

Posterior Direct Reduction of Lateral Atlantoaxial Joints for Rigid Pediatric Atlantoaxial Subluxation

A Fulcrum Lever Technique

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Study Design. Clinical case series.

Objective. To present a surgical technique and results of posterior direct reduction of lateral atlantoaxial joints for rigid pediatric atlantoaxial subluxation (AAS) using a fulcrum lever technique.

Summary of Background Data. The surgical treatment of pediatric rigid AAS is still technically challenging. Several factors contribute to the surgical difficulty, such as small vertebrae, incomplete bone formation, dysplasia, the difficulty of reduction and external fixation are considered as a surgical daunting challenge. Herein, the surgical technique of posterior direct reduction of lateral atlantoaxial joints for rigid pediatric AAS using a fulcrum lever technique is presented.

Methods. This retrospective study included 10 pediatric patients with rigid AAS who underwent posterior direct reduction of bilateral C1/2 facet joints via a fulcrum lever technique. The indication for surgery was the presence of neurological symptoms and spinal cord atrophy with an intramedullary high signal at the C1 level on T2-weighted magnetic resonance (MR) images. The surgical procedure consisted of three steps: (1) opening and distraction of the C1/2 facet joints and placement of tricortical bone as a spacer and fulcrum; (2) placement of C1 and C2 screws; and finally, (3) compression between the C1 posterior arch and C2 lamina and constructing C1/2 fusion. All

patients underwent the neurological and radiological evaluations before and after surgery.

Results. Eight of 10 patients demonstrated genetic disorders, either Down syndrome or chondrodysplasia punctate. Besides, all cases documented congenital anomaly of the odontoid process. Bilateral C1 lateral mass screws were successfully placed in all cases. No evidence of postoperative neurovascular complications. Radiological evaluation showed the corrections and bony fusions of C1/2 facet joint in all cases.

Conclusion. The fulcrum lever technique for rigid pediatric AAS can be one of the effective surgical solutions to this challenging pediatric spinal disorder.

Key words: atlantoaxial subluxation, chondrodysplasia punctate, down syndrome, fulcrum lever technique, halo-vest orthosis, lateral atlantoaxial joint, lateral mass screw, pediatric spine, pedicle screw, posterior fusion, translaminar screw.

Level of Evidence: 4

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The atlantoaxial complex consists of extremely mobile 3 synovial joints, structurally weak, and located between two stiff structures (occipital–C1 and C2–3 joints).¹ Rigid atlantoaxial subluxation (AAS) in pediatric patients appears to be a dynamic process, and the dislocation may aggravate with time. As the center of gravity of the head shifts anteriorly, lordosis of the cervical spine increases in compensation. This further aggravates anterior slippage of the atlas over the axis. The atlas along with the head may slip anteriorly over the axis in a vertical orientation. Slippage between the atlas and axis will finally become rigid. The increased anterior angulation at the craniovertebral junction leads to compression of the cervicomedullary junction, giving rise to critical clinical symptoms. The surgical treatment of rigid pediatric AAS is still technically challenging. Rigid AAS often occurs in children with Down syndrome (DS), which might carry a high risk of myelopathy with severe neurological condition. Historically, the choice of surgical treatments includes anterior transoral decompression followed by posterior occipito-cervical fusion.^{2–5} However, the procedure may be technically demanding in pediatric patients because of

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TABLE 1. Lower and Upper Extremity Motor Function Based on the Neurosurgical Cervical Spine Scale (NCSS)

| Score | Function |
|--------------------------------|--|
| Lower extremity motor function | |
| 1 | Total disability: chair-bound or bedridden |
| 2 | Severe disability: needs support walking on flat surfaces, and unable to ascend or descend stairways |
| 3 | Moderate disability: difficulty walking on flat surfaces, and needs support ascending or descending stairways |
| 4 | Mild disability: no difficulty walking on flat surfaces, but mild difficulty ascending or descending stairways |
| 5 | Normal: normal walking, with or without abnormal reflexes |
| Upper extremity motor function | |
| 1 | Total disability: unable to perform daily activities |
| 2 | Severe disability: severe difficulty in daily activities with motor weakness |
| 3 | Moderate disability: moderate difficulty in daily activities with hand and/or finger clumsiness |
| 4 | Mild disability: no difficulty in daily activities, but mild hand and/or finger clumsiness |
| 5 | Normal: normal daily activities, with or without abnormal reflexes |

their small size, congenital variations, possible growth potential, and immature ossification.⁶ Furthermore, another concern is that posterior occipito-cervical fusion may result in severe restrictions to neck movements, and potential postoperative dysphagia or dyspnea.⁷ Recently, posterior direct distraction of the lateral atlantoaxial joints for irreducible AAS or basilar invagination has been shown to be possible.^{3,8–17} However, these prior techniques have a limited impact on rigid pediatric AAS. The intraoperative difficulty in reduction is likely attributed to the shape of the lateral atlantoaxial (C1/2 facet) joints in pediatric patients. C1/2 facet joints might be gradually deformed. Angulation of the facet joints causes progressive slippage of C1 over C2 with subsequent rigidity of the C1/2 facet joints. Intraoperative compression force without a fulcrum between C1 posterior arch and C2 lamina may cause deterioration of cord compression because of slippage of C1 over C2. To overcome these difficulties, we applied a fulcrum lever technique. Here, we present a case series of consecutive patients who underwent posterior direct reduction of C1/2 facet joints using a fulcrum lever technique for rigid pediatric AAS to discuss outcomes as well as describe the technique in detail.

MATERIALS AND METHODS

Patient Population

This retrospective study included 10 patients (six males, four females) with rigid AAS who were younger than 15 years old at the time of surgery. Mean age was 9.6 years old (range, 5–15 yrs). All surgeries were conducted during the 5-year study period between 2015 and 2019. Postoperative follow-up ranged from 10 to 57 months (mean, 31.2 mos). Rigid AAS was defined as an atlantodental interval (ADI) longer than 5 mm in the extension position on cervical lateral radiography. The indication for surgery was the presence of neurological symptoms and spinal cord atrophy with an intramedullary high signal at the C1 level on T2-weighted magnetic resonance (MR) images. All medical records were analyzed retrospectively on a computerized medical records system.

Clinical Assessment

All patients underwent a comprehensive evaluation before surgery, at postoperative day 1 (POD 1), 90 days (POD 90), and the most recent follow-up after surgery. Neurological status was assessed using the Neurosurgical Cervical Spine Scale (NCSS) (Table 1),¹⁸ nevertheless, sensory function was not included because of the difficulty in examining the sensory condition. The ADI was measured in the neutral position using cervical lateral radiography. The space available for the spinal cord at the C1 spine level (C1-SAS) was measured using cervical lateral radiography and reconstruction sagittal images of cervical computed tomography (CT) (Figure 1A). The C1 line was defined as that line connecting the centers of the C1 anterior and posterior arches. C1–2 height was defined as the distance between the C1 line and the midpoint of the C2 inferior surface (Figure 1A). The C2 line was defined as the line perpendicular to the posterior line of the odontoid process. The inclination angle at C1 (C1-IA) was defined as the angle between the C1 line and the C2 line (Figure 1A).¹⁶ The inclination angle at the C1/2 lateral joints (C1/2 LJ-IA) was defined as the angle between the McRae line and the C2 facet line in the neutral position (Figure 1B). Osseous bony fusion after surgery was evaluated strictly using CT and lateral dynamic radiographs of the cervical spine. Osseous union was defined as the presence of complete bony bridges in the C1/2 lateral joints or interlaminar space.

Surgical Techniques

The principle of the surgical procedure consisted of three steps: (1) opening and distraction of the C1/2 facet joints and placement of tricortical bone as a spacer and fulcrum (Figure 2A, B); (2) placement of C1 and C2 screws; and finally, (3) compression using Kocher forceps between the C1 posterior arch and C2 lamina and constructing C1/2 fusion (Figure 2C) (See Supplemental Video, <http://links.lww.com/BRS/B575>). Under general anesthesia, intubation was performed using a fiberoptic device without any manipulation to the neck. Patients were placed in the prone position. The head and cervical spine was fixed in the

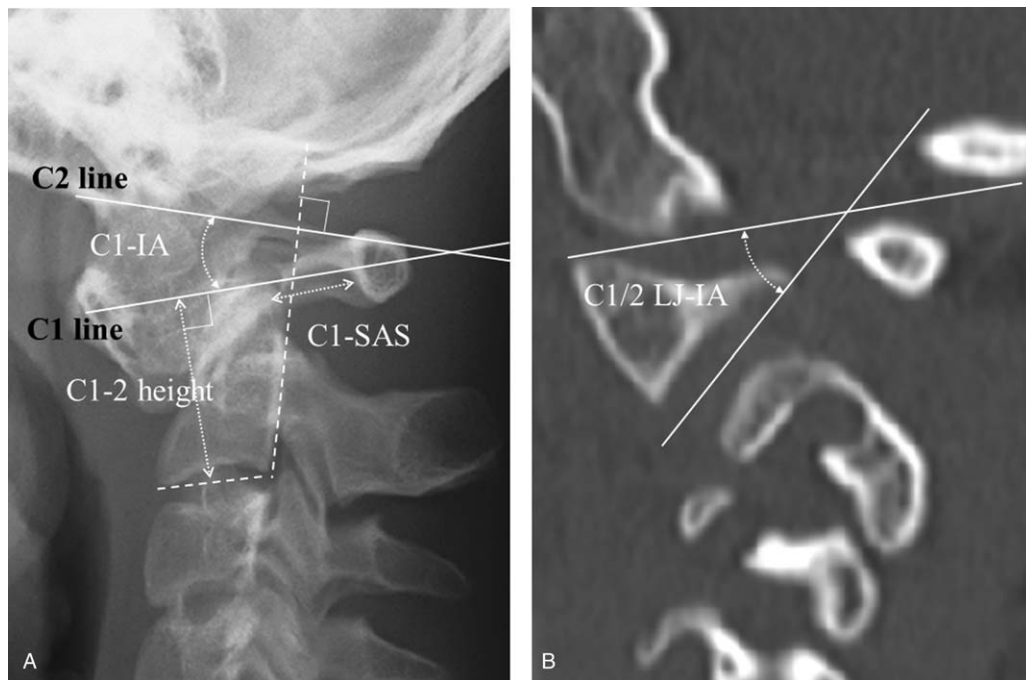


Figure 1. Radiological evaluation. The C1 line was defined as that line connecting the centers of the C1 anterior and posterior arches. The C2 line was defined as the line perpendicular to the posterior line of the odontoid process. Space available for the spinal cord at the C1 spine level (C1-SAS) was defined as the distance between C1 posterior arch and odontoid process. C1–2 height was the distance between C1 line and midpoint of the C2 inferior surface. Inclination angle at C1 (C1-IA) was the angle between C1 line and C2 line. Inclination angle at the C1/2 lateral joints (C1/2 LJ-IA) was defined as the angle between the McRae line and C2 facet line in the neutral position.

neutral position using fluoroscopic image guidance. Posterior elements of the upper cervical spine were carefully exposed. Bilateral C1/2 facet joints were widely exposed after bilateral transection of the C2 nerve roots at ganglia. C1/2 facet joints were usually tightly closed in case of rigid AAS. A thin raspatory was inserted and rotated in the joint space to open the joints. An interbody spreader was then carefully inserted to open the joint much more widely. Autologous bone grafts were harvested from the posterior iliac crest. The grafting bone size was calculated by the area of facet joint *versus* height using the preoperative CT. Tricortical iliac bone grafts were pushed into the joints as the joint spacer (Figure 3). Arterial flow in the vertebral artery was monitored carefully using a micro-Doppler system. Under full exposure of the C1 lateral mass, C1 lateral mass screws (Vue Point; NUVASIVE, CA) were placed on both sides under fluoroscopic image guidance. C2 screws were carefully placed. C2 pedicle screws were our first choice, when safely available. When the pedicle diameter was smaller than 3.5 mm, a C2 translaminar screw or laminar hook was selected as the alternative anchor screw. Autologous bone grafts were also placed into the space between decorticated C1 posterior arch and C2 lamina. Sublaminar polyethylene cables (NESPLON cable; Alfresa Pharma, Osaka, Japan) were placed under the C1 posterior arch and C2 lamina. Direct reduction of AAS was achieved by compression force between the C1 posterior arch and C2 lamina. Tricortical iliac bone was used as a fulcrum for leverage. Alignment of the midpoint of the C-1 posterior

arch and C-2 spinous process suggested complete reduction. When reduction of the atlantoaxial joints was achieved, the final tightening was completed. The entire procedure was performed under fluoroscopic guidance. All patients were electively ventilated overnight and slowly weaned off the ventilator and extubated the next day. In general, 3-month Halo-vest was applied postoperatively.

Statistical Analysis

All data are expressed as means \pm standard deviation. The Steel-Dwass multiple comparison was performed to assess differences in averaged values of radiological parameters among the preoperative state, POD 1, POD 90, and the most recent follow-up after surgery. JMP version 14 software (SAS Institute Japan, Tokyo, Japan) was used for all statistical analyses, with values of $P < 0.05$ considered significant.

RESULTS

All cases had no basilar invagination but odontoid dysplasia or os odontoideum. Case 9 showed C2–7 vertebral body dysplasia and cervical kyphosis. Eight of the 10 patients demonstrated a genetic disease as either Down syndrome or chondrodysplasia punctate (CDP). CDP subtypes were represented by three cases of X-linked CDP1 (CDPX1) and one case of brachytelephalangic CDP. Eight of the 10 patients presented with relatively short stature. On admission, two patients showed respiratory dysfunction and underwent tracheostomy. Preoperative assessments are summarized in Table 2.

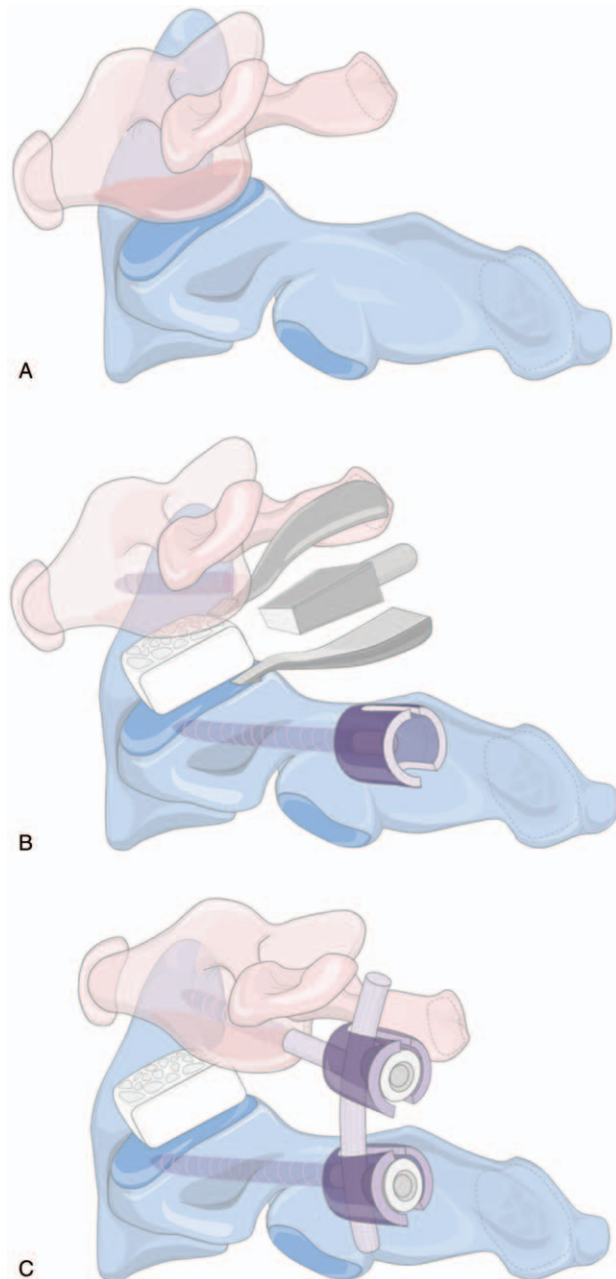


Figure 2. Schematic drawings showing the principle of the surgical procedure. **A**, Before surgery. **B**, Opening and distraction of lateral C1/2 facet joints and placement of tricortical bone as a spacer and fulcrum. Careful compression between C1 and C2 laminae after placement of C1 and C2 screws. **C**, Creation of a construct of C1/2 fusion.

Clinical Outcome

Eight of the 10 patients underwent posterior C1–2 reduction and fusion, and the remaining two patients underwent posterior C1–4 fusion and C1–Th1 fusion. Bilateral C1 lateral mass screws were placed in all cases. Bilateral C2 screws were placed into the laminae or into the short pars in eight cases. A C2 laminar hook combined with translaminar screw was placed in one case, and bilateral C2 laminar hooks were placed in one case. No cases demonstrated

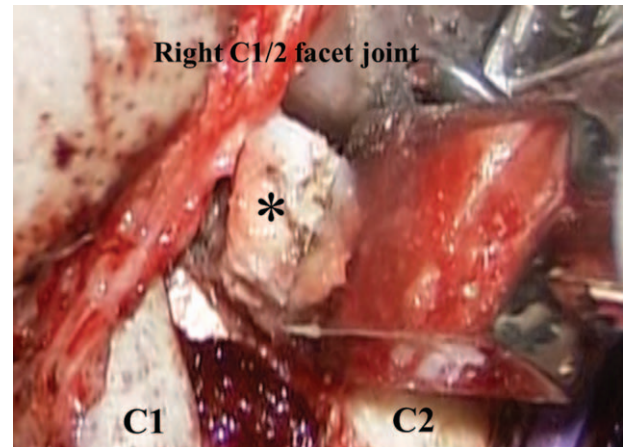


Figure 3. Intraoperative video-captured photographs showing the surgical steps. Bilateral C1/2 facet joints were widely exposed after transection of the C2 nerve roots at the ganglion on each side. An interbody spreader was then carefully inserted into the joint to make the joint much more open. Tricortical iliac bone (*) was pushed into the joints not only as grafting bone, but also as a fulcrum for leverage.

surgery-related neurological or vascular injuries as complications. Nine of the 10 patients needed to wear a Halo-vest after surgery, followed by cervical-shoulder bracing for at least 3 months after surgery. Cervical-shoulder bracing after surgery was selected for the remaining one patient because of severe respiratory dysfunction. Spinal external orthosis was continued until confirmation of spinal osseous stability. Bony fusion was confirmed in all cases. NCSS was improved in six cases and maintained in four cases, postoperatively. Surgical data are summarized in Table 3.

Radiological Outcomes

Radiological analysis included C1–2 height, inclination angle at C1 (C1-IA), C1-SAS, and ADI before surgery, at POD 1, POD 90, and the most recent postoperative follow-up. The average preoperative C1–2 height was 16.4 ± 4.2 mm, improving to 21.4 ± 4.6 mm at POD 1, 20.3 ± 4.5 mm at POD 90, 20.1 ± 4.6 mm at the recent follow-up. Mean preoperative C1-IA was $-22.6 \pm 10.5^\circ$, and significantly improved to $2.1 \pm 10.7^\circ$ on POD 1, 0.6 ± 11.3 mm at POD 90, 0.3 ± 12.1 mm at the recent follow-up. Mean preoperative C1-SAS was 9.4 ± 2.0 mm, improved significantly to 14.5 ± 2.2 mm on POD 1, 14.2 ± 2.7 mm at POD 90, 14.0 ± 2.8 mm at the recent follow-up. Besides, mean preoperative ADI was 10.3 ± 2.3 mm, significantly improved to 2.8 ± 2.2 mm on POD 1, 2.9 ± 2.1 mm at POD 90, 3.0 ± 2.3 mm at the recent follow-up. The Steel-Dwass multiple comparison revealed that these postoperative C1-IA, C1-SAS, and ADI were significantly improving compared with the preoperative parameters ($P < 0.05$). Radiological analysis was demonstrated in Figure 4. Mean C1/2 LJA was $-29.0 \pm 10.1^\circ$ preoperatively. Preoperative C1-IA correlated significantly

TABLE 2. Patient characteristics

| Case | Age/ Sex | Height (cm) /Body Weight (kg) | Genetic disorder | Odontoid dysplasia | Preoperative respiratory dysfunction (Tracheostomy) | Follow-up (months) |
|------|-------------|-------------------------------------|--|-----------------------|---|-----------------------|
| 1 | 12/M | 135/35 | Down syndrome | + | — | 57 |
| 2 | 6/F | 100/14 | — | + | + | 42 |
| 3 | 12/M | 117/16 | Chondrodysplasia punctate (CDPX1) | + | + | 41 |
| 4 | 7/M | 105/16 | — | + | — | 35 |
| 5 | 15/F | 133/40 | Down syndrome | + | — | 33 |
| 6 | 14/M | 156/56 | Down syndrome | + | — | 32 |
| 7 | 5/F | 108/16 | Down syndrome | + | — | 30 |
| 8 | 9/M | 131/36 | Chondrodysplasia punctate (Brachytelephalangic CDP) | + | — | 21 |
| 9 | 9/F | 116/26 | Chondrodysplasia punctate (CDPX1) | + | — | 11 |
| 10 | 8/M | 116/23 | Chondrodysplasia punctate (CDPX1) | + | — | 10 |

with preoperative C1/2 LJ-IA ($R^2=0.7043$) (Figure 5). Lower (negative) C1-IA correlated with lower (negative) C1/2 LJ-IA, suggesting significant dislocation of the C1/2 joint.

Illustrative Case

Case 8: A 9-year-old boy of CDP presented with bilateral progressive upper and lower motor weakness. His past medical history revealed that he was born at 38 weeks' gestation and suffered from respiratory distress and tetraparesis soon after his birth. He was intubated and treated with mechanical ventilation, but able to be extubated 2 months later. CDP was diagnosed based on typical clinical and radiological findings. He received conservative therapy such as cervical orthosis and rehabilitation. Although he could stand independently at 5 years old, he gradually demonstrated that he could not stand up himself. He was referred to our institute at 9 years old where severe spastic tetraparesis was confirmed on neurological examination. Imaging studies demonstrated an odontoid dysplasia and

rigid AAS, and severe compression of the spinal cord at the level of C1/2 (Figure 6A).

The patient underwent posterior C1–2 reduction and fusion. Initially, intraoperative lateral radiographs showed difficult reduction without bone grafting of bilateral atlantoaxial lateral joints (via compression of C1/2 laminae using Kocher forceps) which was attributed to slippage of C1 over C2. On the other hand, proper reduction with bone grafting was possible using the fulcrum lever technique. Spinal fusion was selected for both C1 lateral mass and C2 translaminal screws. Reduction and fusion were successfully performed. Follow-up neuroimaging at 8 months documented normal atlantoaxial alignment with successful decompression at C1–2 level (Figure 6B).

DISCUSSION

Surgical Challenges in Pediatric AAS

The surgical management of rigid AAS in pediatric patients is truly technically demanding. Transoral decompression as

TABLE 3. Surgical summary

| Case | Preoperative NCSS | | Postoperative NCSS | | Fusion level | Screws | | | | | Postoperative Halo-vest |
|------|-------------------|-------|--------------------|-------|--------------|----------|----------------|----------|----------|--------|-------------------------|
| | Upper | Lower | Upper | Lower | | C1 | C2 | C4 | C6 | Th1 | |
| 1 | 4 | 4 | 5 | 4 | C1-2 | LMS, LMS | Hook, Hook | — | — | — | + |
| 2 | 2 | 2 | 5 | 5 | C1-2 | LMS, LMS | Hook, TLS | — | — | — | + |
| 3 | 1 | 1 | 2 | 2 | C1-4 | LMS, LMS | TLS, TLS | LMS, TLS | — | — | — |
| 4 | 3 | 1 | 3 | 1 | C1-2 | LMS, LMS | TLS, TLS | — | — | — | + |
| 5 | 4 | 4 | 5 | 4 | C1-2 | LMS, LMS | TLS, TLS | — | — | — | + |
| 6 | 4 | 5 | 4 | 5 | C1-2 | LMS, LMS | TLS, TLS | — | — | — | + |
| 7 | 4 | 5 | 4 | 5 | C1-2 | LMS, LMS | Pars S, Pars S | — | — | — | + |
| 8 | 3 | 1 | 3 | 1 | C1-2 | LMS, LMS | TLS, TLS | — | — | — | + |
| 9 | 4 | 4 | 4 | 4 | C1-Th1 | LMS, LMS | TLS, TLS | TLS, TLS | TLS, TLS | PS, PS | + |
| 10 | 4 | 4 | 4 | 4 | C1-2 | LMS, LMS | TLS, TLS | — | — | — | + |

NCSS indicates Neurosurgical Cervical Spine Scale; LMS, lateral mass screw; TLS, translaminal screw; Pars S, pars screw; PS, pedicle screw.

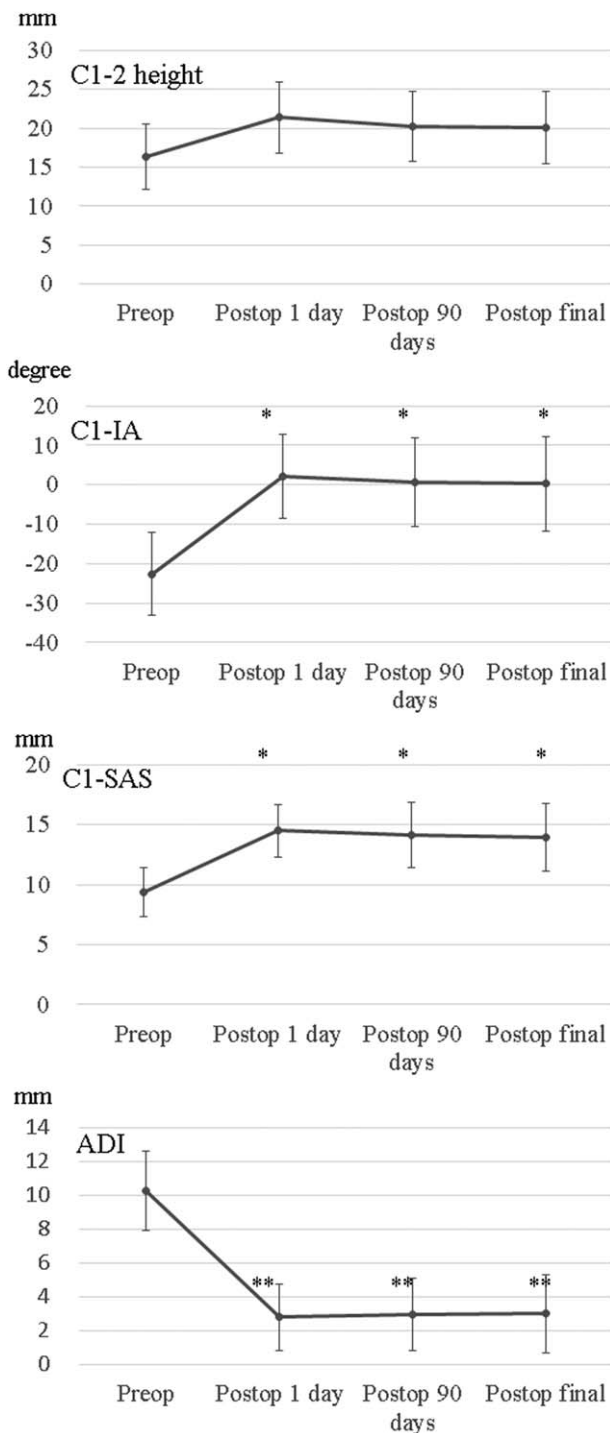


Figure 4. Summary of radiological analysis. **A**, The average preoperative C1–2 height was 16.4 mm, improving to 21.4 mm on POD 1, 20.3 mm on POD 90, and 20.1 mm on the recent follow-up. **B**, Mean preoperative C1-IA was -22.6° , and significantly improved to 2.1° on POD 1, 0.6° on POD 90, and 0.3° on the most recent follow-up. **C**, Mean preoperative C1-SAS was 9.4 mm, improved significantly to 14.5 mm on POD 1, 14.2 mm on POD 90, and 14.0 mm on the most recent follow-up. **D**, Mean preoperative ADI was 10.3 mm, significantly improved to 2.8 mm on POD 1, 2.9 mm on POD 90, and 3.0 mm on the most recent follow-up. The Steel-Dwass multiple comparison revealed that these postoperative C1-IA, C1-SAS, and ADI were significantly improving compared with the preoperative parameters (*, $P < 0.05$).

another option might be really limited in pediatric patients. Posterior surgical fusion of the upper cervical spine has been accomplished by on-lay or wiring techniques in conjunction with external rigid orthosis using Halo-vest orthosis. Biomechanical and clinical studies have shown that rigid screw fixation can provide better outcomes regarding stabilization and fusion rates compared with other techniques.^{6,19–28} On the other hand, surgical fixation of the upper cervical spine is challenging in pediatric patients due to the smaller bone size, relatively large head, increased flexibility, immature bone formation, and variable anatomy.^{24,25} Previous clinical reports have suggested that upper cervical spine instrumentation in pediatric patients may carry a high risk of surgery-related complications.^{24,29} Taking into consideration all daunting challenges and based on a thorough survey of the English literature, the principle of the index surgical procedure presented here involved three steps: (1) opening the C1/2 facet joints by distraction of the C1/2 facet joints; (2) placement of tricortical iliac bone as a spacer and fulcrum; and finally (3) compression between C1 and C2 laminae. The present study included short fusion without OC fusion. Bone grafting to both C1/2 facet joints and C1/2 laminae were added along with spinal instrumentation. Moreover, tricortical iliac bone was used as a joint spacer and grafting bone instead of a metal cage to avoid any possibility of spacer dislodgement and to allow removal of spinal instrumentations after bony fusion if needed late after surgery.

Specific Concerns and Study Limitations

Correction loss early after surgery or postoperative spinal deformity needs to be carefully considered. In our study correction loss occurred early after surgery in spite of Halo-vest orthosis. Halo-vest orthosis may be essential for pediatric patients. Another concern regarding resection of the C2 nerve root may be suggested. Routine bilateral C2 nerve root resection was conducted to provide wide exposure of the lateral C1–2 joints in all cases in the index study. C2 nerve root resection is contentious and may not be fully justified. Our previous study focusing on intentional resection of the C2 nerve root in adult cases, suggesting that C2 nerve root resection does not always result in significant morbidity and can be an option for surgical resolution to achieve safe, wide exposure of the lateral C1/2 joints.³⁰ We are aware that the small number of cases and relatively short follow-up period might be considered as a limitation for this study.

CONCLUSION

Surgery for rigid AAS in pediatric patients is still technically challenging because of the smaller immature spine. Although specific concerns and limitations were present in this technical study, such as the small number of cases and shorter follow-up period, posterior direct reduction of the lateral atlantoaxial joints for rigid pediatric AAS using a fulcrum lever technique can be considered as

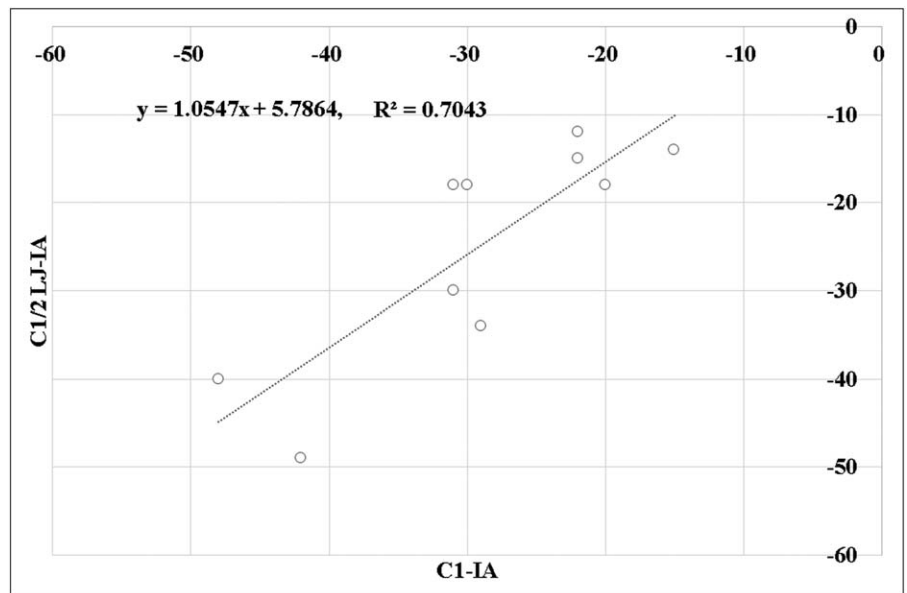


Figure 5. Correlation between preoperative C1-IA and preoperative C1/2 LJ-IA. Preoperative C1-IA correlated significantly with preoperative C1/2 LJ-IA ($R^2 = 0.7043$).

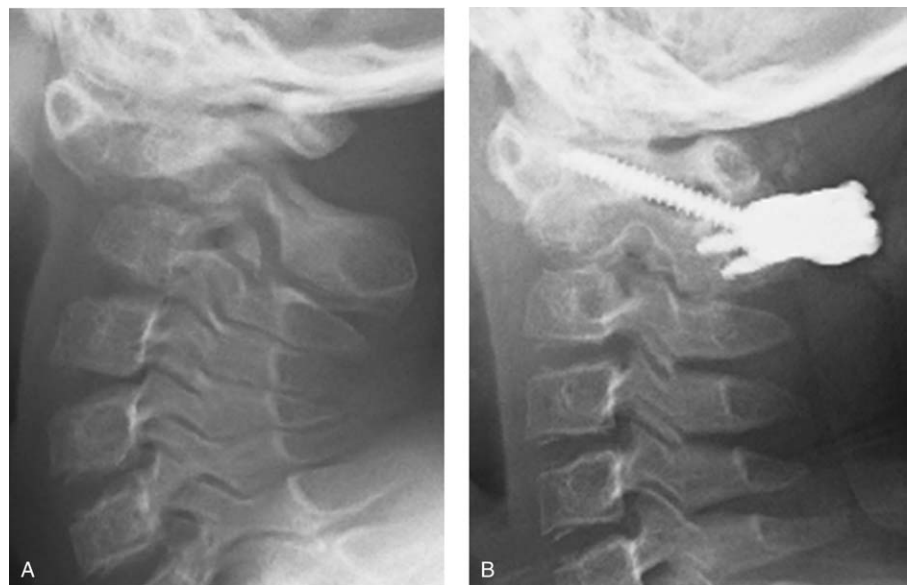


Figure 6. An illustrative case: case 8. Posterior direct reduction of C1/2 joints was performed. Cervical lateral radiography. **A**, Preoperative. **B**, Postoperative.

a surgical solution for this difficult pediatric spine deformity.

➤ Key Points

- ❑ The surgical treatment of pediatric rigid AAS is still technically challenging.
- ❑ The surgical procedure consisted of three steps: (1) opening and distraction of the C1/2 facet joints and placement of tricortical bone as a spacer and fulcrum; (2) placement of C1 and C2 screws; and finally, (3) compression between the C1 posterior arch and C2 lamina and constructing C1/2 fusion.

- ❑ Eight of 10 patients demonstrated genetic disorders, either Down syndrome or chondrodysplasia punctate. Bilateral C1 lateral mass screws were successfully placed in all cases.
- ❑ Radiological evaluation showed the corrections and bony fusions of C1/2 facet joint in all cases.
- ❑ The fulcrum lever technique for rigid pediatric AAS can be one of the effective surgical solutions to this challenging pediatric spinal disorder.

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