Eur Spine J
DOI 10.1007/s00586-015-3928-8

CASE REPORT

Solitary sacral osteochondroma without neurological symptoms: a case report and review of the literature

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Received: 9 September 2014 / Revised: 31 March 2015 / Accepted: 1 April 2015

Abstract
Purpose To report a case of solitary sacral osteochondroma without neurological symptoms and describe the en bloc excision of the tumour, as well as review the literature on osteochondroma involving the sacrum.
Methods Summary of the background data: although osteochondromas are among the most common benign tumours of the bone, they uncommonly involve the spine. Its occurence in the sacrum is rare, accounting for only 0.5 % of the osteochondromas involving spine. All previous cases of sacral osteochondroma have reported neurological symptoms on presentation.
Case report A 21-year-old male presented with a palpable, painless mass in the left side of the sacral region of 1 year duration, without neurological symptoms. Radiological studies showed a well-circumscribed lesion with bony osteoid within arising from the sacrum at S3–S4 level left to midline, with features suggestive of osteochondroma. The tumour was excised en bloc through posterior approach. A literature review of sacral osteochondroma was conducted using MEDLINE search of English Literature and bibliographies.
Results Histopathological studies showed the lesion to consist mature bone trabeculae with active enchondral ossification with cap of normal hyaline cartilage. Literature review yielded only 8 cases of sacral osteochondroma reported earlier.
Conclusion This is the 9th case of solitary osteochondroma of the sacrum to be reported, the first to be reported without any neurological symptoms, and third case reported for which en bloc excision was performed.

Keywords Osteochondroma · Exostosis · Spine · Sacrum · En bloc excision

Introduction

Osteochondromas, the most common benign bone tumours [1–3], commonly involve the appendicular skeleton. Its occurrence in spine especially the sacrum is rare [2, 3]. In cases where the spine is involved, compression of the spinal cord or cauda equina may occur. To prevent this as well as to make a tissue diagnosis and exclude malignancy, excisional biopsy is usually necessary. We are reporting a case of solitary sacral osteochondroma, without neurological symptoms in a 21-year-old male.

Materials and methods

A 21-year-old male presented with a palpable, painless mass in the left side of the sacral region for 1 year with insidious onset and gradually increasing size. There were no neurological symptoms and the bladder and bowel function was normal. On examination, a bony hard, nontender spherical mass of 3 cm diameter was palpable over left side of S3–S4 region, with smooth margins and normal overlying skin. Rest of the spine and other parts of the skeletal system did not reveal such bony swelling. His

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Published online: 10 April 2015

neurological system was normal. The X-ray of sacrum showed a bony growth in S3–S4 region left to the midline (Fig. 1). MRI was done to see the soft tissue extension, possible neural compression and signs of malignant transformation. MRI showed a 36 × 22 mm, well-circumscribed, expansile lesion with bony osteoid within, arising from the sacrum at S3–S4 level to the left of midline, protruding posteriorly having hypointense rim with heterogenously hyperintense centre on T1 and T2WI indicating marrow: features suggestive of osteochondroma (Fig. 2a, b). Since the mass was progressively increasing in size making it uncomfortable for the patient to lie down as well as for local tumour control and diagnostic purpose the patient and his family wished to proceed with an excisional biopsy of the tumour.

Through a longitudinal paramedian incision over the mass, the tumour was seen arising from conjoint laminae of sacral S3–S4 vertebrae and removed en bloc in which whole tumour mass and cartilaginous cap along with healthy margin and base were removed as a single piece (Figs. 3a, b, 4). Care was taken to further clear the margins of tumour-bed meticulously, protecting the vessels and nerves and the sacro-iliac joints throughout the procedure.

The literature was reviewed by use of the following pertinent keywords—osteochondroma, exostosis and sacrum using MEDLINE search of English Literature and bibliographies of published manuscripts. Abstracts of interest were screened, and full articles were selected that were pertinent to the literature search. References of articles selected were further explored for additional relevant references. Selection of articles was not restricted because of study design or level of evidence. The extensive search revealed only 7 previous publications reporting a total of only 8 cases of sacral osteochondroma reported previously.

Results

Histopathology report showed a benign tumour composed of a cap of hyaline cartilage with mature bone trabeculae underneath having active enchondral ossification, confirming the diagnosis of osteochondroma (Fig. 5).

The postoperative period was uneventful. Follow-up at 3 and 8 weeks showed complete healing of skin wound, without any palpable swelling or any neurological dysfunction. At 6 months there was no evidence of any recurrence; however, further evaluation was not possible as the patient was lost to follow-up.

The Literature Review yielded 7 relevant articles, one case series and six case reports, which are summarized in Table 1. A total of 8 cases of sacral osteochondroma have been reported till date. However, patient details, exact tumour location and symptoms have not been described by Pugh et al. [4] and Sung et al. [5]. The predilection for age or sex could not be discerned from the review. Of the reported cases, two originated from lamina, one from articular process and one from ala of the sacrum. In our study, the exostosis has been found to originate from the conjoint laminae of S3 and S4 vertebra. Also, all cases reported previously had presented with neurological symptoms. Ours is the 9th case of solitary osteochondroma of the sacrum to be reported, first to be reported without any neurological symptoms, and third case reported for which en bloc excision was performed.

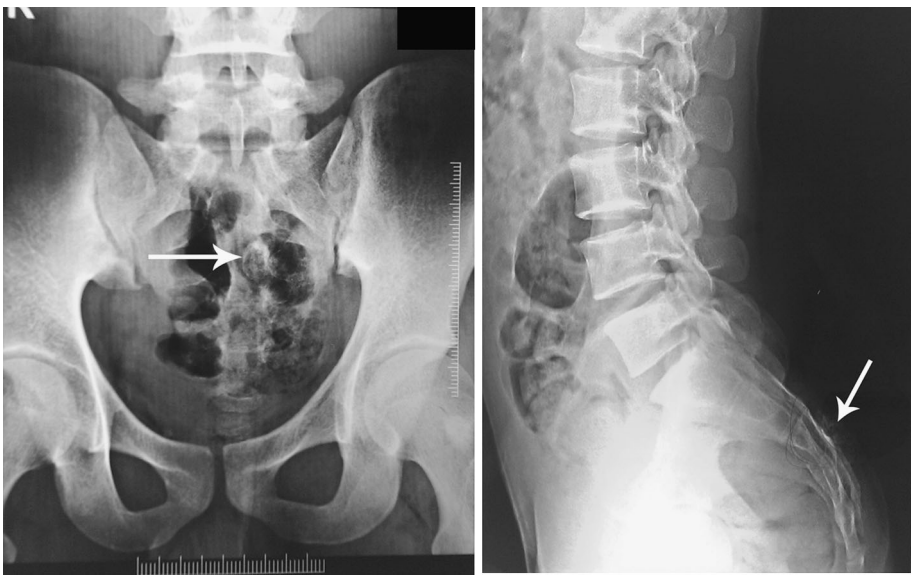

Fig. 1 X-ray of sacrum in antero-posterior and lateral views showing a rounded bony growth at S3–S4 level to the left of *midline*

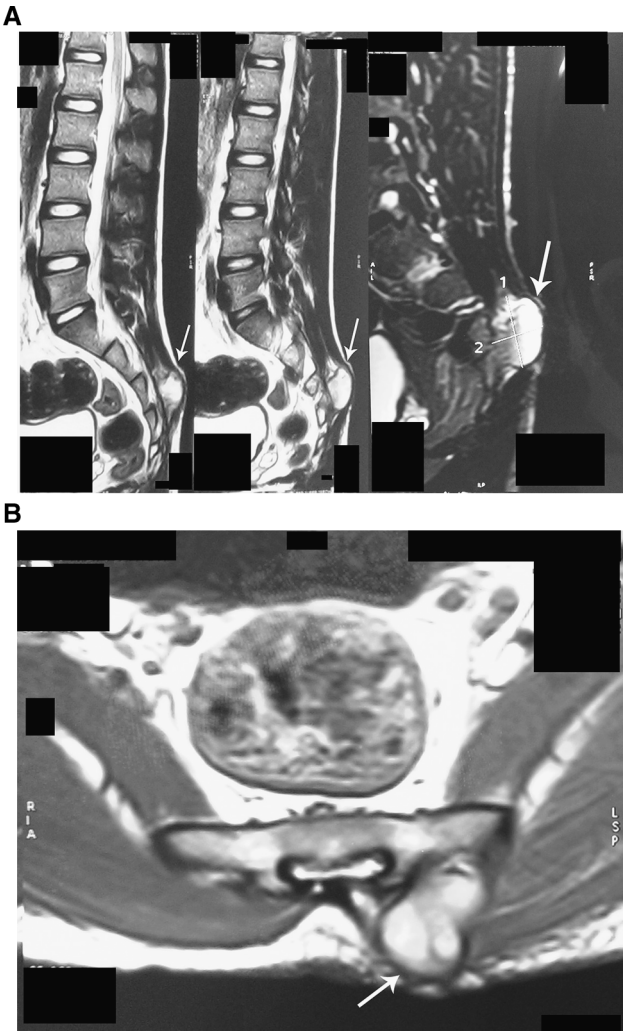

Fig. 2 **a** Sagittal section of MRI showing 36 × 22 mm, well-circumscribed bony lesion protruding posteriorly at S3–S4 level without any anterior extension. **b** Cross section of MRI showing bony lesion arising from left of *midline* with thin hypointense rim and hyperintense *centre* indicating marrow

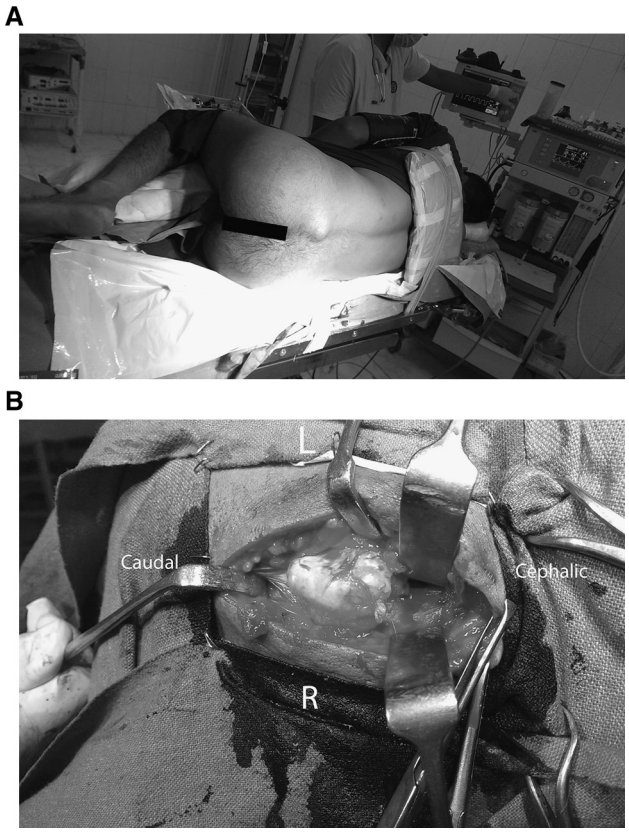

Fig. 3 **a** Under general anaesthesia, patient was placed in right lateral position in the operation theatre. **b** Exposure of the tumour by a longitudinal incision over it and by splitting and retracting the erector spinae and gluteal muscles of the left side

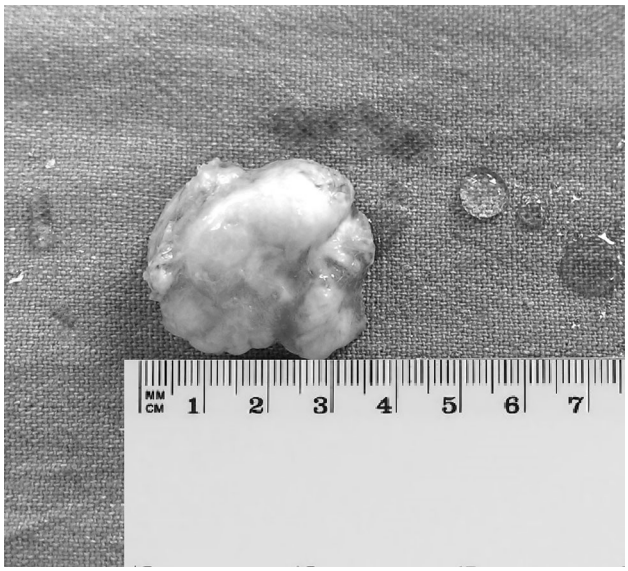

Fig. 4 En bloc excision of the tumour having cartilage capping

Discussion

Osteochondroma is caused by overgrowth of cartilage at the edge of the growth plate, which separates and grows by progressive endochondral ossification into a bony protuberance covered by cartilaginous cap [9], and has a fatty or haematopoietic marrow containing medulla [10] which is in continuity with the medulla of the bone from which it arises [2, 11]. It may be solitary or multiple [1, 12]; multiple being often associated with hereditary multiple exostoses (HME), an autosomal dominant disorder [2, 13, 14]. Osteochondromas involve the spine in approximately 3 % of cases [1, 3], with solitary lesions involving

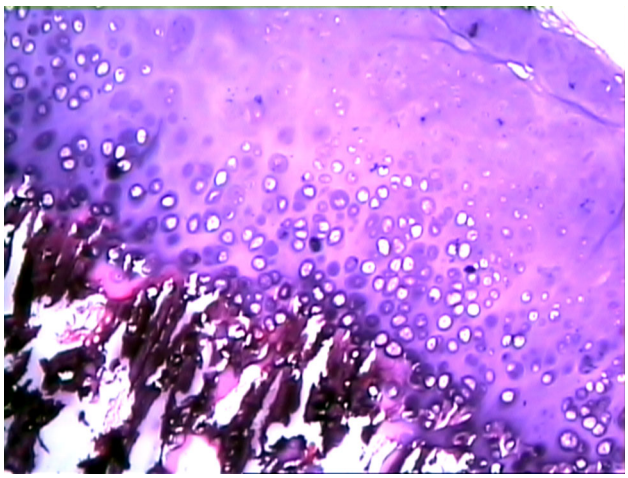

Fig. 5 Histopathology examination shows cap of hyaline cartilage with mature bone trabeculae underneath with active enchondral ossification confirming the diagnosis of osteochondroma

1.3–4.1 % and HME involving 3–9 % of the cases [2, 3, 15]. In spine, they arise from the neural arch [16] and the predominant sites are cervical and upper thoracic vertebrae. Less than 0.5 % of osteochondromas of the spine occur in the sacrum [1, 3].

The most common presenting symptom of spinal exostoses is painless mass as the tumour grows posteriorly into the soft tissue [17]; symptoms consistent with spinal cord or nerve root compression, though rare, can occur if the tumour grows anteriorly into the spinal canal compressing the neural structures [17, 18], with reported incidence of 0.5–1 % [18, 19], and more commonly associated with HME [16]. All previous cases of sacral exostoses have reported neurological symptoms on presentation, in contrary to our case.

X-rays and computed tomography (CT) scans are diagnostic in most cases and allow anatomical delineation of the lesion. However, MRI is important to visualise the size of the cartilaginous cap, the spinal cord and other neural structures compression and also to differentiate exostoses from chondrosarcoma [20]. Differential diagnosis from other neoplasms involving the sacrum like chordoma, giant cell tumours and bone cysts is not difficult because of their typical clinical features and radiologic appearances. Osteochondromas are mainly differentiated from juxtacortical chondroma, myositis ossificans and parosteal chondrosarcoma. Juxtacortical chondromas usually have a scalloped cortical defect with a sclerotic margin, whereas in myositis ossificans the apparent tumour does not blend with the cortex. Parosteal chondrosarcomas are suspected if increasing size is noticed after skeletal maturity and best differentiated with help of MRI [9].

Surgical excision of spinal tumours is challenging given the anatomic peculiarity and hypervascularity. In sacrum, it is no different with proximity of lateral sacral vessels and exiting nerve roots when the tumour is located posteriorly. In anteriorly located tumours the presence of cauda warrants further precision.

Surgical excision of sacral osteochondroma with free margins usually constitutes definitive treatment. Although in our case we have excised the tumour by a paramedian

Table 1 Summary of published literature on sacral osteochondroma

References	No. of cases	Age/sex	Level	Location	Presentation	Treatment
Pugh et al. [4]	1					
Sung et al. [5]	2		Below S3			Surgery. Tumour excision from posterior approach
Hanakita and Suzuki [6]	1	42 years/F	L5–S1	Lamina	Low back ache with urinary disturbance and cauda equina compression	Surgery. Tumour excision with hemilaminectomies of left L4–L5 and right L5–S1
Agrawal et al. [7]	1	14 years/M		Right ala of sacrum	Low back ache with right sided radiculopathy	Surgery. Tumour excision with posterior approach
Samartzis and Marco [1]	1	11 years/M	S2	Lamina	Right posterior thigh pain	Surgery. En bloc excision with right S1–S4 laminectomy
Chin and Kim [8]	1	54 years/F		Sacrum	Low back ache with left lower limb radiculopathy	Surgery. En bloc excision through abdominal-retroperitoneal approach
Kuraishi et al. [2]	1	63 years/F	S1	Articular process	Drop foot, numbness	Surgery. Right partial hemilaminectomy at L5–S1 level
Present case	1	21 years/M	Conjoint laminae of S3–S4	Left of midline	Painless mass	Surgery. En bloc excision via posterior approach

incision, a midline incision would have been ideal, so that future access to extensile approaches to the revision spine surgery is not altered and would deal with contamination of the surgical bed adequately, if required. A frozen section biopsy should be performed ideally to confirm margin free of tumour, which could not be done in our case due to unavailability. During excision, care should be taken to preserve the pelvic organs, blood vessels and sacral plexus [21]. Complete excision of the cartilaginous cap and its overlying periosteum is necessary to decrease the likelihood of recurrence [1]. In our case, tumour was excised en bloc.

Conflict of interest None of the authors has any potential conflict of interest.

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