



Postoperative screw pullout of severe spondylolisthesis in osteogenesis imperfecta: a case report with 3-year follow-up

Zhongtai Zhang, MD^a, Feng Ling, MD^c, Kangwu Chen, MD, PhD^a, Yuxuan Liu, MD^b, Qingfeng Ding, MD, PhD^{a,*}, Zhigang Zhang, MD, PhD^{a,*}

Introduction and importance: Osteogenesis imperfecta (OI) is a rare skeletal disorder characterized by bone fragility and deformities in both paediatric and adult populations. The occurrence of severe spondylolisthesis in OI patients is even more infrequent. However, there is no consensus regarding the optimal treatment approach for OI patients afflicted with severe spondylolisthesis. The selection of surgical procedures and the effective management of postoperative complications present significant challenges in this context.

Case presentation: A 30-year-old male patient diagnosed with OI type IV (Sillence classification) underwent the lumbar laminectomy and postero-lateral fusion due to severe spondylolisthesis (grade III). Following the surgery, the patient experienced postoperative screw pullout while on bedrest. However, aside from experiencing back pain, there were no neurological symptoms present. To address this issue, the patient received salvage treatment in the form of cast immobilization combined with bisphosphonates. At the 3-year follow-up, the patient exhibited absence of sciatic nerve pain and reported mild numbness in the lower extremities. Moreover, the patient demonstrated the ability to ambulate a distance exceeding 1500 m. Nevertheless, the persistence of sexual dysfunction was observed.

Clinical discussion: This study presented the initial instance of surgical complications observed in patients with severe spondylolisthesis and OI. This highlights the importance to exercise meticulous caution and thoroughness when assessing surgical interventions.

Conclusion: In cases where the fixation fails to offer adequate biomechanical stability, the administration of bisphosphonates and robust immobilization remains crucial, even in the presence of complications.

Keywords: case report, immobilization, osteogenesis imperfecta, postoperative complication, spondylolisthesis

Introduction

Osteogenesis imperfecta (OI) is a heritable spinal disease characterized by multiple fractures caused by a genetic mutation in Collagen I. The incidence is estimated to be ~1 in 10 000–20 000 births^[1,2]. OI is classified into various types, with the Sillence classification being the most widely used system. This classification

^aDepartment of Orthopedics, The First Affiliated Hospital of Soochow University,

^bThe Affiliated Stomatological Hospital of Soochow University, Suzhou

Stomatological Hospital, Suzhou and ^cDepartment of Orthopedics, Jiangsu Taizhou People's Hospital, Taizhou, Jiangsu Province, China

Z.Z., F.L. contributed equally to this study and share first authorship.

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*Corresponding authors. Address: Department of Orthopedics, The First Affiliated Hospital of Soochow University, 188 Shizi Street, Suzhou, Jiangsu Province 215000, China.

Tel.: +86 0512 6778 0249; fax +86 0512 6778 1165. E-mail: ddt0222@163.com

(Q. Ding), and Tel.: +86 0512 6778 0101; fax +86 0512 6778 1165.

E-mail: zhg_zhang@outlook.com (Z. Zhang).

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HIGHLIGHTS

- Osteogenesis imperfect with severe spondylolisthesis and early fixation failure is extremely rare.
- Surgeons must exercise meticulous caution when assessing surgical appropriateness.
- Normal or below-normal level of activity may contribute to internal fixation failure.
- Biomechanical stability holds significant importance for osteogenesis imperfecta patients after surgery.
- Cast immobilization is a viable alternative for complications of internal fixations.

divides OI into four types (type I–IV) based on clinical and radiographic features^[3]. In 2000 and 2002, three additional types (type V–VII) were incorporated into the classification system^[4–6]. However, there is a scarcity of reports regarding the treatment of spondylolisthesis in individuals with OI. In 1963, Newman and Stone documented a case of a 6-year-old girl afflicted with OI and spondylolisthesis^[7]. Rask *et al.*^[8] later reported a case of a 40-year-old man with grade II spondylolisthesis in the lumbosacral joint resulting from a pars defect. Basu *et al.*^[9] presented two cases of spondylolisthesis (grade III and IV) attributed to significant elongation of pedicles. Additionally, Ivo's report indicated that one out of every three OI children with spondylolisthesis underwent lumbar postero-lateral fusion^[10]. In a retrospective study conducted in 2014, a total of 195 patients diagnosed with OI were

included. Among these patients, 19 cases of spondylolisthesis were identified^[11]. To the best of our knowledge, there is a lack of literature reporting postoperative surgery-related complications in OI patients with spondylolisthesis.

In our clinical practice, we encountered a rare case of OI with severe spondylolisthesis, in which the patient experienced screw pullout following posterior lumbar fusion. The management of such exceptional cases has not been previously discussed. Therefore, this case report aims to describe this rare occurrence and provide insights based on our experiences.

This case report has been reported in line with the SCARE 2023 criteria^[12].

Case report

This is a 30-year-old male patient who diagnosed with OI type IV (Sillence classification). The family history was normal, he was born by natural delivery with no siblings. In his trauma history, the patient suffered from several fractures in the left elbow, left ankle and right femoral shaft after minor trauma in adolescence. All these fractures were treated conservatively. The patient also underwent a stent implantation in the iliac artery due to the deep venous thrombosis at the age of 28.

Also, at 28 years of age, the patient strained his waist when shifting heavy loads. He complained about lower back pain and limited mobility. All symptoms were relieved after 3 days' bedrest. Nevertheless, 2 months later, progressive weakness and numbness in the left lower limb appeared, which affected his walking ability. Nineteen months after that, this patient developed additional sciatic pain, sexual dysfunction as well as the dysfunction of urination and defecation. He was then referred to orthopaedic department of our hospital. The computed tomography (CT) and MRI showed spondylolisthesis (grade III) of the fifth lumbar vertebrae (L5) and spinal stenosis, furthermore, both the fifth lumbar vertebrae and the first sacral vertebra (S1) showed different degrees of abrasions. (Fig. 1A) The physical examination showed the lumbosacral kyphosis, mild tenderness in L5–S1, loss of perineal sensation and perianal reflex. The straight leg raising test: left 70°, right 70°; the hypaesthesia presents in both plantar and lateral side of the dorsal feet, the strength of left ankle dorsiflexors was grade IV. The deformity of left elbow was obvious, the flexion/extension of the elbow was 120°–40°. The hypaesthesia can be found in the lateral elbow, but no local tenderness or percussion pain existed, there was no ankle clonus and pathological sign.

To relieve the relative symptoms, a lumbar laminectomy and postero-lateral fusion was suggested. However, because of the abrasion, there was no enough space for S1 screws. An additional pelvic fixation is not suitable due to the poor bone quality of OI patient, which may lead to additional surgical trauma and unpredictable fractures. Therefore, we placed screws in L4, L5 and S2 in this surgery and require postoperative bedrest immobilization. The surgery proceeded uneventfully uneventful without any intraoperative complications. After the surgery, the lumbar pain, sciatic pain and numbness in the lower limb had improved remarkably, but sexual dysfunction remains. The postoperative X-ray and CT scan showed a satisfactory reduction in L5/S1 (Fig. 1B). However, 13 days after the surgery, the patient felt a sudden back pain in the sacrococcygeal region during defecation in bed. Subsequent radiographs indicated screw

pullouts in the S2 (Fig. 1C), but the patient showed no signs of neurological symptoms other than the back pain. Due to his poor general condition, another revision surgery was not recommended immediately. As a result, to maintain the stability of L5/S1, cast immobilization was introduced. The patient was secured with plaster bandages from the buttocks to the chest for 3 months and was treated with bisphosphonates. After the cast immobilization, the patient got the pain relief and had no more complains. One year later, the instrumentations were removed in another surgery successfully. At the 3-year follow-up, The CT scan (3 years after the second surgery) showed the fusion of the L5 and S1 (Fig. 1D). The patient presented no pain in the sciatic nerve and experienced faint numbness in the lower limbs. He was able to walk for more than 1500 m. The dysfunction of excretion was greatly improved. However, the sexual dysfunction still remained a problem.

Discussion

OI, also known as osteogenesis imperfecta, is a rare skeletal disorder that is characterized by bone fragility and deformities in both children and adults. In patients with OI, spondylolisthesis is typically of low grade and can often be managed without surgical intervention^[13]. However, the number of OI patients with severe spondylolisthesis requiring surgery is relatively small. While several studies have reported on the occurrence of spondylolisthesis in OI patients, only Basu and Ivo have described a successful treatment^[9,10]. Basu's case involved the utilization of anterior and posterior fusion with posterior instrumentation, which resulted in a satisfactory outcome during the 3-year follow-up period. In Ivo's report, it was observed that the patient underwent a lumbar laminectomy and postero-lateral fusion, leading to a significant amelioration of their symptoms. To the best of our knowledge, this case represents the initial documentation of surgical complications in individuals with OI and spondylolisthesis.

We present the first instance of a patient with severe spondylolisthesis and spinal stenosis who experienced screw pullout following the surgical procedure. This particular case has not been previously addressed in the existing literature concerning OI patients. The occurrence of screw pullout in the early postoperative stage was infrequent, even in patients without OI, let alone those with rare OI. Nevertheless, despite its rarity, screw pullout holds considerable clinical significance in treating similar patients.

Various factors contributed to the occurrence of postoperative screw pullout. The primary cause may be attributed to the bone fragility observed in OI patients, wherein low bone mass results in structural deficiencies and a significant reduction in bone mineral density (BMD)^[14]. Furthermore, the mechanical integrity of bone in individuals with OI is diminished as a result of genetic abnormalities in type I collagen. The inadequate production of collagen adversely affects the material properties^[15]. Consequently, the fixation of slipped vertebrae using conventional screws becomes more challenging, primarily due to the instability observed within the vertebrae.

In order to mitigate potential complications, surgeons commonly employ an extended fusion technique to enhance biomechanical stability through increased points of fixation. Notably, certain studies suggest that osteoporosis patients should



Figure 1. (A) CT and MRI showed spondylolisthesis of the L5 (grade III) and spinal stenosis. (B) The postoperative radiography showed a satisfactory reduction in X-ray and CT. (C) The radiographs taken 13 days after surgery indicated screw pullout in the S2. (D) Three years after the second surgery, the X-ray and CT showed the fusion of the L5 and S1. CT, computed tomography.

consider utilizing a minimum of three fixation levels both above and below the identified segment^[16]. Additionally, the application of bone cement to reinforce pedicle screw fixation has proven effective in preventing screw pullout. Lehman and colleagues reported a significant 149% increase in pullout strength through the implementation of polymethyl methacrylate (PMMA) augmentation^[16,17].

Prior to the surgical procedure, we had taken into account the potential for instability associated with the use of three pairs of screws. However, due to limited alternatives, pelvic fixation was deemed necessary, despite the potential for increased surgical trauma and heightened risks of additional fractures. Consequently, we opted for this particular protocol in the treatment of the patient, necessitating postoperative bed rest.

In this particular case, the occurrence of screw pullout transpired shortly after the surgical procedure, with the accompanying symptom being limited to lower back pain rather than neurological manifestations. Given the patient's compromised physical state and bone integrity, formulating an optimal therapeutic approach poses considerable challenges. It is our belief that an additional surgical intervention would not alleviate the patient's complaint of back

pain alone, nor can we endorse bed rest as a viable solution due to the potential for inadequate immobilization of the lumbosacral joint, thereby increasing the risk of nonunion.

Therefore, we opted for cast immobilization and prescribed bed rest for a duration of 3 months. Previous literature has documented cast immobilization as a successful conservative approach for treating spine fractures due to its ability to maintain spinal stability, limit vertebral mobility, and facilitate fusion^[18]. Additionally, the patient underwent anti-osteoporosis therapy (medical treatment with bisphosphonates). Following the completion of all conservative treatments, the patient experienced complete alleviation of symptoms. During the three-year follow-up period, the patient exhibited a favourable outcome characterized by the absence of pain in the sciatic nerve and only minimal numbness in the lower limbs.

One of the limitations of this case report is the underestimation of the patient's bone mass weakness in the initial treatment plan. However, it also serves to validate the efficacy of bisphosphonates and cast immobilization, even in the presence of complications. This particular aspect highlights the intricacy involved in managing patients with OI.

Conclusion

This case raises concerns regarding the therapeutic approaches for individuals with severe spondylolisthesis in OI patients. It is imperative to thoroughly assess the surgical procedures, considering the potential utilization of longer fusion or bone cement augmentation to mitigate the risk of screw loosening or pullout. However, in cases where the fixation fails to offer adequate biomechanical stability, the administration of bisphosphonates and the implementation of potent immobilization remain crucial for promoting bony union, particularly in the presence of complications.

Ethical approval

The ethical approval is waived for case report in our institution. Written informed consent from the patient has been obtained for publication of this report.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

Z.Z.: patient follow-up, writing the paper and investigation. F.L.: patient follow-up, figure preparation and writing the paper. K.C.: investigation and supervision. Y.L.: manuscript editing and critical revision. Q.D.: study design and critical revision. Z.Z.: study design supervision and manuscript revision.

Conflicts of interest disclosure

The authors declare that they have no conflict of interest.

Research registration unique identifying number (UIN) (for case reports detailing a new surgical technique or new equipment/technology)

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Data sharing is not applicable to this article (No data included).

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