




# Revision surgery after instrumental fixation in patient with butterfly vertebra: a case report

Jiheng Zhan<sup>1</sup> · Yufeng Wang<sup>2</sup> · Bolai Chen<sup>2</sup> · Dingkun Lin<sup>1</sup> 

Received: 12 April 2018 / Accepted: 7 September 2018 / Published online: 15 September 2018  
© Belgian Neurological Society 2018

**Keywords** Butterfly vertebra · Revision surgery · Osteotomy

## Introduction

Butterfly vertebra consists of two hemivertebrae with a sagittal cleft between them, resulting in the special appearance of butterfly wings on anteroposterior spine radiograph. This rare congenital anomaly is not only been reported in clinical cases, but also in dry bone specimens from archaeological excavations. To date, most clinical case reports on butterfly vertebra have mainly focused on the imaging findings, and few studies discuss on its treatment strategy. Notably, a minority of patients may experience slowly progressive spinal deformity after the initial surgery due to the great growth potential of butterfly vertebra and implant failure. Many of these patients suffer from further deterioration of symptoms with time, thus require repeat surgical intervention. Our objective was to determine the causes of failure of primary surgery and to explore new treatment strategy. Herein, we report an unusual case of butterfly lumbar vertebra underwent revision surgery and perform a review of the literature concerning this condition. To the best of our knowledge, this is the first case report issued on the radiographic and clinical outcomes of patient underwent secondary surgery for butterfly lumbar vertebra.

## Case report

A 34-year-old man was first diagnosed with butterfly L2 vertebra 15 years earlier, and underwent short-segment transpedicular fixation at the local hospital. Following an initial excellent result with correction of spinal deformity, he developed recurrent back and leg pain since half a year ago. The pain typically became worse in the evening and was mildly improved with analgesics. In the physical examination, the internal fixation protruded to the surface of the waist skin without obvious red and swollen (Fig. 1), and the range of motion for lumbar flexion–extension was limited. As shown in anteroposterior radiograph of lumbar spine, the implant failed and accompanied by mild kyphosis of the thoracolumbar spine (Fig. 2). Computed tomography (CT) showed a sagittal-cleft in the body of L2 vertebra, separating two unconnected wedge-shaped vertebral bodies (Fig. 3a, b). Magnetic resonance (MR) of lumbar spine was performed due to the clinically evident lumbar radicular pain, and images showed a funnel-like sagittal cleft in L2 vertebral body, as well as adjacent segment degeneration (Fig. 3c, d).

The repeated procedure was performed along the original surgical incision. Dense scar tissue proliferated extensively and tightly adhered to normal fascia. The screw cap of the left L1 pedicle loosed and displaced upward, and there was bony fusion at the bilateral lumbar facet joints of L1–L3 vertebra bodies. Pedicle subtraction osteotomy (PSO) and Smith-Petersen osteotomy (SPO) were applied for correction of kyphosis by a single posterior approach (Fig. 4). Briefly speaking, the bilateral pedicles and posterior elements of L2 were excised first, then removed partial facet joints of T12 to L5 vertebra. The spinal cord and the nerve roots were identified and protected. Gradual correction of kyphosis was achieved by replacing the pedicle screws and installing pre-bending rods.

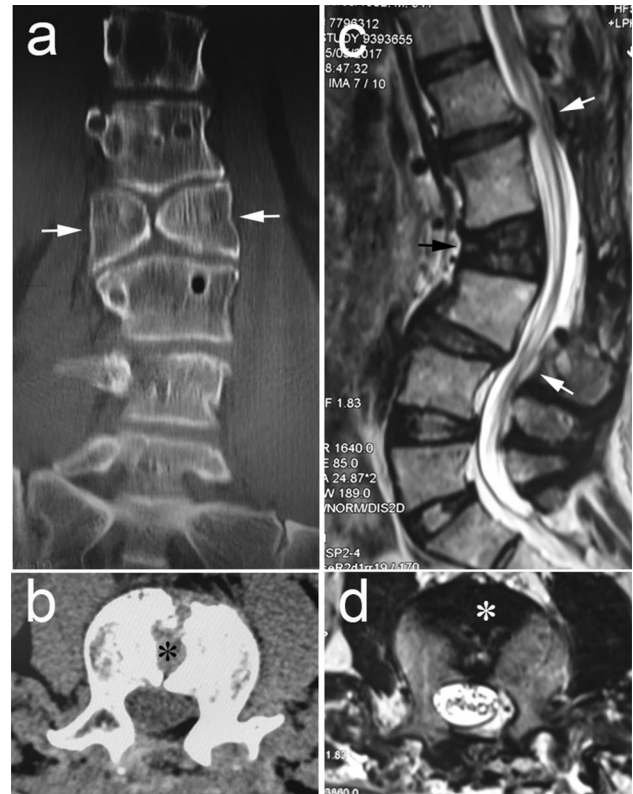
✉ Dingkun Lin  
lindingkun@126.com

<sup>1</sup> Second College of Clinical Medicine, Guangzhou University of Chinese Medicine, Guangzhou, China

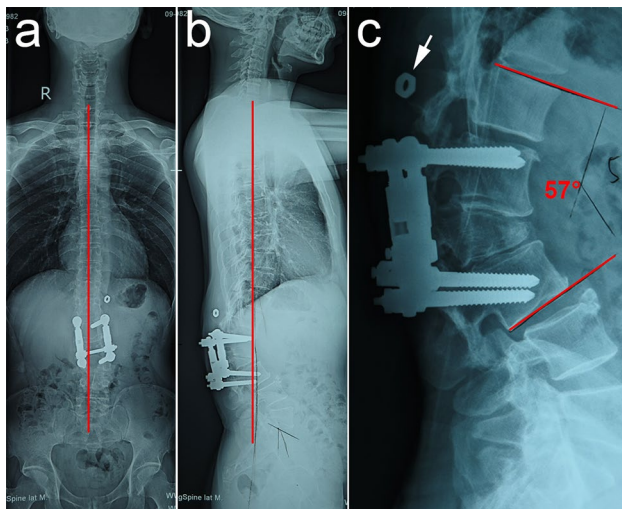
<sup>2</sup> Department of Spine Surgery, Guangdong Provincial Hospital of Chinese Medicine, Guangzhou, China



**Fig. 1** Preoperative appearance of the patient. **a, b** A mild thoracolumbar kyphotic deformity. **c, d** The internal fixation protrudes to the surface of the waist skin without obvious red and swollen (black arrows)



**Fig. 3** Computed tomography (CT) and magnetic resonance (MR) imaging of the lumbar spine. **a** Coronal CT image of the bone window demonstrates the L2 vertebra consists of two hemivertebrae (white arrows) with a sagittal cleft between them, **b** axial cuts showing sagittal cleft (black asterisk) at the L2 level. **c** Lumbar spine sagittal T2-weighted image showing adjacent segment degeneration at T12/L1 and L3/L4 (white arrows), **d** axial cuts at the L2 level showing a funnel-like sagittal cleft (white asterisk) separates two unconnected wedge-shaped vertebral bodies



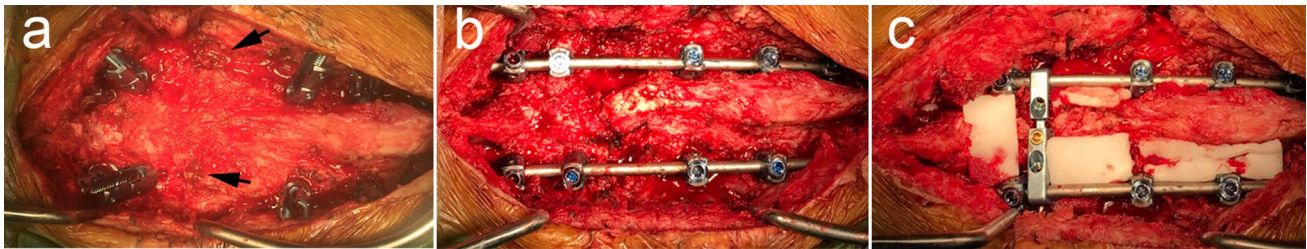
**Fig. 2** Full-length anteroposterior and lateral spine radiographs before revision surgery. **a, b** Kyphosis of thoracolumbar spine. **c** The screw cap of the left L1 pedicle loosened and displaced upward (white arrow)

The patient was mobilized on the fifth postoperative day with a rigid spinal brace and brace was continued 3 months to promote spinal fusion. There was obvious alleviation of low back pain without any neurological or vascular complications at 6-month follow-up. His range of motion of lumbar spine was preserved and physiological curvature of

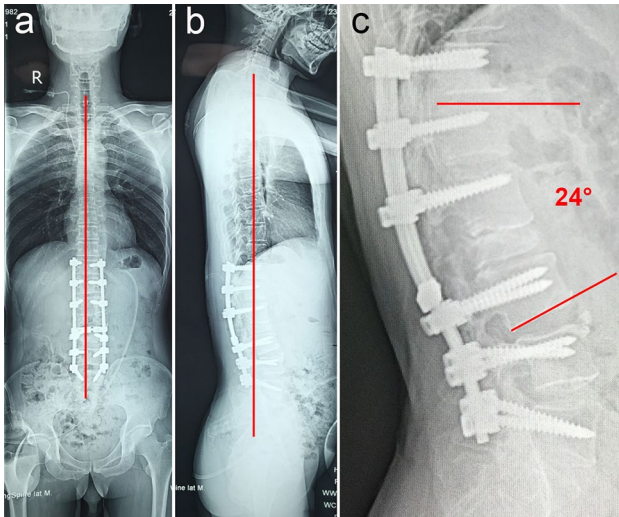
thoracolumbar spine was maintained. A 1-year follow-up radiograph showed good coronal and sagittal plane alignment of the spine following repeated operation. Additionally, the angle in thoracolumbar kyphosis has reduced from 57° to 24° after revision surgery (Fig. 5).

## Discussion

Butterfly vertebra, considered to be incidental and usually asymptomatic, can give rise to chronic low back pain closely related to abnormal spinal curvatures. Hopkins et al. indicated that axial pain is mostly related to the biomechanical alteration caused by butterfly vertebra [4]. Cho et al. confirmed by discography that the disc fragments that cause lumbosacral radicular pain originate from the sagittal cleft [1]. Previous studies suggested that single-stage partial vertebra resection, interpedicular graft, and instrumentation via a posterior approach could represent an efficient and safe



**Fig. 4** Intraoperative images of the patient. **a** Application of L2 PSO by a single posterior approach (black arrows). **b** Installation of double pre-bending rods and orthopedic. **c** Complete correction of kyphosis



**Fig. 5** Postoperative anteroposterior and lateral radiographs. **a**, **b** Images showing good coronal and sagittal alignment of the spine. **c** The kyphosis angle of thoracolumbar has reduced from 57° to 24° after revision surgery

treatment for symptomatic butterfly vertebra [2, 6]. However, very few research on the causes for the failure of initial operation and progressive deformity and the specific revision procedure.

The causes of internal fixation failure in this case can be summarized as follows. The patient's age at the time of primary surgery has missed the optimal correcting timing. And butterfly vertebra has intact intervertebral discs and endplates, thus exhibiting great growth potential and lead to the progression of spinal deformity. In other words, the first operation should have been a butterfly vertebra resection or at least a posterior epiphysiodesis, which can eliminate the asymmetric growth of the anterior and posterior column of spine, not just the simple posterior short-segment fixation. The patient had been engaged in physically demanding work for a long time, and the intensive stress load of thoracolumbar segment accelerated the wear of pedicle fixation. The kyphosis is mainly located between the fixed thoracic vertebrae and the active lumbar

vertebrae, and this biomechanical difference may lead to implant failure. Moreover, the level of diagnosis and treatment of this disease 15 years ago was limited, especially the development of orthopedic surgery was not mature enough.

Pseudarthrosis, instrumentation failure, progressive deformity, and exacerbation of clinical symptoms are the most common causes of revision spine surgery [7]. Individualized revision surgical intervention should be developed based on the initial surgical procedure and the specific cause of the failure. This patient with failed lumbar fusion showed progression of thoracolumbar kyphosis with adjacent segment degeneration. His kyphotic deformity was caused by single-segment vertebral disease, so combination therapy with PSO plus multi-segment PSO was reasonable and efficient [5]. Under the premise of ensuring corrective effect, the operation time is shortened and excessive blood loss is avoided using modern surgical techniques. The number of fusion segments in spinal surgery has long been controversial. Recently, a retrospective observational study revealed that multilevel vertebral osteotomies with instrumented fusion provides excellent restoration of sagittal and coronal balance in adults with spinal deformity compared to the short-segment ones [3]. Given that the patient's main symptom was low back pain, long-segment fixation, which effectively reconstruct three-dimensional alignment of the spine, seems to be the best choice.

In summary, this case report suggests that epiphysiodesis should be done during the initial operation. If internal fixation fails, individualized revision surgery should be developed based on the specific cause of failure. This case report showed that a satisfactory clinical result was achieved in kyphosis owing to butterfly vertebra by PSO combined with multi-segment SPO, which may be reliable and effective for the reconstruction of spinal balance.

**Funding** This study was supported by the National Natural Science Foundation of China (no. 81673992) and the Guangdong Provincial Science and Technology Department Program Foundation of China (no. 2016A020226008).

## Compliance with ethical standards

**Conflict of interest** The authors declare that they have no conflict of interest.

**Ethical approval** All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

**Informed consent** Informed consent was obtained from the patient involved in this study.

## References

1. Cho HL, Kim JS, Paeng SS, Lee SH (2011) Butterfly vertebra with lumbar intervertebral disc herniation. *J Neurosurg Spine* 15:567–570
2. Cui G, Watanabe K, Ishii K, Toyama Y, Chiba K, Matsumoto M (2011) Interpedicular graft using a titanium mesh cage in a patient with lumbar scoliosis associated with a congenital butterfly vertebra. *J Neurosurg Spine* 14:215–218
3. Ghobrial GM, Lebowitz NH, Green BA, Gjolaj JP (2017) Multi-level Schwab grade II osteotomies for sagittal plane correction in the management of adult spinal deformity. *Spine J* 17:1594–1600
4. Hopkins RM, Abbott Jh (2015) Congenital ‘butterfly vertebra’ associated with low back pain: a case report. *J Man Manip Ther* 23:93–100
5. Park JH, Hyun SJ, Kim KJ, Jahng TA (2017) comparative study between pedicle subtraction osteotomy (PSO) and closing-opening wedge osteotomy (fish-mouth PSO) for sagittal plane deformity correction. *Spine (Phila Pa 1976)* 42:E899–E905
6. Patra DP, Salunke P, Pramanick G (2013) Butterfly C2 vertebra: an unusual finding in a case of os odontoideum. *Pediatr Neurosurg* 49:320–323
7. Zhu F, Bao H, Liu Z, Bentley M, Zhu Z, Ding Y, Qiu Y (2014) Unanticipated revision surgery in adult spinal deformity: an experience with 815 cases at one institution. *Spine (Phila Pa 1976)* 39:B36–B44