Giant Osteochondroma in the Pelvis of a Skeletally Mature Patient: Case Report and Review of Literature

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Abstract

Large pelvic osteochondroma is a rare presentation in adult patients, we present case of giant pelvic osteochondroma in a 52 years old male patient, symptomatic at presentations with pain and discomfort during sleeping, all radiology imaging and biopsy were consistent with benign osteochondroma with no malignant features. Patient underwent complete excision and delayed primary closure utilizing skin flaps and grafts. The final histopathology of the resected specimen showed osteochondroma with no suspicious signs of malignant degeneration. Upon follow up patient is doing well with complete wound healing, no signs of recurrence and back to normal functional life.

Keywords: Osteochondroma; Pelvic; Multiple Hereditary Exostosis

Abbreviations

MHE: Multiple Hereditary Exostosis; CT: Computerized Tomography; MRI: Magnetic Resonance Image

Introduction

Osteochondromas are the commonest bone tumors and often present as solitary lesions at the end of long bones. Hereditary multiple exostosis (HME), a type of rare benign bone tumor, is an inherited autosomal dominant disorder characterized by the presence of multiple exostoses or osteochondromas. Osteochondromas commonly develop during the first decade of a person’s life and most commonly arise around the knee joint. Those lesions usually stop growing when the individual reaches skeletal maturity [1,2]. Osteochondroma of the Pelvis is a rare tumor with an incidence of 5% [3] and rarely often present in elderly population due to the cosmetic and functional results that they can cause.

We report an extreme rare case of giant, neglected Osteochondroma of the posterior iliac crest in a 52-year-old man with Hereditary multiple exostosis that grew after skeletal maturity and was treated with wide local excision.

Case Presentation

A 52-year-old male was referred to the orthopedic oncology clinic for evaluation and management regarding a mass in the gluteal region. The patient is a known case of multiple hereditary exostosis and had undergone many surgeries for removal of variable size
osteochondromas in the lower and upper extremity. The patient reported a history of huge swelling in the lower back and gluteal region since childhood. Since then the swelling started to increase in size till it reached enormous size that bothers his sleeping position. For the past 4 months the swelling started to ulcerate and with foully smell and discharge associated with occasional pain and discomfort. He didn't report any constitutional symptoms. Patient does have similar swellings in other boney parts of the body including the groin, the right distal femur, right ankle. The patient has four children, two of them has the same condition as well his uncle has the same disease. Clinical examination revealed multiple osteochondroma lesions in the lower and upper limbs. And local examination of the gluteal regions showed enormous swelling over the lower back and gluteal region arising from the posterior iliac crest. The mass is firm, non-tender, and fixed to the bone (Figure 1A and 1B). There was marked skin thinning and multiple area of skin ulcerations with foully smell and discharge. No regional lymph nodes enlargement and had intact distal neurovascular examination. Laboratory tests showed mildly elevated C-Reactive protein elevated Erythrocyte sedimentation rate. Other hematological tests were insignificant. Plain radiographs, Computerized tomography scan and Magnetic resonance imaging were done and showed features consistent with osteochondroma (Figure 1C-1F).

Figure 1: A, B: Clinical picture showing large exophytic lesion arising from the right iliac crest posteriorly with multiple ulcerations. C: Plain radiographs demonstrates large radiopaque lesions projecting over the pelvis, the largest is seen over the right iliac region. D: Axial CT image showing direct intramedullary communication between the large pedunculated chondroid bone neoplasm and the right iliac bone posteriorly. E: Axial non-fat suppressed T1-weighted image showing a large chondroid lesion with direct intramedullary communication with the right iliac bone posteriorly. F: Axial fat suppressed fluid sensitive image showing the lesion, there is a T2 hyperintense cartilaginous cap of 1.5 cm thickness.

Multiple incisional biopsies were done to roll out sarcomatous degeneration and the results came back with benign osteochondroma with no features of malignant degeneration, so surgery was planned and done with resection of the bone lesion together with the overlying skin and skin ulcers. The defect was covered with vacuum dressing till obvious granulation tissue started to form and them plastic surgery covered the wound with multiple rotational flaps and skin grafts and was followed up till complete healing (Figure 2E and 2F). He is back to function with no functional deficits.

The final pathology was read finally as osteochondroma with no sarcomatous degeneration.

Histopathological examination of the resected specimen showed grossly rounded mass measuring 24.5 cm x 22 x 10 cm with multiple overlying ulcers. Final histopathological diagnosis was consistent with osteochondroma with no malignancy seen.

**Figure 2:** A, B: Pre surgery images. C: Intra operative image showing the tumor resection bed. D: The surgical bed was covered by VAC dressing. E: Multiple rhomboid flaps and skin grafts for coverage. F: Final clinical picture showing acceptable healing of the flaps and skin grafts.

**Figure 3:** A, B: Histopathological findings revealed a benign osteochondromatous lesion with dystrophic calcification. There was an area of abscess formation. There was no evidence of cytological atypia, necrosis or bone permeation to suggest malignant transformation. It measured 24.5 cm x 22 x 10 cm. Multiple skin ulcers were identified. The diagnosis was consistent with an osteochondroma.
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Discussion

Osteochondroma, commonly referred to as bony exostosis, is the most commonly encountered bone tumor accounting for more than 43% of all bony neoplasms [2]. The disease has obvious male predominance [4] and usually presents in the second decade of life with prominent bony swelling around the joints mainly the knee, shoulder and ankle joints. It is usually asymptomatic however some patients may complain of pain due to irritation or compression of major structures around the swelling like tendons, muscles and nerves. The disease could either present as a solitary lesion or part of multiple hereditary exostosis MHE.

MHE is an autosomal dominant disorder caused by genetic mutations of the tumor suppressor genes EXT1, EXT2, and EXT3. Affected patients with MHE usually present early in childhood with multiple osteochondromas and usually undergo many surgeries for resection during their lifetime. MHE has a higher chance of deep hidden osteochondromas as well as higher chance of malignant degeneration into chondrosarcoma.

The majority of osteochondromas arise from the lower end of the long bones particularly around the knee and shoulder joint. Pelvic involvement is rare and more common as part of MHE disease where the lesions arise from the inner side of pelvis reaching larger size before they are symptomatic with compression of the bladder, bowel and the neurovascular bundle.

Lesions arising from the posterior iliac crest usually present with nerve compression and usually present early due to the cosmetic and functional effect that they cause.

The diagnosis is often clear from plain radiography with the characteristic finding of corticomedullary continuity of the lesion and the native bone. Both sessile and pedunculated variants osteochondromas can be present with the latter being more common and accounting for more than 80% of the cases [5].

CT and MRI are often needed to assess the lesion and to roll out malignant degeneration by looking at further cortical details and cartilage cap thickness. The Definitive diagnosis is usually established on histopathological examination of the lesion and will help rule out malignant degeneration which can occur in 5% of cases of MHE and in 1% of solitary osteochondromas. Sudden and radio growth or continued growth after skeletal maturity should rise the suspicion of malignancy and trigger further diagnostic tests and biopsy.

Most of the osteochondromas can be managed by observation alone. Surgical treatment in the form of en-bloc resection is usually indicated for pain, cosmetic reasons, neurovascular compromise, abnormal growth, skeletal deformity, decreased motion of the adjacent joint or in cases with evidence of malignant transformation.

In our patient, the swelling has increased in size after skeletal maturity and was symptomatic and cosmetically unappealing, hence, we have considered surgical resection. The base of the tumor was reached, and en-bloc resection was performed with saucerization of the base of the tumor to ensure that no cartilage remnants are left behind. Recurrence after complete surgical resection is rare and are probably caused by failure to remove the entire cartilaginous cap [6].

Conclusion

Pelvic osteochondroma is a rare disease and more rarely seen is gigantic osteochondroma which was left untreated especially in the outer side of the pelvis where these can be symptomatic and cosmetically unacceptable. Successful surgery and soft tissue management usually leads to excellent results.

Summary

Osteochondroma is a common benign bone tumor most commonly arise around the knee joint; the presentation can be solitary or part of multiple hereditary exostosis. Pelvic osteochondroma, large size at presentation, is very rare especially in adult patients.

Consent
Written informed consent was obtained from the patient for publication of this case report along with the images.

Bibliography

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