the left side of the outer

cortex of the iliac wing (Fig. 1). The iliac crest physis is unfused (Risser stage 3). MRI was performed to confirm the continuity of the cortex/medulla with adjacent bone, the size of the stem, and surrounding soft tissue (Fig. 2). Furthermore, we have also observed that the stem connecting the tumor to the ilium with no evidence of bone destruction and invasion of the surrounding tissue. According to these imaging findings, the patient was diagnosed with osteochondroma. We did not perform a CT scan or 3D model printing as it was not required for surgical planning due to the relatively safe location of the tumor.

Surgical technique

The patient was planned for an en bloc excision biopsy of the tumor after all the necessary basic investigation, pre-anesthetic fitness, and consent. Spinal anesthesia was given and the patient was lying down in the right lateral position. Painting and drapping were done under aseptic precautions. The operating surgeon and assistant were both standing at the patient's back/posterior side. Tumor boundaries were marked in circular manner by a sterile surface marker to determine the extent and direction of the incision (Fig. 3). A central curvilinear skin incision (11 cm) was made parallel to the long axis over the mass. Fascia-like

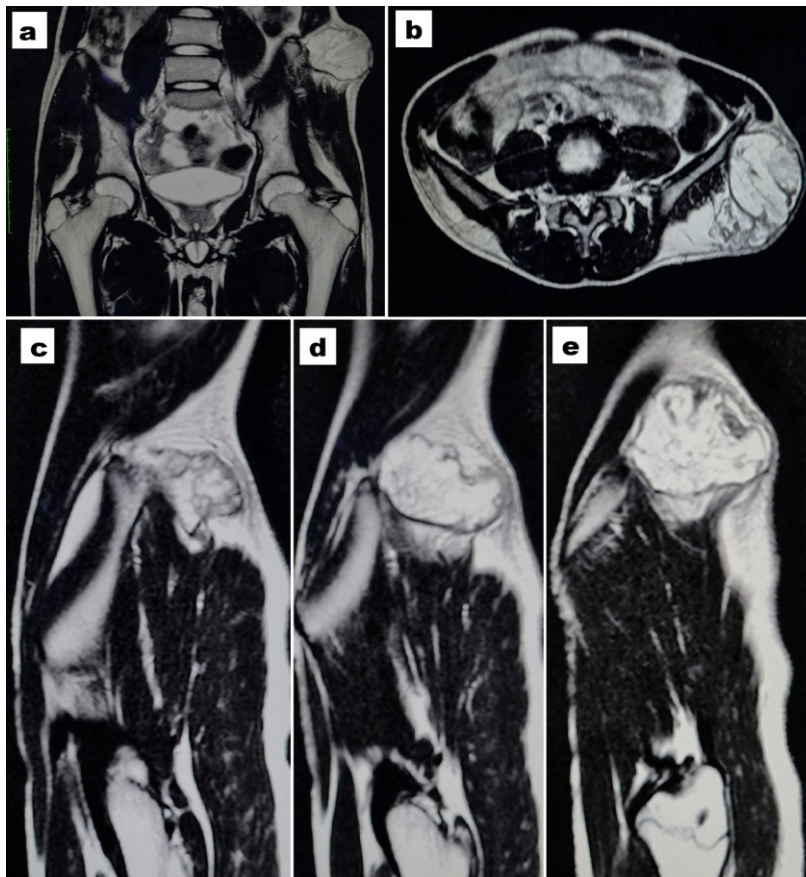


Figure 2: MRI showing pedunculated exostosis mass arising from the outer table of the ilium (a) coronal view, (b) axial view, and (c-e) sequential sagittal view images.



Figure 3: Surface marking of the tumor site before incision.

covering of the tumor was removed until the solid cartilaginous part was exposed. Then, the muscles were separated by a periosteum elevator from the basal surface of the mass and the outer cortex of the iliac wing until the tumor pedicle was exposed (Fig. 4a). Then, the whole mass was resected through the base of the pedicle over the posterior iliac fossa with the help of 25 mm curved osteotomes (Fig. 4b). A normal periosteal margin of around 0.5 cm was also removed from the area surrounding the base of the pedicle (Fig. 4c). We found a fleshy cartilaginous cap of variable thickness overlying the mature bone in the gross specimen highly suggestive of osteochondroma, which ruled out any need for a frozen section

or wide excision. We did not dissect the iliac crest and thus, avoided the risk of physal damage. The hemostasis of oozing bone was achieved by bone wax over the resection area (Fig. 4d). No drain tube was kept considering the minimal blood loss. The actual size of the osteochondroma was measured and documented as 8.5 cm × 6 cm × 5 cm (Fig. 5). C-arm X-ray imaging was used during the surgery to confirm complete resection of the osteochondroma. Finally, the subcutaneous tissue and skin were sutured in layers (Fig. 6). The immediate post-operative A-P view confirmed that the osteochondroma was completely resected (Fig. 7).

Post-operative protocol

The excision biopsy specimens were sent to laboratories in diluted formalin. Histopathologic examination was performed by two experienced

independent oncohistopathologists (one was blinded about clinical and radiological findings, and the other was non-blinded) to confirm the osteochondroma. Mature hyaline cartilage with fibrous perichondrial covering over the mature bone without any malignancy confirmed a pedunculated osteochondroma (Fig. 8). The patient was discharged on the 3rd post-operative day along with an active and passive physiotherapy regimen. The sutures were removed after 2 weeks after surgery. The patient was followed up telephonically for 2 years, and there were no clinical signs of recurrence of the osteochondroma after the surgery. We could not get the follow-up X-ray to rule out the radiological signs of recurrence,

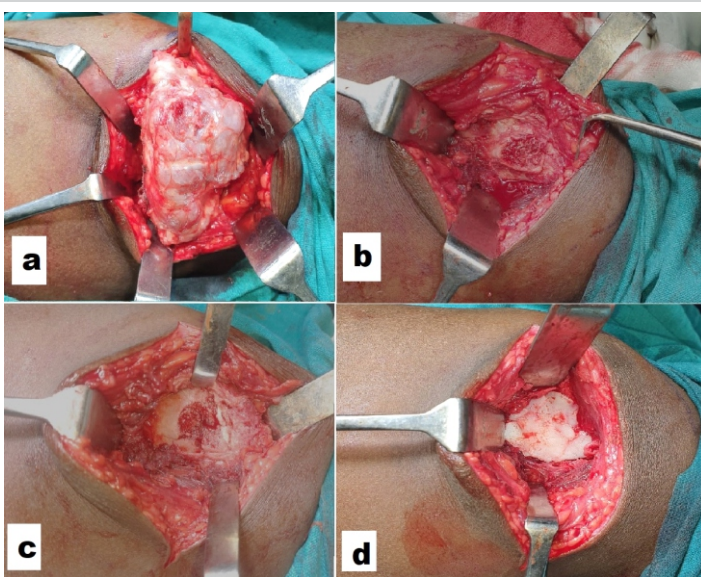


Figure 4: Surgical steps for en bloc excision of osteochondroma (a) completely exposed tumor, (b) tumor stem after excision through curved osteotomes, (c) exposed base of the stem after safe periosteal outer table margin removed, and (d) bone wax applied over the oozing bone.



Figure 5: Gross view of surgical specimen (9 cm × 6 cm × 5 cm) obtained by en bloc resection showing a cartilaginous bone mass with an irregular surface.



Figure 6: Curvilinear incision (11 cm) closed with ethilon 2.0 suture.



Figure 7: Post-operative X-ray of the pelvis anteroposterior view with both hips showing complete resection of the osteochondroma.

however, the risk of further growth of the tumor is minimal considering that patient's age is very close to physal fusion.

Literature review

As far as our knowledge is concerned, our's is the first study that performed a review solely on osteochondroma arising from the ilium. We performed literature search using Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. We conducted the literature search through PubMed, EMBASE, Scopus, and Web of Science using the terms "osteochondroma," "exostosis," "pelvis," "ilium," and "solitary" with various spellings and Boolean logical operator. We reviewed 95 abstracts and included cases after reading the

full text with a comprehensive description of the occurrence, symptoms, physical examination, radiologic characteristics, treatment, and follow-up results. We excluded duplicate articles, other language articles (where the English translation is unavailable), those discussing pelvic osteochondroma other than iliac wing (i.e., pubic and ischial region), cases of multiple exostosis, and osteosarcoma cases. A total of 21 cases (including ours) are summarized in Table 1. All of these cases were osteochondroma originating from the iliac wing with a mean age of 26 (range 11–74) years. Among these 21 cases, 18 were male and 3 were female. The disease duration varied from a few months to many years. Slowly progressive, painless swelling

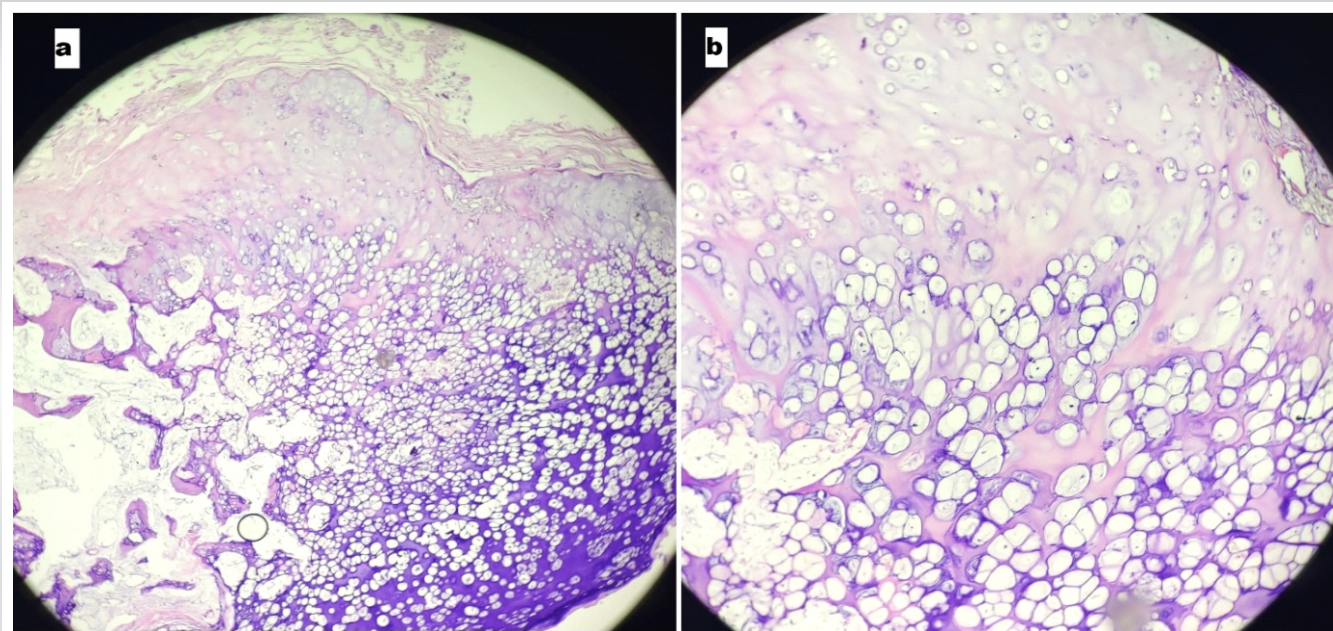


Figure 8: Histopathological appearances in hematoxylin-eosin stain (a) Low-power ($\times 20$) view: variable thickness cartilage cap made up of abundant cellular hyaline cartilage and chondrocytes; and (b) high-power ($\times 100$) view: highly active enchondral ossification.

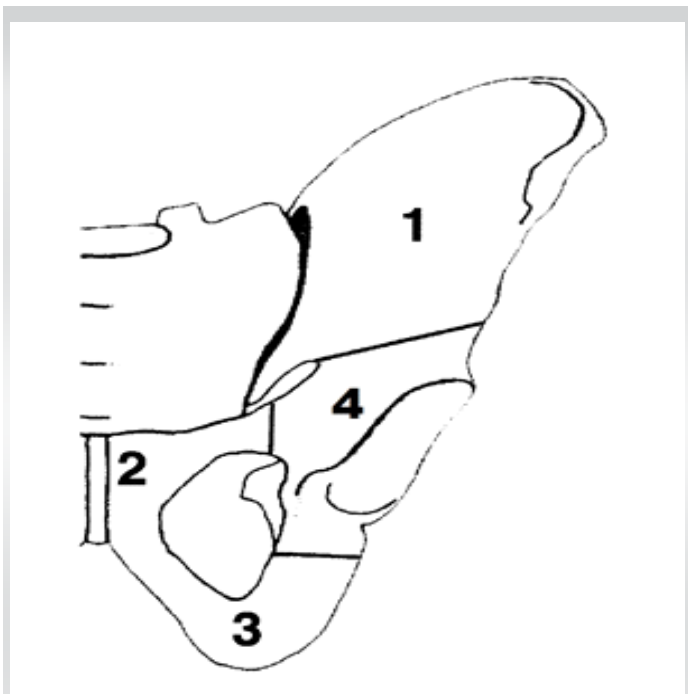


Figure 9: Schematic illustration of the different zones defining pelvic tumor location. Zone 1: ilium; zone 2: pubis; zone 3: ischium; and zone 4: periacetabular region.

over time was the most common symptom. On physical examination, the non-tender bony hard mass was the most common positive finding (18 out of 21). Osteochondroma was arised from the “inner cortex” (unlike the crest or outer cortex) of ilium in two out of 21 cases. The tumor size varied widely (minimum 2.3 cm × 1.4 cm; maximum 16 cm × 15 cm × 10 cm). All (19 out of 21) except two patients (one incidentally diagnosed and one with lost follow-up) underwent successful compete en bloc tumor excision. Two cases out of 19 had to undergo extensive reconstruction. One had L4-S1 transforaminal lumbar interbody fusions and another underwent abdominopelvic reconstruction. No recurrence was encountered in any of the cases.

Discussion

Osteochondroma is considered as an osteocartilaginous aberrant growth of normal physeal growth plates and, hence, believed to be a developmental lesion [6]. This theory is supported by the cases of osteochondroma reported following trauma or irradiation. However, recent research suggests that

S. No.	Case	Age/gender	Symptoms and duration	Physical examination	Size and location in ilium	Imaging	Treatment
1	Trotter et al., 1984 [12]	11/M	Gait disturbance; 1 year	Hamstring tightness	Small, near Rt. PSIS	CT scan	En bloc excision
2	Larson et al., 2002 [13]	29/M	Hip and lower extremity radiating pain; 2 years	L-3 radiculopathy and diminished patellar reflex	10 cm, inner cortex of Lt. mid iliac wing	CT scan, MRI	En bloc excision
3	Kim et al., 2009 [14]	33/M	Lower extremity radiating pain, paresthesia, and limping; several months	L-5 radiculopathy	5 cm×5 cm×3 cm, near Rt. SI joint	MRI	En bloc excision
4	Sharma et al., 2009 [15]	21/M	Lateral waist swelling, 1 year	Non-tender bony hard mass	4 cm×3 cm, near mid -iliac crest	None	En bloc excision
5	Ali et al., 2014 [16]	16/M	Posterolateral waist swelling, 2 years	Non-tender bony hard mass	Not mentioned	None	En bloc excision
6	Nerurkar et al., 2015 [17]	17/M	Anterolateral waist swelling, 1 year	Non-tender bony hard mass	5 cm×4 cm, near Rt ASIS	MRI	En bloc excision
7	Yilmaz et al., 2015 (Turkish) [18]	12/M	Anterolateral waist swelling, 4 years	Non-tender bony hard mass	5 cm×4 cm×3 cm, near Lt. SI joint	CT scan, MRI	En bloc excision
8	Doomra and Kumar, 2016 [19]	17/M	Posterolateral waist swelling, 1 year	Non-tender bony hard mass	3 cm×3 cm; postero -lateral iliac wing	CT scan	En bloc excision
9	Chun et al., 2016 [20]	32/F	Groin pain with restricted hip mobility; 10 years	Non-tender bony mass in groin with snapping hip	3.3 cm×2.8 cm×2.0 cm; right ilium near AIS	CT scan, MRI	En bloc excision
10	Singh et al., 2016 [21]	21/F	Posterolateral waist swelling, 4 year	Non-tender bony hard mass	6 cm×4 cm near mid -iliac crest	CT scan	En bloc excision
11	Zhang et al., 2017 [11]	36/M	Pelvic pain with radiation and numbness in lower extremity; 5 months	Non-tender bony hard mass	Left ilium near PSIS	CT scan, MRI, 3D printing	En bloc excision, hemipelvic resection and L4 -S1 TLIF by T shape posterior only approach
12	Nekkanti et al., 2018 [22]	15/M	Anterolateral waist swelling, 4 years	Non-tender bony hard mass	4 cm×3 cm×1 cm, near postero -lateral iliac crest	None	En bloc excision
13	Kunjappan, 2019 [23]	18/M	Posterolateral waist swelling, 2 years	Non-tender bony hard mass	10 cm×8 cm, near mid -iliac crest	CT scan	En bloc excision
14	Magalhães et al., 2019 [24]	18/M	Paresthesia in the lateral right thigh, which was worse in the right lateral decubitus position, 9 months	Paresthesia over the lateral aspect of right thigh and thickening of the right iliac bone	Small, inner cortex of left ilium	CT, MRI, NCV for LFCN	Pt refused surgical treatment and follow-up got lost
15	Moreno-Ballesteros et al. 2020 (Spanish) [25]	74/M	Incidentally found	Known case of prostate cancer	2.3 cm×1.4 cm, near AIS of ilium	CT scan	No treatment indicated
16	Thomas et al., 2020 [26]	18/M	Painful left iliac wing mass with difficulty in clothing and wearing belt; 3 years	Tender bony hard mass	3 cm×2 cm×1.6 cm, left mid iliac crest	MRI	En bloc excision
17	Kulkarni et al., 2021 [27]	16/M	Anterolateral waist swelling, 8 months	Non-tender bony hard mass	4 cm×3 cm×3 cm, near right ASIS	CT scan	En bloc excision
18	Sun et al., 2021 [28]	45/F	Groin mass swelling, 20 years	Non-tender bony hard mass	16 cm×15 cm×10 cm, right ilium near ASIS	CT scan, MRI	En bloc excision
19	Fox and Kanawati, 2021 [29]	40/M	Left-sided lower back and flank pains radiating to thigh and knee along with numbness; 2 years	Non-tender bony hard mass	8 cm×5 cm×4 cm, near PSIS compressing L4, 5 nerve roots	CT scan, MRI, 3D printing	En bloc excision
20	Olivero et al., 2022 [30]	47/F	Inguinal swelling	Non-tender bony hard mass	Approx 7 cm×6.8 cm×6.5 cm	CT scan	En bloc excision and abdominopelvic reconstruction with mesh anchored harpoons and TFL graft
21	Our study, 2023	16/M	Posterolateral waist swelling, 3 year	Non-tender bony hard mass	8.5 cm×6.5 cm near mid -iliac crest	MRI	En bloc resection

CT: Computed tomography, MRI: Magnetic resonance imaging, ASIS: Anterior superior iliac spine, PSIS: Posterior superior iliac spine, TFL: Transforaminal lumbar interbody fusions, NCV: Nerve Conduction Velocity, LFCN: Lateral Femoral Cutaneous Nerve

Table 1: Review of published literature for osteochondroma of the ilium.



osteochondromas may truly be neoplasms as genetic mutations have been found in both MHE and solitary forms [7]. Despite the oldest article, we found about pelvic osteochondroma was by Ghormley et al., in 1946, there have been very few cases reported so far which suggest how rare is the pelvis for such a common tumor [8]. With their initial experience with 37 patients at the Mayo clinic between 1910 and 1943, they concluded that “chondromatosis of the pelvis must always be considered as serious surgical lesions.” The majority of osteochondroma arises from the ilium of the pelvis. However, ischium, pubis, and periacetabular region may rarely give rise to osteochondroma (Fig. 9).

Our case was unusual in a number of ways. The patient’s age at presentation was younger than the typical age group. Besides, it is again very rare for it to let alone develop into such a huge tumor within few years of course as in our case. In our case, the tumor size was increasing, which began causing significant impairment with daily living. Pedunculated morphology as in our osteochondroma case is less likely to undergo shrinkage as compared to sessile ones [9]. Osteochondroma with continuous growth after skeletal maturity should raise the possibility of malignant chondrosarcoma. However, we decided not to wait due to constant discomfort in wearing pants or the lower extremity outfits and near total skeletal maturity. Besides, despite the rare location, pelvic osteochondroma has more chance of malignant transformation into secondary osteosarcoma than any other site and it also is associated with worse outcome [10]. Other serious complication in these regions can lead to fractures, hip joint mobility restriction, vascular impairment, neurological sequelae, hematuria, and sexual dysfunction. X-ray itself can be diagnostic in most cases. However, we recommend that CT scan or MRI as it allows anatomic delineation necessary for the surgical planning. Moreover, 3D printing is of immense value in

case or osteochondroma causing nerve root compression [11]. Surgical en bloc excision is the treatment of choice for symptomatic osteochondroma. Our patient showed complete resolution of problems postoperatively with no post-operative recurrence. Similarly, all the cases we reviewed recovered fully after en bloc resection of tumor, and hence, it should be offered whenever indicated. Finally, there is no substitution for microscopic examination to prove that the chondrocytes grew actively and did not deteriorate. We do recommend two independent reporting to avoid any sampling or human error in definitive diagnosis.

Conclusion

Even though the majority of osteochondromas arise from the metaphysis of long bones, case reports have shown that osteochondromas presenting in atypical locations such as the pelvis do occur. Iliac osteochondroma can greatly impair the quality of life of the patients even after being asymptomatic clinically. Better recognition and more comprehensive evaluation of these rare cases should be highlighted to avoid misdiagnosis during clinical practice. All providers, particularly those in primary care, should be aware of these conditions as patients with asymptomatic iliac mass lesions will most likely initially present to them.

Clinical Message

We suggest a high index of suspicion for pelvic osteochondroma in skeletally immature patients and its en bloc excision is indicated whenever the atypical location of the tumor compromises the quality of life.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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