Proximal Curve Decompensation Following a Local Resection and Selective Fusion of Lumbar Spine Osteoblastoma with Scoliosis: A Case Report

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Abstract: ***Background**: The recommended treatment for Osteoblastoma with scoliosis is surgical resection, correction of scoliosis with posterior spinal instrumentation and fusion. There were reported risk of recurrence of osteoblastoma but no published report on proximal curve decompensation when selective fusion of lumbar spine is chosen. **Case presentation**: A 12-year-old girl presented with low back pain for 3 years and was found to have back deformity. She was diagnosed with thoracolumbar scoliosis with apex at L2 vertebra with Cobb's angle of 43^0 secondary to Osteoblastoma pedicles of left L2 and L3. Tumour excision with hemilaminectomy left L2 and L3, posterior instrumentation and selective fusion from T12 to L4 was done. Unfortunately, the Cobb's angle worsened to 65^0 proximal to the upper instrumented vertebra (UIV) with proximal thoracic curve of 30^0 after 2 years follow up. An extension of posterior spinal instrumentation, curve correction and fusion up to T3 was performed. Latest review 6 months after the surgery revealed no aggravated curve or shift, no back pain and no neurological deficit. **Conclusions**: Scoliosis secondary to osteoblastoma need to be monitored closely following resection and selective fusion in skeletally immature patient. Risk of proximal curve decompensation as well as secondary surgery need to be aware of.*

Keywords: Osteoblastoma, Scoliosis, Proximal curve decompensation and Selective fusion

1. Introduction

Osteoblastoma is a known osteoblastic and locally aggressive benign primary bone tumours that affect predominantly children aged 10–15 years [1]. The most common location is the vertebral column (28–36%) and usually found at the posterior elements, especially the lamina and pedicles [2, 3]. Eventhough it is a benign tumour, it can behave aggressively causing bone destruction, soft tissue infiltration, epidural extension, malignant transformation and metastatic disease [4]. Surgical resection is a mainstay of treatment in neurologically impaired patient with back pain to improve symptoms [5]. There were reports on osteoblastoma with severe scoliosis, surgical management and risk of osteoblastoma recurrence but none about the risk of worsening scoliosis or proximal curve progression following surgery. Here, we describe a complication of proximal curve decompensation following a local tumour resection and selective fusion of spinal osteoblastoma with scoliosis that have not been reported before.

2. Case Presentation

A 12-year-old girl presented with low back pain for 3 years with no neurological abnormality and no constitutional symptoms. She did not attain menarche at the time of presentation and was found to have back deformity only during her visit to a hospital due to persistent back pain.

Examination revealed atypical scoliosis curve with right sided lumbar curve with lumbar hump. The left shoulder was 1 cm higher but the pelvic was balanced and no lower limb length discrepancy. Mild tenderness at L2 and L3 region. The neurological examination was normal with unremarkable laboratory studies.

The plain radiographs showed thoracolumbar scoliosis with apex at L2 vertebra on the right side with Cobb's angle of 43^0 with secondary compensatory left sided thoracic curve of 20^0 (**Figure 1**). The radiographic shoulder height (RSH) was 1cm higher on the left side and the sagittal balance was normal from lateral radiograph.

CT scan revealed thoracolumbar scoliosis with well-defined expancile lytic lesion with internal matrix mineralization and sclerotic changes at left lamina of L2. MRI demonstrated an irregularly shaped bony lesion from left lamina of L3 and heterogeneously hypointense on T1, T2 and TIRM sequences. The lesion also involve the left L2 and L3 pedicles with diffuse surrounding soft tissue inflammatory changes.

Open biopsy from left L2 lamina was done and the histopathological examination (HPE) showed features of osteoblastoma with irregular trabeculae woven bone, rimmed by osteoblasts. The osteoblasts are large, polyhedral in shape with vesicular nuclei, prominent nucleoli and abundant eosinophilic cytoplasm. Loose fibrovascular stroma are present at intertrabecular space with infrequent mitosis.

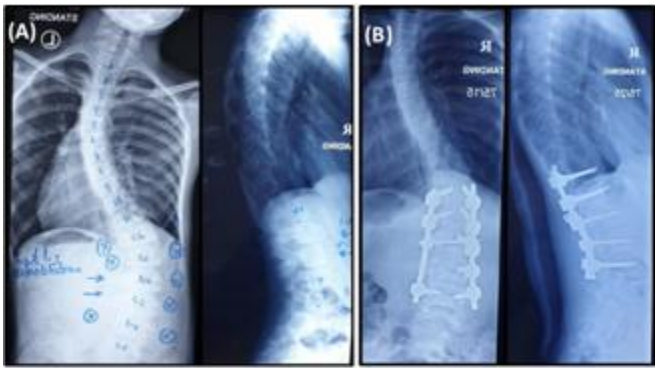

Figure 1: (A) Radiographs of thoracolumbar scoliosis with osteoblastoma of L2 and L3, and secondary proximal thoracic curve. (B) Postoperative radiographs after tumour resection and selective fusion T12 to L4.

She underwent left L2 and L3 resection with hemilaminectomy with posterior spinal instrumentation and fusion from T12 to L4. Intraoperatively, the lamina of L2 and L3 were soft with high vascularity and was removed intralesionally. Estimated blood loss was 1500ml and neuromonitoring showed no abnormal signal change throughout the surgery. She was then on thoracolumbar spinal orthosis (TLSO) cast to prevent curve progression. The repeated intraoperative HPE confirmed osteoblastoma.

Unfortunately, despite the surgery and TLSO, the scoliosis curve progressed with left truncal shift, left thoracic hump and left shoulder higher on follow up 1 year after the surgery. There was minimal mechanical back pain and the neurological examination were unremarkable. She had attained menarche during the review and was planned for another surgery. Repeated plain radiographs showed worsening Cobb's angle of 65^0 proximal to the upper instrumented vertebra (UIV) with thoracic curve of 30^0 Cobb's angle on side bending radiograph (**Figure 2**).

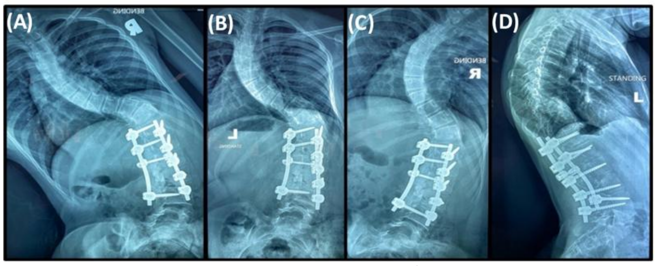

Figure 2: Radiographs 2 years after 1^{st} surgery (A) left sided bending, (B) AP radiograph, (C) right sided bending and (D) lateral view.

Therefore, an extension of posterior spinal instrumentation and fusion up to T3 was done 2 years after the 1^{st} instrumentation. Intraoperative finding showed well fused vertebrae along the previous instrumentation and no recurrence of osteoblastoma. The curve were able to be reduced in order to correct the shoulder balance and truncal shift (**Figure 3**). There were no perioperative complications. Latest review 6 months after the surgery revealed no aggravated curve or shift and she had no back pain with intact neurology.

Figure 3: Radiographs after the extension of instrumentation and fusion up to T3.

3. Discussion

There were many published data regarding the incidence, natural history, character and treatment of osteoblastoma in the spine [5, 6]. The incidence of scoliosis in osteoblastoma was reported to be as high as 63% and the asymmetric location of osteoblastoma in the spine leads to abnormal curve progression especially in growing spine, leading to the formation of scoliosis [7].

In term of treatment, surgical resection of osteoblastoma in the spine is the most effective treatment [5]. Boriani et al. found that intralesional curettage was effective for early stage osteoblastoma but intact resection is needed for more aggressive stage to minimize recurrence [1]. Early intervention is needed in order to alleviate pain and prevent any possible neurological injury [8].

In our case, the initial surgery was resection of L2 and L3 lesion with selective fusion from T12 to L4. The decision was made because she was skeletally immature during the first review with unfused triradiate cartilage. The aim was to preserve the motion segments and allowing the rest of spine to grow as well as prevention of crankshaft phenomenon from long segment fusion. The risk of progression was anticipated, thus, TLSO cast was applied for 4 months.

Eventhough the curve progression was expected, but no published data was found on proximal curve progression or decompensation following selective fusion of lumbar osteoblastoma with scoliosis. This phenomenon did reported in adolescent idiopathic scoliosis (AIS) in few studies and it is also known as proximal adding on [9, 10]. Craig Louer et al. reported 10% (3/30) patients with primary thoracolumbar/lumbar AIS underwent selective fusion had progression of curve after 10 years follow up [10]. According to Ding Q et al., 8.5% (11/130) patients following anterior selective fusion in Lenke 5 AIS developed proximal adding on with lower Risser grade, upper instrumented vertebra (UIV) lower than upper end vertebra (UEV) and C7 plumline (PL) away from UIV were the associated risk factors [9].

The possible explanation for our case curve progression are skeletally immature spine and truncal shift to the left (C7PL away from UIV) eventhough no literature to support this theory with osteoblastoma of spine. Extension of fusion to T3 with correction of curve was delayed to allow for skeletal maturity during the definitive 2^{nd} surgery and to prevent crankshaft deformity. Delaying the surgery further will lead to difficulty in curve correction due to rigid spine. Thus, more complex surgery with osteotomy maybe required which carries higher morbidity.

4. Conclusion

This case emphasizes that proximal curve decompensation is a phenomenon that can happen following selective fusion of scoliosis secondary to osteoblastoma especially in skeletally immature patient and presence of truncal shift. Secondary surgery for long fusion is required at appropriate time to reduce morbidity.

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