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Case Report

Posterior Hemivertebrectomy for Correction of Congenital Kyphosis in Adulthood. Case Report and Literature Review

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Abstract

Introduction: Congenital kyphosis is an infrequent cause of kyphosis with a potential severe progressive deformity and neurological damage. There are few reports in the literature about congenital kyphosis focusing on adults. **Case presentation**: 24-year-old male with progressive dorsal pain, paresthesias, and numbness in his legs. Physical examination demonstrated a rigid kyphotic deformity with tenderness on the apex, neurologically intact. The diagnosis was a T10 butterfly posterior hemivertebra with segmental kyphosis of 48 degrees. The patient was treated surgically with a single posterior procedure, performing a complete T10 hemivertebrectomy and instrumented fixation from T8 to T12. The postoperative assessment demonstrated a segmental kyphosis of 14 degrees with 34° of correction, without surgical complications, and good functional results during 4 years of follow-up. **Conclusion**: Posterior-only hemivertebra resection is an effective procedure for treating congenital kyphosis, providing an excellent correction of the deformity with a good functional outcome. Intraoperative imaging tools, such as CT scans and navigation systems, can aid in performing the surgery and improve outcomes.

Keywords: Case Report; Congenital Kyphosis; Adulthood; Butterfly Hemivertebra

Introduction

Congenital Kyphosis is an uncommon spinal deformity characterized by abnormal kyphotic angulation secondary to an alteration of the embryonic development and growth of one or more segments that fails formation or segmentation of one or more vertebral bodies and disks [1]. Generally, the diagnosis is made during childhood, and the treatment is performed early[2]. Literature regarding the outcome of treatment of this condition in skeletally mature patients is scarce, making it challenging to recommend a treatment guide for adults.

We present the results of the surgical treatment of a 24-year-old male patient with a history of congenital kyphosis secondary to a T10 butterfly hemivertebra, diagnosed at 12 years of age,

treated conservatively until he developed intractable dorsal pain, paresthesias, and numbness on his lower extremities.

This case report aims to show an option to resolve this complex problem with a one-stage posterior surgical technique obtaining good clinical and radiological results. We prepared this article following the CARE Guidelines.

Material and Methods

The patient approved the publication of his case, signed the authorization form of the Informed Consent, and sent it to us through email. Confidentiality was fully protected. Clinical and radiological patient information is presented in an anonymized form.

Case Presentation

A 24-year-old Latin American male has been diagnosed with T10 butterfly posterior hemivertebra. This diagnosis was made when he was 12 years old based on X-rays and computed tomography scan (CT scan). He has been undergoing treatment with anti-inflammatory medications and physical therapy, with regular monitoring of the progression of his spinal deformity through X-ray imaging during his adolescent years.

Ten months prior to the medical consultation, the patient began experiencing progressive back pain. Three months before the consultation, he developed paresthesias and numbness in his legs, which were aggravated by prolonged sitting. A rigid kyphotic back deformity was noticed on the physical examination, with pain at palpation on the apex. No neurological findings (myelopathy or radiculopathy) were observed; neither had renal, cardiovascular, or pulmonary abnormalities.

X-ray assessment revealed an abnormal T9-T11 kyphosis. The coronal balance was neutral, and the sagittal balance was negative (Figure 1A-B). Sagittal magnetic resonance imaging (MRI) shows the T10 butterfly vertebra characteristics (Figure 1C). Focused dynamic X-rays showed a deformity reduction from 48º to 40º with hyperextension over a bolster (Figure 2A-B). The CT scan confirmed a T10 hemivertebra abnormality with a butterfly morphology in the sagittal and coronal projections, with a local T9T11 kyphosis of 48º, without any other abnormal findings (Figure 3A-B).

The patient was disabled based on patient-reported outcome measurement (PROM). His Oswestry Disability Index (ODI) score was 44, his 36-item Short Form Health Survey (SF-36) score was 47, and his low back pain score was 7 on the Visual Analogue Scale (VAS).

Based on clinical and image findings it was decided to treat the patient surgically with a single posterior procedure, with a complete T10 hemivertebrectomy and instrumented fixation from T8 to T12.

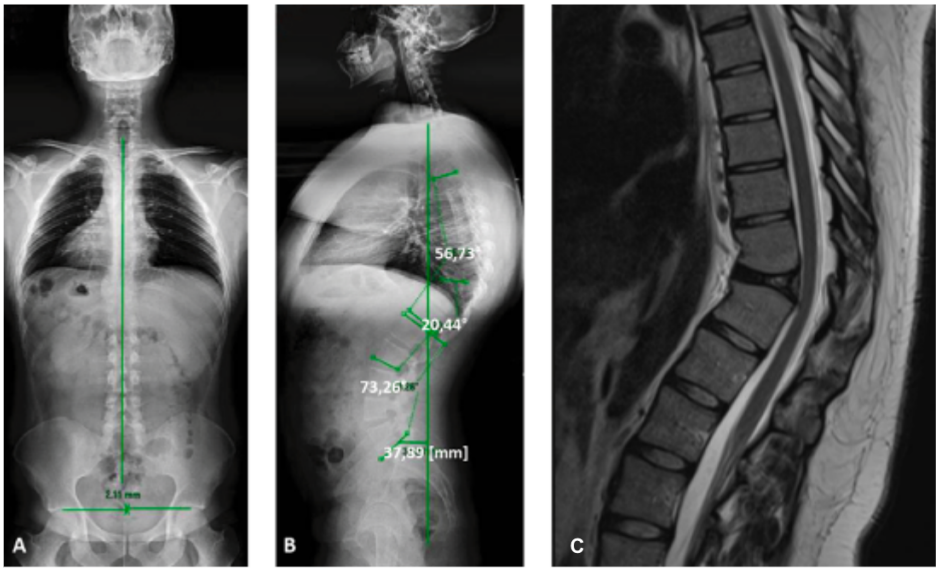

Figure 1: Anterior-posterior X-ray shows an aligned spine (A). Lateral X-ray shows T10 butterfly vertebra with abnormal T9T11 thoracic kyphosis. It also observed 37 mm negative sagittal balance, 73° of lumbar lordosis, 56° of thoracic kyphosis (B). Sagittal magnetic resonance imaging shows the T10 butterfly vertebra characteristics (C).

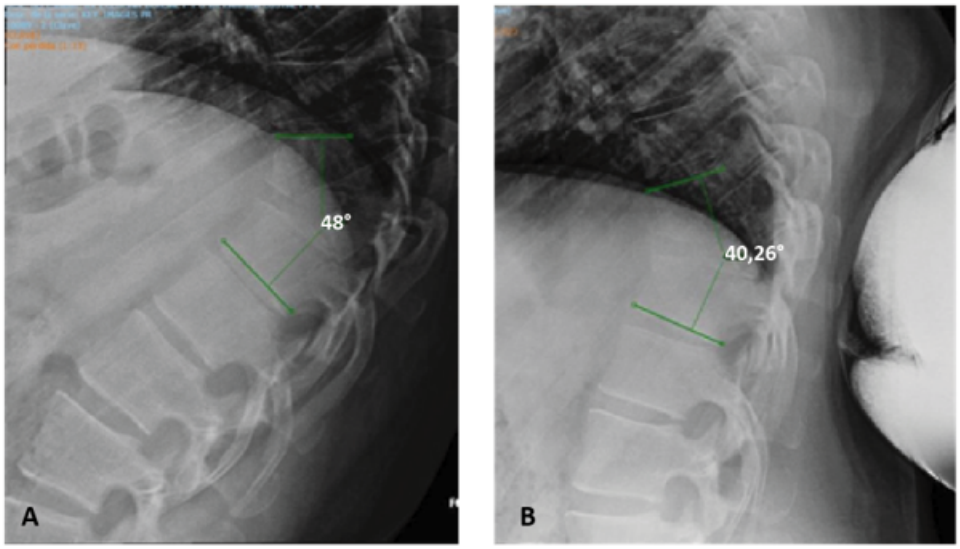

Figure 2: Focused pre-operative dynamic X-rays shows T9T11 kyphotic angle of 48˚on flexion view (A) and 40˚ on extension view (B)

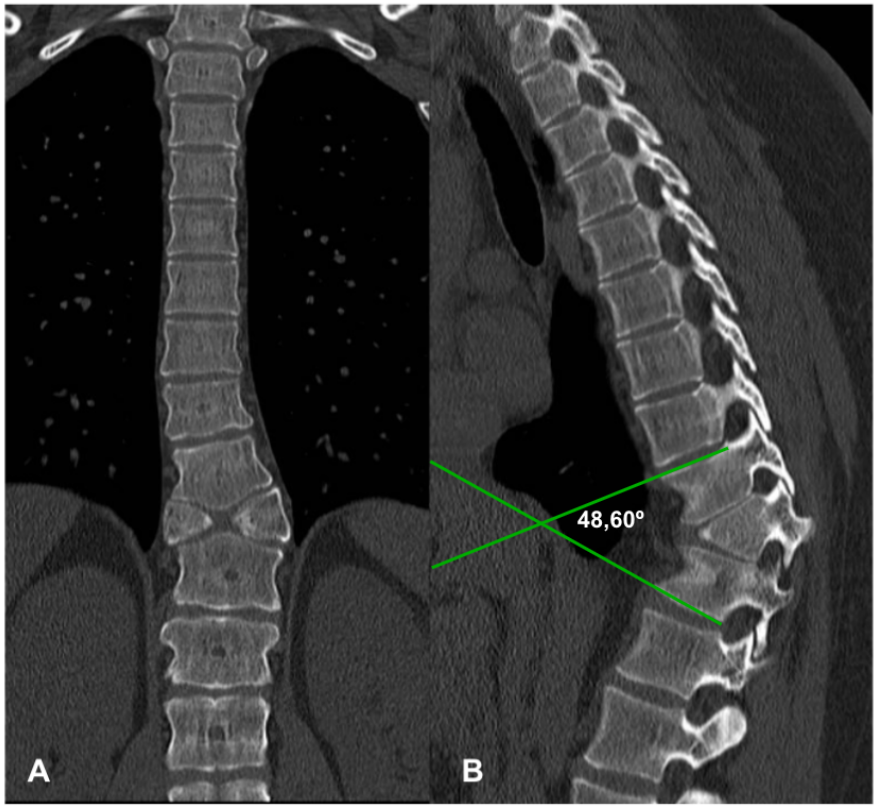

Figure 3: CT scan of the dorsal spine, with coronal (A), and sagittal (B) section with a T10 hemivertebra abnormality with a butterfly morphology.

Surgical Treatment

Under general anesthesia in a prone position with intraoperative neurophysiologic monitoring (IONM), the spinous processes and laminae from T9 to T11 were removed through a posterior midline incision. The O-arm navigation system guided the instrumentation positioning the monoaxial screws at T8, T9, T11, and T12 vertebrae.

Transpedicular decancellation with complete hemivertebra resection ("eggshell" procedure) was performed through a bilateral costotransversectomy without IONM alarms. The dural sac and bilateral T9 and T10 roots were fully exposed (Figure 4). Finally, each side of the instrumentation was connected with a rod bar using controlled compression to close the gap formed by the hemivertebrectomy, obtaining a satisfactory deformity reduction. Posterolateral arthrodesis was done with autografts from the iliac crest and posterior arch after decortication from T8 to T12. Pedicle screw positioning and kyphotic correction were confirmed by intraoperative CT scan. Surgical time was 330 minutes, and blood loss was 1100 ml. Surgery was finalized without complications. The patient was immobilized with a TLSO brace during the first 1-month postoperative period.

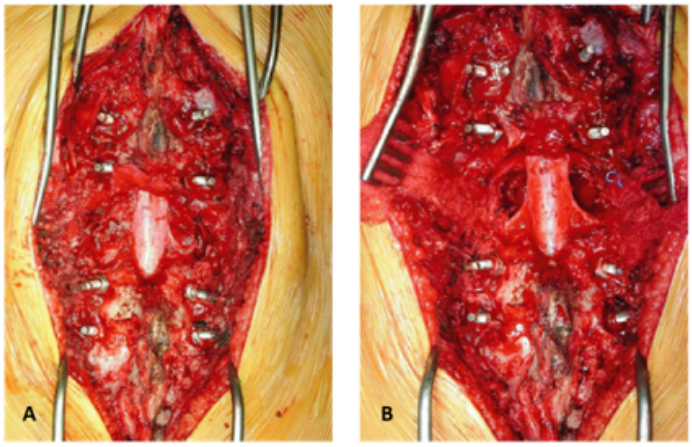

Figure 4 (A, B): Intraoperative image: Laminectomy from T9 to T11 and transpedicular decancellation with complete excision of T10 hemivertebra, and instrumented fixation from T8 to T12

Follow-Up and Outcome

The patient evolved neurologically intact, and his posture significantly improved. No wound complications or other surgical or medical complications were presented. The patient progressed favorably after a regular rehabilitation program and ambulated independently without pain. The patient was discharged from the hospital five days post-surgery

Based on X-rays the post-operative thoracolumbar angles and spinopelvic parameters were: 48º dorsal kyphosis, 18º T9T11 kyphosis, 63º lumbar lordosis, 54º pelvic incidence, 36º sacral slope, 19º pelvic tilt, 1 mm coronal balance and 12 mm negative sagittal balance. The CT scan assessment demonstrated a significant kyphotic T9T11 correction of 34° (48° pre-operative to 14° postoperative), with better global dorsal kyphosis and lumbar lordosis, restoring the sagittal balance (Figures 5 and 6).

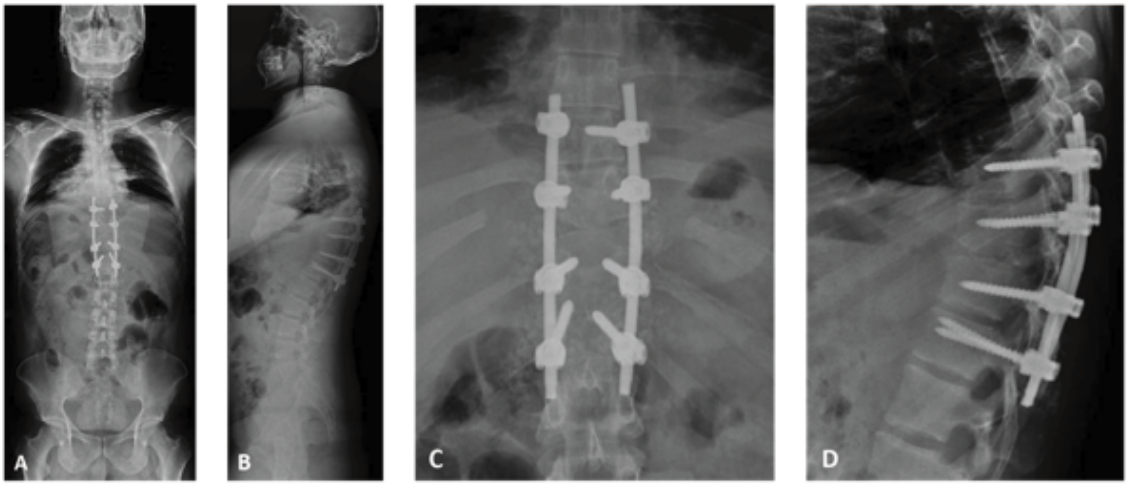

Figure 5: Postoperative total anterior-posterior and lateral X-rays (A and B) and focused thoracolumbar spine (C and D).

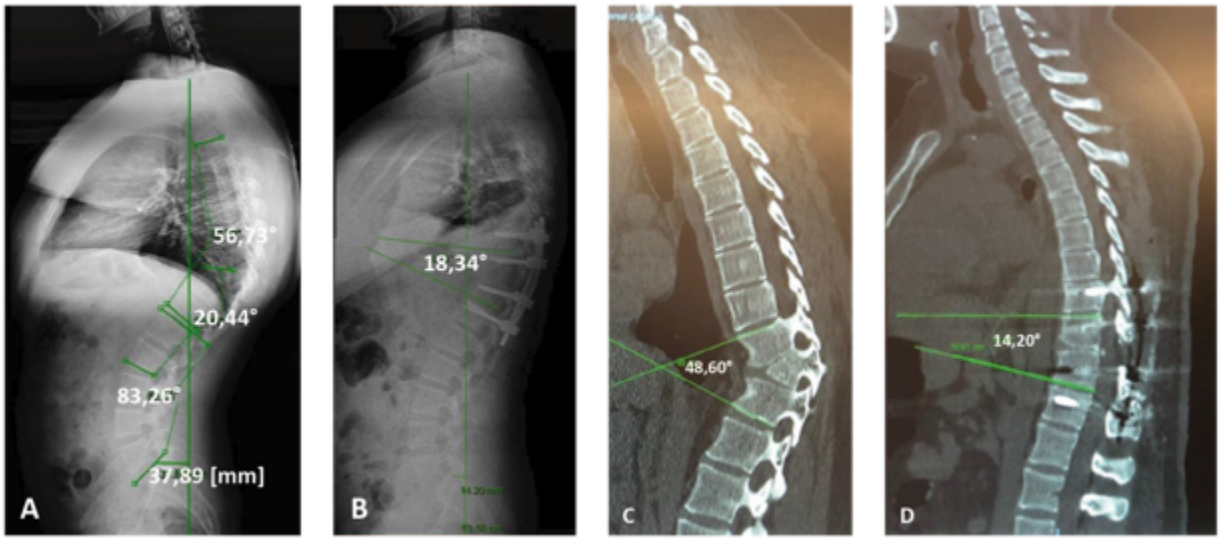

Figure 6: Pre and Postoperative X-rays show a significant thoracic deformity improvement (A and B). Pre and Postoperative CT scan show a T9T11 kyphotic correction from 48° to 14° (C and D).

Follow-up was made at one, three, six, and twelve months and up to 4 years postoperatively without complications or adverse events reported. The radiographic study demonstrated maintenance of the post-operative correction with screws and bars in a good position and no signs of screw loosening or failure.

At the 6-month follow-up, CT images showed complete fusion from T8 to T12. Clinical outcomes were evaluated using Oswestry Disability Index (ODI), 36 Item Short Form Health Survey (SF-36) and Visual Analogue Scale (VAS score). Significant improvement can be seen in the patient-reported outcome measurement (PROM) comparing pre versus post-operative. At the 4-year follow-up, the patient continued to exhibit positive results post-surgery (Table 1).

	Pre	6 Months Post	1 year Post	4 years
ODI score	44/100	32/100	12/100	15/100
SF-36 score	47	60	73	75
VAS score Back	7/10	3/10	1/10	2/10
VAS score Leg	0/10	0/10	0/10	0/10

Table 1: Outcome measures pre-operative, 6 months, 1 and 4 years post-surgery. (ODI: Oswestry Disability Index; SF-36: 36 Item Short Form Health Survey; VAS: Visual Analogue Scale)

Discussion

Congenital kyphosis is an uncommon sagittal plane deformity resulting from vertebral anomalies that impair the longitudinal development of the anterior vertebral body. Winter [3] and McMaster [4] classified it into three types: Anterior failure of vertebral body formation (I); Anterior failure of vertebral body segmentation or anterolateral bars (II); and a mix of both: failures of formation and segmentation (III). McMaster's study found a prevalence of 61% of the cases for Type I.

According to the pattern of failure of the vertebral body formation, deformities are divided into posterolateral quadrant vertebra, butterfly vertebra, posterior hemivertebra, and anterior or anterolateral wedge vertebra.

Winter and McMaster's [3,4] studies found that the risk of neurological compromise is substantially higher than in congenital scoliosis, and the limited blood circulation of the upper thoracic spine increases its susceptibility to spinal cord impairment. In worst cases, paraplegia could result in 10 to 12%, which occurred on average at the age of 12 years.

When congenital kyphosis is produced by a failure of formation with an unsegmented bar, it can progress into a sharp angular kyphosis by as much as 10° per year [2]. McMaster [4] study authors determined that congenital kyphosis presents rapid progression during the adolescent growth spurt; they concluded that posterolateral deformity progressed by 2.5° annually before the age of ten and progressed by 5° per year thereafter. The formation defect in two adjacent vertebrae determined kyphotic progression much faster if the failure was in a single one [5].

Regarding the treatment, Mayfield et al. [6] showed that the use of a brace would not prevent the progression of the deformity; instrumentation alone or with simple posterior soft tissue release is insufficient to correct these types of deformities, so spinal osteotomies are needed to obtain a successful alignment.

The main objective of surgical treatment for spinal deformity is to provide adequate sagittal and coronal balance without causing a neurological deficit. Currently, we have different techniques for the correction of kyphosis as closing wedge osteotomy (CWO), pedicle subtraction osteotomy (PSO), closing-opening wedge osteotomy (COWO), and posterior vertebral column resection (PVCR). Some studies have demonstrated that these procedures can provide correction rates of 53 to 89% depending on the technique used [7-9].

Risk factors for surgical complications include complex procedures (such as combined anterior-posterior surgery), severe deformities, previous cord compression, and older age [10-11].

According to the literature reviewed, few reports investigated the outcomes of surgical treatment of congenital kyphosis in adults, focusing mainly on the pediatric and adolescent populations. We present a 24-year-old male with a T10 butterfly hemivertebra that complains of back pain, reduced mobility of the spine in extension, paresthesias, and numbness, without neurological examination anomalies.

Our main challenge was defining the indication for surgical treatment and the technique to use. Wang et al. [12] reported a retrospective assessment of 24 patients with an average age of 13 years, treated with a posterior-only vertebral column resection obtaining a correction of the segmental kyphosis of more than 60° with a high rate (16.6%) of significant complications: incomplete spine injury, root injuries, proximal junctional kyphosis, and screws pull-out. Arriagada et al. [13] reviewed seven patients (1-7 years old) treated for congenital kyphoscoliosis with hemivertebra resection via "eggshell" procedure (transpedicular decancellation), without significant complications but with an inferior correction magnitude. In our case, we performed a posterior-only procedure with complete hemivertebra excision after transpedicular decancellation, and O-arm navigated guided instrumentation from T8 to T12, obtaining a satisfactory deformity correction and good functional results without perioperative complications.

We recognize the limitations of a case report and its restrictions on providing scientific data. However, we want to emphasize that congenital kyphosis is a rare complex pathology that requires demanding surgical techniques, where spinal cord and radicular injuries remain potential catastrophic complications. We believe that our results during the four-year follow-up are due to an adequate selection of the surgical technique and the use of technological aids such as intra-operative CT scans and navigation.

This is a starting point for more thorough prospective multicenter clinical studies to assess adult patients treated with a posterior-only hemivertebra resection technique and to characterize the role of intraoperative imaging aids in terms of facilitating and making the procedure safer.

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Conflicts of Interest: The authors declare no conflict of interest.

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