Review Journal of Clinical Orthopaedics 2021 July-Dec;6(2):45-48

Osteoblastoma of the lumbar spine in an adolescent: A case report and review of literature

Bhushan Sagade[1], Sarang Rokade[1], Arjun Dhawale[1], Abhay Nene[1]

Abstract

Introduction: Osteoblastomas are primary bone tumors representing 1% of all bone tumors and 10% of all spinal osseous neoplasms with a predilection for posterior elements.

Case report: A 13-year-old boy with insidious backache for six months presented with progressive radiating paraesthesia and claudication, restricted lumbar motion and positive straight leg test bilaterally with weakness of left ankle dorsiflexion. Radiograph showed an subtle expansile lytic lesion in the L3 posterior elements. CT and MRI revealed a space-occupying lesion of the L3 vertebra lamina, involving the left pedicle causing severe spinal canal stenosis. Excision of the posterior elements of the L3 vertebra including the facet and left pedicle and short segment fixation from L2-L4 using autogenous rib was done.

At two years postoperatively, he was asymptomatic, neurologically normal, showing radiographic evidence of union with no recurrence.

Conclusion: Autogenous structural rib can be used for posterolateral fusion after osteoblastoma excision with potential instability.

Keywords: Osteoblastoma, back pain, rib graft, postero-lateral fusion, en-bloc resection.

Introduction

Osteoid osteoma was first described by Jaffe in 1935 [1], osteoblastomas were described by Jaffe and Lichenstein in 1956 independently [2]. They are benign bone tumours constituting about 11 % of all primary bone tumours [1-3]. The condition is more frequent in males, (male to female ratio of 2:1) and occurs during the second decade of life [1-3]. The most common location for these lesions is the spine with a preponderance for posterior elements of the spine constituting 30-40 % of all the cases [1,3,4]. Two types of osteoblastoma have been described in the literature; conventional osteoblastomas and aggressive osteoblastomas, the latter being characterised by high alkaline phosphatase levels and size of more than 1.5 cms with paravertebral or epidural extension and lytic changes on radiographs [4].

Treatment of osteoblastomas of the spine involves en bloc excision of the lesion in Enneking stage 3 lesions and intralesional curettage in Enneking stage 2 lesions [5,6]. Radiotherapy is considered as an adjuvant or an alternative to surgical excision if excision demands unacceptable functional sacrifice or in non-accessible locations [7]. Recurrence can occur if excision is inadequate. Overall recurrence rates reported for osteoblastomas has been around 10 – 15% [2]. Recurrences are typically seen 5-10 years after index procedure [4].

We describe a case of osteoblastoma of posterior elements of L3 vertebra in a 13-year-old boy treated with en bloc excision and posterolateral fusion using rib autograft.

[1]Department of Paediatric Orthopaedics, Bai Jerbai Wadia Hospital for Children, Parel, Mumbai 400012

Address of Correspondence
Dr. Dhawale Arjun A,
Department of Paediatric Orthopaedics, Bai Jerbai Wadia Hospital for Children, Parel, Mumbai 400012
E-mail: arjundhawale@hotmail.com

Case report

A 13-year-old boy presented to clinic with complaints of low back pain of six months duration which was insidious in onset, gradually worsening with time. Pain was present during rest, it radiated to both lower limbs associated with parasthesia, and claudication symptoms with walking, with relief on lying down with the hips and knees flexed. There was no history of trauma, heavy weightlifting, no unaccustomed activity, no history of fever, weight loss, loss of appetite, tuberculosis, no morning stiffness, or small joint pains. Past medical history was unremarkable. On examination there was restriction of movements of the lumbar spine and focal tenderness over the lumbar region. Power of ankle dorsiflexion on the left side was grade 3. Ankle jerk on the left side was diminished. Rest of the neurological examination was unremarkable.

Radiograph showed a lytic lesion in posterior elements of L3. MRI revealed a space occupying lesion in the posterior

Submitted Date: 4-Sep 2021, Review Date: 12-Nov-2021, Accepted Date: 12-Nov-2021 & Published Date 31-Dec-2021

Figure 1: Plain radiograph of the lumbosacral spine Antero-posterior and Lateral view. The Antero-posterior view shows widening and irregularity of the left pedicle of L3. There is irregularity and widening visible in the posterior elements of L3 on the Lateral radiograph

Figure 2: Preoperative MRI of the Lumbosacral spine sagittal and axial sections showing isointense lesion involving the lamina and pedicles of the L3 vertebra with expansion of the bony posterior elements causing severe compression of the left sided L3 nerve root

Figure 3: Preoperative axial and sagittal sections of CT scan of the Lumbar spine showing an expansile bony lesion with lytic and sclerotic components arising from the spinous process and extending into the left pedicle of L3 vertebra causing severe stenosis of the neural canal

elements of L3 involving the left pedicle and causing severe spinal canal stenosis. CT revealed a heterogeneous expansile bony lesion with lytic and sclerotic component arising from the spinous process of the L3 vertebra extending into the left pedicle (Figure 1-3). A PET-CT scan showed the solitary lumbar lesion with no other lesion elsewhere. Alkaline phosphatase level was 224 IU/L(Normal- 38 – 94 IU/L).

After pre-operative work-up, consent and general anaesthesia, patient was positioned prone; exposure of L2-L4 levels was done after level confirmation. En bloc excision of the L3 lamina including the facet joint and left pedicle was done as these were involved. Posterior stabilisation with pedicle screws at L2 and L4 was done. Posterolateral fusion was performed to prevent iatrogenic instability and autogenous rib graft (left 10th rib) was harvested for fusion (Figure 4). The estimated blood loss was 200 ml. Closure was performed and the patient was extubated, the post-operative period was uneventful.

Histopathological examination of the resected specimen confirmed the lesion to be osteoblastoma (Figure 5).

Patient followed up every six months and at two years' post-operatively has had no symptoms or radiological evidence of recurrence of the disease or implant loosening (Figure 6, 7, 8).

Discussion

Clinical presentation: Osteoid osteomas and osteoblastomas of the spine are uncommon tumours which may present with atypical symptoms and normal radiological findings in the initial course of the disease and thus may lead to delays in diagnosis [2]. It may present as persistent, dull back pain. Other presentations may include a painful scoliosis. Scoliosis is usually convex

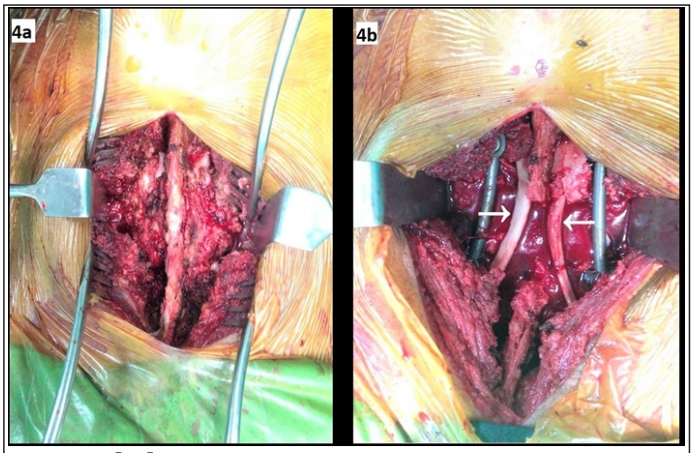

Figure 4: [4a] showing posterior exposure of the lumbar vertebrae. [4b] demonstrates the insertion of autogenous rib graft (white arrows) after resection of the lesion to enhance the posterolateral fusion of the spine

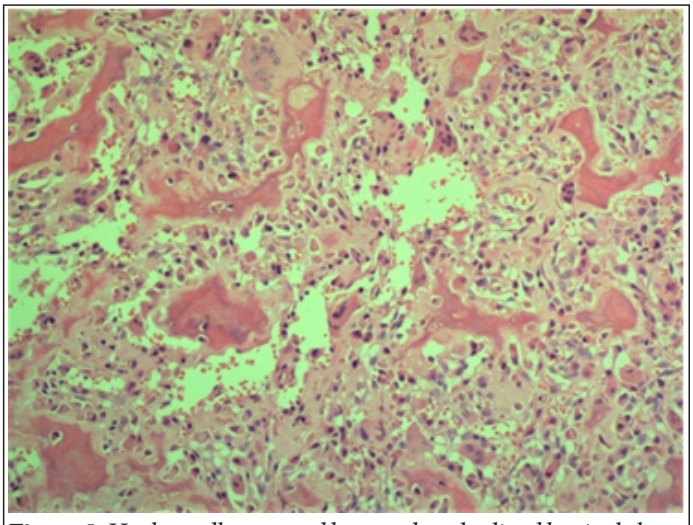

Figure 5: Haphazardly arranged bony trabeculae lined by single layer of osteoblasts are seen (H & E x 100)

Figure 6: Postoperative radiograph showing short segment fixation from L2 to L4 with pedicle screws in situ with satisfactory incorporation of the rib grafts 2 years after surgery and no evidence of recurrence

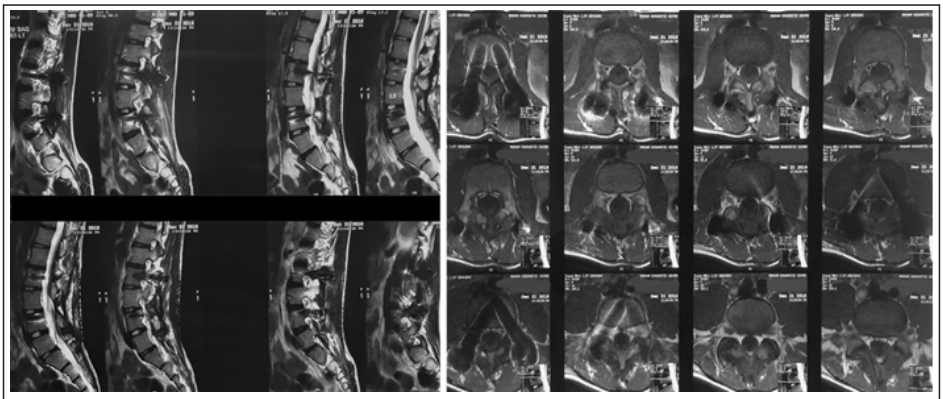

Figure 7: Sagittal and Axial sections of MRI 2 years after surgery demonstrating adequate decompression of the spinal cord, well positioned implants and no evidence of recurrence of the lesion

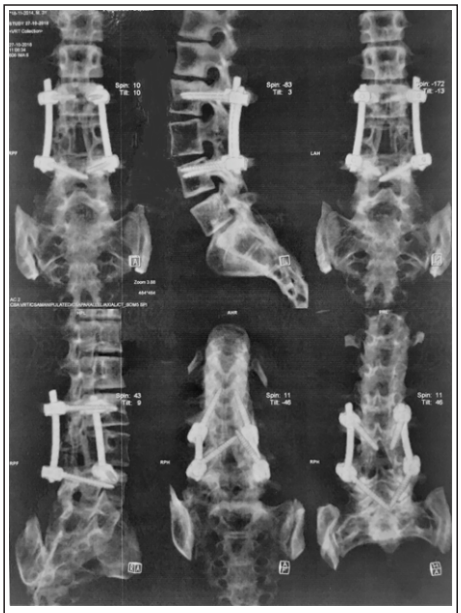

Figure 8: Postoperative CT scan demonstrating uptake of the rib graft and posterolateral fusion

opposite to the side of the lesion. Radiculopathy and neurological deficit with cauda equina syndrome can occur if the lesion impinges on the nerve roots or the spinal cord [3]. Thus, backache should not be presumed to be postural or inflammatory if long-standing and with red flags.

Investigations: Plain radiographs maybe less sensitive in picking up the lesion in early stages as lytic lesions cannot be identified on radiographs unless there is approximately 50% bone destruction [4]. CT scan and MRI provide detailed information of the extent of the lesion, involvement of adjacent structures, distortion of local anatomy and intra-spinal extent of the lesion. A PET-CT scan detects involvement of other regions and allows staging.

Pathology: Osetoblastomas are known to be more aggressive tumours. Locally aggressive tumours can cause mass symptoms. Malignant transformation of osteoblastomas has also been reported [2].

Literature summary is shown in Table 1.

Treatment: Enneking staging has been used by many authors to guide the method of treatment [4,7]. Intra-lesional excision has been advised for Enneking stage 2 lesions and en-bloc resection for stage 3 [7]. Pre-operative arterial embolization has been shown to reduce intra-operative blood loss [8]. Surgical resection is the conventional treatment of choice for spinal osteoblastomas after meticulous surgical planning. Intraoperative use of navgation provides accurate localisation facilitating complete removal [9]. In this case interbody fusion was avoided as it would violate compartments. Stabilisation is warranted if excision of intervertebral joints or facets is done [5].

Minimally invasive options like CT-guided radiofrequency ablation and image guided cryoablation may avoid need for fusion. Recently fully endoscopic resection has been described for spinal osteoblastoma. Use of denosumab preoperatively has been reported to regress tumour, ossify and facilitate resection [10].

To conclude, Autogenous rib graft can be used as a structural graft for posterolateral fusion after osteoblastoma excision with potential instability.

Clinical relevance

Back pain in adolescents should not be considered as postural or inflammatory especially when associated with red flags. The patient should undergo appropriate investigations to reach a diagnosis. Autogenous rib can be reliably used as a structural graft for posterolateral fusion.

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

Conflict of Interest: NIL; **Source of Support:** NIL

Table 1: Review of Literature

NAME	AUTHOR/ JOURNAL/ YEAR	SAMPLE	SEX	MEAN AGE (yr)	LEVEL	INVOLVED AREAS OF THE VERTEBRA	SYMPTOMS	ADJUNCTIVE THERAPY	TECHNIQUE	BIOPSY	IMPLANTS	COMPLICATIONS	CONCLUSION
Osteoblastoma: A 30 year study of 99 cases	Micah Berry et al/ Journal of Surgical Oncology/2008	99	69,M: 30,F	24	28 in the vertebra, rest in the extremities	NA	NA	NA	Curretage and en-bloc resection, auto/allograft	NA	NA	Recurrence	OBL frequently affects the long bones and the spine. Recurrence rates following curretage is relatively high and can be minimised by resection in selected cases
Management of osteoblastoma and osteoid osteoma of the spine in childhood	Sasha Burn et al/ Journal of Neurosurgery: Pediatrics/2009	33	20,M : 12,F	13: OO, 12: OBL	11 C, 3T, 15 L, 1 Clivus	Body and posterior elements	Pain, radiculopathy, stiffness, scoliosis, absent reflexes,	NA	Subtotal or total resection	NA	NA	Paralytic ileus in 1, surgical site infection in 3,	Spinal OO and OBL can be a challenging management problem in pediatric patients. If conservative therapy fails, surgery using modern intra-operative imaging and spinal instrumentation can provide symptom relief and tumour control
Staging and treatment of osteoblastoma in a mobile spine: a review of 51 cases	Stefano Boriani et al/ Eur Spine Journal/2010	51	34,M: 17,F	24.1	11 C, 17 T, 23 L	NA	Pain, scoliosis in 2	Pre-operative embolisation, Postoperative radiotherapy	Intra-lesional or en-bloc excision	NA	NA	Death, progressive kyphosis in 2, local recurrence and implant failure, infection, screw breakage	Intra-lesional excision is effective in Enneking stage 2 and en-bloc resection in stage 3
Osteoblastoma of the sacrum: report of 18 cases and review of literature	Pietro Ruggieri et al/Spine/2014	18	16,M: 2,F	8.4	L5-S4	Anterior and posterior	Low bak pain and radiculopathy, neurodeficit	Radiotherapy in 1, phenol in 7, and cryotherapy in 1	Curettage, intra-lesional excision, wide resection	Needle or open biopsy or intraoperative frozen section	NA	Massive bleeding, wound dehiscence, infection requiring 2 surgical procedures and external radiotherapy	Sacral osteoblastomas are rare, difficult to diagnose and complicated treatment choices, intralesional ressection was successful, local adjuvants did not reduce recurrence. Preoperative embolisation is recommended,
Surgical resection of osteoid osteoma and osteoblastoma of the spine	Muayad Kadhim et al/ Journal of Pediatric Orthopedics B/2016	17	6,M: 4,F	11.5	6C, 1T, 3L: OO. 1C, 2T, 2L: OBL	Only posterior elements in osteoid osteoma, posterior elements and	Pain, stiffness, radiculopathy, scoliosis	NA	Intralesional excision, enbloc excision	NA	None, cable wire , halo	Persistent pain in 1, recurrence in 2	O-arm provided easy and accurate localisation, and verification of complete excision of tumour
Surgical management of osteoblastoma of the spine: case series and review of literature	Benjamin Elder et al/ Turkish Neurosurgery/ 2016	5	4, M: 1, F	28.4	3C, 1L, 1S	Posterior elements of cervical spine	Pain, neurodeficit	Preoperative embolisation in 1	Intra-lesional and en-bloc excision	CT guided biopsy	Pedicle screws, Allograft,	Recurrence	Aggressive surgical resection can minimise risk of recurrence although it may still occur after en-bloc resection. Removal of structural elements surgically makes fusion often necessary. OO can progress to OBL in the spine despite previous resection.
CT-guided radiofrequency ablation of spinal osteoblastoma: treatment and long-term follow up	Francesco Arrigoni et al/ International Journal of Hyperthermia/ 2018	11	7,M: 4,F	26	5T, 4L, 2S	NA	Pain	NA	CT guided radiofrequency ablation	CT guided	NA	NA	CT-guided radiofrequency ablation is safe and effective for spinal OBL with the advantage of being minimally invasive
Percutaneous image guided cryoablation of osteoblastoma	Roberto Cazzato et al/ Amereican Journal of Radiology/ 2019	10	7M; 3 F	21 (median)	3C, 1T, 1L, 2 S	NA	Pain	Pre-operative embolisation in 2 cases	Cryoablation	In 7 patients	NA	Permanent sensory deficit in the arm, transient horners syndrome,	Percutaneous image guided cryoablation represents an effective option for patients affected by painful osteoblastomas. It can be safely performed with comprehensive protective measures
Can osteoblastoma evolve to malignancy? A challenge in the decision-making process of a benign spine tumour	Addisu Mesfin et al/ World Neurosurgery/ 2019	2	M	25, 34	L5, T10	Body, lamina and pedicles	Pain, weakness	Post-operative radiotherapy	Radiofrequency ablation, intralesional excision, enbloc resection	CT guided biopsy	Pedicle screws, cage	Recurrence and death	Conversion of OBL to osteosarcoma stresses upon performance of appropriate enneking staging for benign aggressive tumours as well as always performing a biopsy particularly at recurrence if imaging is not pathognomonic for a benign primary tumour

ROM: Range of motion, OBL: osteoblastoma; OO: osteoid osteoma;

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Conflict of Interest: NIL
Source of Support: NIL

How to Cite this Article

Sagade BS, Rokade SN, Dhawale AA, Nene AM. Osteoblastoma of the lumbar spine in an adolescent: A case report and review of literature. Journal of Clinical Orthopaedics July-Dec 2021;6(2):45-48.