Radiographic Outcomes of Upper Cervical Fusion for Pediatric Patients Younger Than 10 Years

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Abstract:

Purpose: This study aimed to investigate radiographic outcomes after posterior spinal fusion (PSF) for pediatric patients younger than 10 years with upper cervical disorders. Methods: Thirteen patients (mean age at surgery, 5.9 years; range, 1 to 9 years) who underwent PSF with a minimum of 2 years of follow-up (mean, 5.8 years) were included. Diagnoses were atlanto-axial instability due to congenital disorders for 11 patients and atlanto-axial rotatory fixation for 2 patients. The fusion area was occipito-cervical for 7 patients and C1/2 for 6 patients. PSF was performed using rigid screw-rod constructs for 6 patients and conventional techniques for 7 patients. Ten patients required halo immobilization after surgery. Fusion status, perioperative complications, radiographic alignment, and range of motion (ROM) from C2 to C7 were evaluated. Results: Twelve patients successfully achieved bony fusion (fusion rate, 92%), but complications occurred in 5 patients. Regarding radiographic measures (preoperative/postoperative/final follow-up), the mean atlanto-dental interval was significantly reduced (8.0 mm/2.7 mm/3.5 mm) and the C2-7 ROM was increased (from 49.4 degrees to 66.0 degrees) at the final follow-up (both comparisons, $p<0.05$). Sagittal alignment was unchanged. Conclusion: Use of rigid screw-rod instrumentation in the upper cervical spine with careful radiological evaluation is amenable for pediatric patients younger than 10 years. However, conventional procedures such as wiring fixation with rigid external immobilization are still alternative options for preventing serious neurological and vascular complications.

Keywords:

pediatric patient, atlanto-axial instability, atlanto-axial rotatory fixation, posterior fusion, upper cervical spine

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Introduction

Upper cervical disorders, including atlanto-axial instability (AAI), atlanto-axial rotatory fixation (AARF), trauma, congenital anomaly, and others, are relatively uncommon diseases in the pediatric population. Spinal arthrodesis, especially with internal fixation, for pediatric patients is challenging given their smaller anatomical structures, immature bone quality, consideration for future growth potential, and difficulty in applying reliable external immobilization.

Traditionally, internal screw-rod instrumentation has not been technically feasible in young children; thus, various wiring techniques, including McGraw, Brook, and Gallie techniques with casting or halo-vest immobilization, were indicated to manage upper cervical disorders[1-3]. However, traditional fusion procedures (as mentioned) showed high nonunion rates, and the use of stable external immobilization for young children had a relatively high complication rate, thus adding to the difficulty during postoperative care[4-6]. Recently, rigid screw-rod instrumentation procedures, including transarticular, lateral mass, laminar, and pedicle screw techniques, as well as surgery-assisting devices, have contributed to remarkable advances[7-11] and have been applicable even for pediatric patients, despite the anatomical variation. However, using rigid screw-rod instrumentation has the potential risk of neurovascular complications and implant failures due to smaller or fragile bone structures, thus restricting its use in the pediatric population. Therefore, spi-

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Table 1. Demographic Data and Surgical Procedures.

No.	Age/Sex	Diag.	Underlying disease	BL (cm)/ BW (kg)	Procedure	Instrument	Bone graft	External Orthosis	Follow-up (month)
1	4/F	AAS	Down synd.	95/14	O-C2 fusion+ C1 laminectomy	Screw-rod	iliac bone	Halo jacket+ Total contact	108
2	3/F	AAS	Down synd.	82/10	O-C2 fusion+ C1 laminectomy	Screw-rod	iliac bone	Halo jacket+ Collar	60
3	5/F	AAS	Down synd.	104/17	O-C2 fusion	none	iliac bone	Halo jacket	24
4	8/M	AAS	Down synd.	113/17.5	O-C2 fusion+ C1 laminectomy	Screw-rod	iliac bone	Halo jacket+ Collar	84
5	4/F	AAS	NF-1[※]	72/10	O-C2 fusion	none	iliac bone	Halo jacket+ Total contact	158
6	9/F	AAS	CP[※※]+ Os odont.[†]	116/20.2	C1/2 fusion	transarticular +wiring (McGraw)	iliac bone	Halo jacket	108
7	5/M	AAS	Larsen synd.	98/14	C1/2 fusion	wiring (McGraw)	iliac bone	Total contact	24
8	1+10/F	AAS	Os odont.[†]	84/12.5	C1/2 fusion	none	iliac bone	Total contact	50
9	9/F	AAS	Os odont.[†]	125/29	C1/2 fusion	Screw-rod (C1LMS+C2PS)	iliac bone	Collar	60
10	9/F	AARF	none	141/52.6	C1/2 fusion+ Ant. release	wiring (Brooks)	iliac bone	Halo jacket+ Collar	84
11	7/M	AARF	none	124/25	C1/2 fusion+ Ant. release	wiring (McGraw)	iliac bone	Halo jacket+ Collar	84
12	6/M	AAS	SMD[††]	91/13	O-C2 fusion	Screw-rod	iliac bone	Halo jacket	36
13	1+6/M	AAS	CDP[$]	77/8	O-C3 fusion+ C1 laminectomy	none	iliac bone +rib strut	Halo jacket	24

※NF-1: neurofibromatosis-1, ※※CP: cerebral palsy, †Os odont.: Os odontoideum, ††SMD: spondylometaphyseal dysplasia, [$]CDP: chondrodysplasia punctata

nal arthrodesis in the skeletally immature cervical spine is still challenging and surgical outcomes are not thoroughly understood. The pediatric spine differs biomechanically from its adult counterpart, and maturation of the spine may be found radiographically as late as at 9 years of age[12]. Most pediatric patients 10 years and older can be surgically treated with the same strategy used for adult patients. In this study, we investigated surgical outcomes after posterior spinal fusion (PSF) for pediatric patients younger than 10 years with upper cervical disorders.

Methods

Internal review board approval was obtained from our hospital. Thirteen consecutive pediatric patients younger than 10 years with upper cervical disorders who underwent PSF at some point since 1995 at a single institution were included. There were 5 boys and 8 girls, with an average age of 5.9 years (range, 1-9 years). Diagnoses were AAI for 11 patients and chronic AARF lasting for more than 6 months for 2 patients. Of the 11 patients with AAI, underlying diseases were Down syndrome for 4 patients, os odontoideum for 2 patients, neurofibromatosis type-1 (NF-1) for 1 patient, spondylometaphyseal dysplasia for 1 patient, Larsen syndrome for 1 patient, chondrodysplasia punctate for 1 patient, and cerebral palsy for 1 patient. The average body length (BL) was 102 cm (range, 72-141 cm), average body weight (BW) was 18.7 kg (range, 8.0-52.6 kg), and average follow-up period was 5.8 years (range, 2-13 years). Demographic data and clinical features of the patients in this study are summarized in Table 1. We investigated fusion status, perioperative complications, and radiographic sagittal alignment. Fusion status was defined as a continuous trabecula on computed tomography (CT) and a range of motion (ROM) ≤3 degrees on flexion-extension lateral radiographs. Radiographic alignment included atlanto-dental interval (ADI), O-C2 angle (the angle between McGregor's line and the lower endplate of C2), C2-7 angle (the angle between the lower endplate of C2 and the lower endplate of C7) on lateral radiograph in the neutral position, and ROM in the subaxial cervical spine (C2-7) on the flexion-extension radiograph. A positive value on radiographic assessment indicated lordosis.

Surgical procedure and postoperative treatment

For AAI, occipito-cervical fusion (O-C fusion) was indicated for irreducible AAI and C1-2 fusion was indicated for reducible AAI based on the preoperative extension radiograph. Preoperative study using thin-slice CT angiography was routinely performed to confirm the osseous anomaly or anomalous course of the vertebral arteries since 2008[13]. The initial surgical procedures were O-C fusion for 7 patients and atlanto-axial fusion (C1-2 fusion) for 6 patients. Regarding O-C fusion, we used occipital plate C2 pedicle screws in 4 patients for O-C2 fusion; we used a simple monocortical iliac or rib onlay graft with morselized cancellous autografts in 3 patients for O-C2 fusion. All patients used halo immobilization after surgery, and the entire period of external orthosis, varying from cervical collar use to halo immobilization, was 7.1 months on average. Regarding C1-2 fusion, we

Table 2. Radiographic Parameters.

	Preop.	Postop.	Final	P value (preop.-final)
ADI※, mean (SD), mm	8.0 (2.2)	2.7 (2.9)	3.5 (3.2)	0.0018
O-C2 angle, mean (SD), °	8.8 (15.8)	19.3 (10.9)	19.1 (17.9)	0.7007
C2-7 angle, mean (SD), °	6.0 (11.3)	10.8 (17.3)	1.4 (34.7)	0.1328
C2-7 range of motion, mean (SD), °	49.4 (23.9)	-	66.0 (19.6)	0.0421

※ADI: atlanto-dental interval

used Brooks' sublaminar wiring techniques or modified McGraw methods for 3 patients; 2 of these 3 patients underwent preceding anterior release through the transoral approach for chronic AARF. We used C1 lateral mass-C2 pedicle screws in 1 patient, transarticular screws in 1 patient, and monocortical iliac bone grafting with morselized cancellous autografts alone in 1 patient. Three patients used halo immobilization after surgery, and the entire period of external orthosis, varying from cervical collar use to halo immobilization, was 4.0 months on average.

Statistical analysis

Statistical comparisons of the radiographic parameters before and after surgery were calculated using the nonparametric Wilcoxon signed-rank test. Statistical comparisons between the radiographic parameters were performed using the nonparametric Spearman's correlation coefficient by rank (r_s). A correlation coefficient with an absolute value of at least 0.5 and $p<0.05$ was considered statistically significant. StatView-J 5.0 (Abacus Concepts, Berkeley, CA, USA) was used for all statistical analyses.

Results

Fusion status

Twelve patients achieved solid bony union that was demonstrated on plain radiographs and CT images. Nonunion occurred in 1 patient with NF-1 (case 5), but additional surgery was not performed for 15 years postoperatively because there was no deterioration of AAI and clinical symptoms were absent. Moreover, unintended spontaneous fusion extending cephalad to the occipital bone in 1 patient and caudal to C3 and C4 in 3 patients and 1 patient, respectively, occurred.

Perioperative complications

Complications related to the surgery occurred in 5 patients, which included liquorrhea during C2 pedicle screw insertion (case 1), recurrent atlanto-axial subluxation immediately after initial surgery (case 3), deep wound infection requiring debridement and revised fusion surgery (case 13), skull penetration of the halo pin requiring revised halo ring application because of the thin and fragile skull cortex (case 13), and pin site infection (cases 5 and 6). There was neither neurological deterioration intraoperatively nor iatrogenic vascular injury.

Radiographic alignment

Radiographic parameter data are summarized in Table 2. ADI was significantly corrected from 8.0 mm preoperatively to 3.5 mm at final follow-up ($p<0.005$). The O-C2 angle was maintained at 8.8 degrees preoperatively to 19.1 degrees at final follow-up, and the C2-7 angle was maintained at 6.0 degrees to 1.4 degrees (both comparisons, $p>0.05$). Comparison between the O-C2 angle change and C2-7 change from the period to final follow-up showed a significant negative correlation between them ($r_s=-0.605$, $p<0.05$). The C2-7 ROM significantly increased from 49.4 degrees to 66.0 degrees ($p<0.05$).

Illustrative case

Case 2

A 3-year-old girl (BL, 82 cm; BW, 10 kg) had spastic gait disturbance due to irreducible AAI associated with Down syndrome (Fig. 1). She underwent C1 posterior arch resection and O-C fusion with a monocortical iliac bone graft using rigid screw-rod instrumentation. Bony fusion was successfully achieved without correction loss, and she could walk independently without myelopathic symptoms at 10-year follow-up.

Discussion

Consideration of anatomy in pediatric patients

For children younger than 3 years, the cervical spine is still undergoing dynamic changes; operative intervention during this time may hasten complications such as progressive cervical kyphosis, instability, and worsening cervical stenosis, all of which may necessitate additional surgical treatment[14]. The pediatric spine differs biomechanically from its adult counterpart, and maturation of the spine may be found radiographically as late as 9 years of age, which is one of the turning points of anatomical development[12]. Kanna et al conducted a morphologic evaluation of the cervical spine using CT in pediatric patients (mean age, 6.7 ± 3.9 years) and reported that more than 75% of adult pedicle widths and lengths were achieved by age 5 years; 90% of those were achieved by age 10 years[15]. Vara et al also conducted a cadaveric evaluation of cervical pedicle morphology in a pediatric population (mean age, 13 years; range, 3-

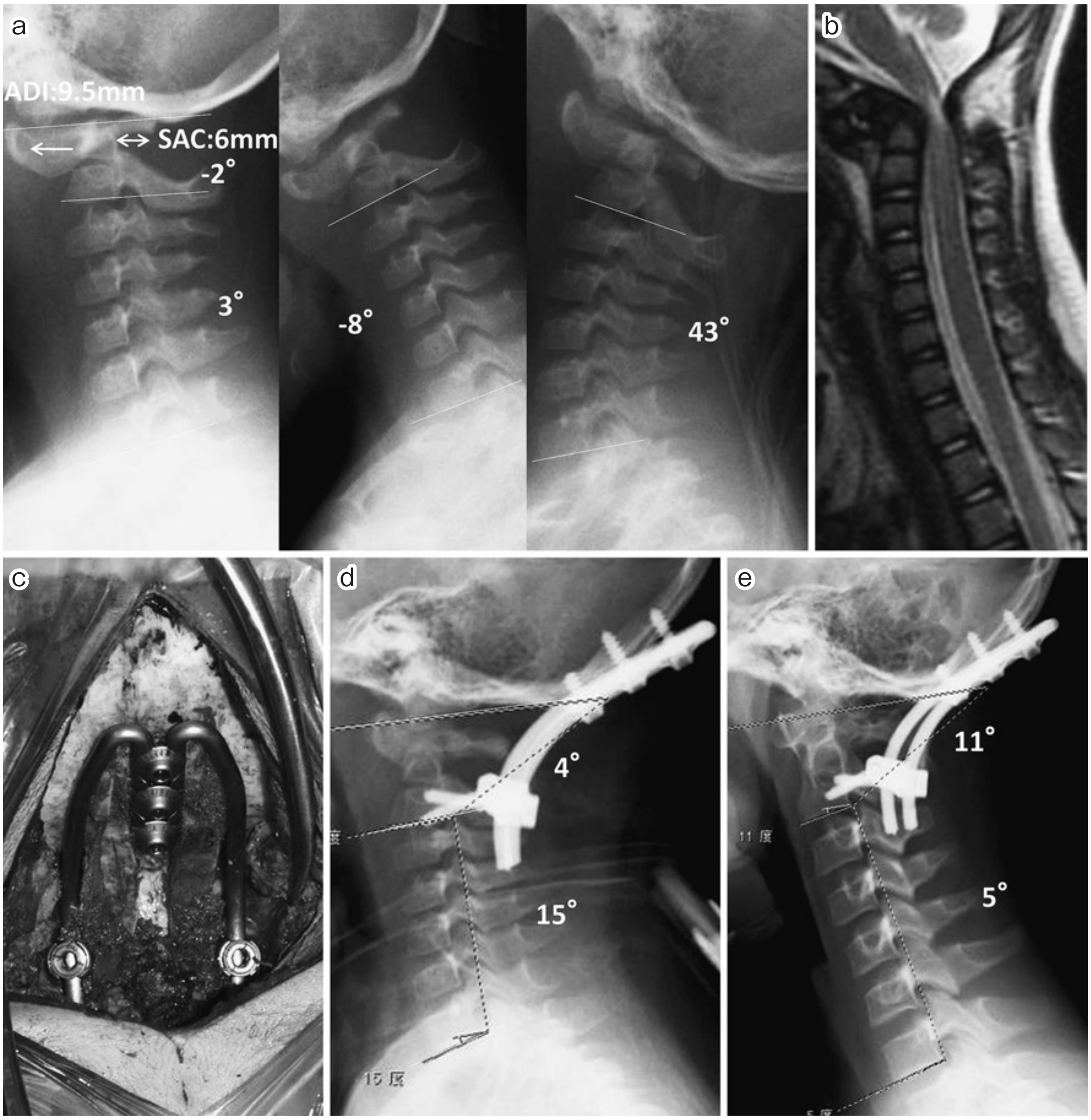

Figure 1. a: Preoperative plain radiographs demonstrating irreducible AAI with kyphosis at the cranio-cervical junction. b: T2-weighted sagittal MR image demonstrating severe spinal cord compression with intramedullary signal change. c: The patient underwent C1 posterior arch resection and occipito-cervical fusion with a monocortical iliac bone graft using rigid screw-rod instrumentation. d: Plain radiograph while wearing the halo jacket immediately after surgery demonstrating kyphosis correction. e: Plain radiograph demonstrating bony fusion without correction loss at 10-year follow-up.

18 years) and indicated that starting from age 6 years or older, the mean pedicle width is at least 3.4 mm (at C3 in the 6- to 8-year-old group)[16]. Regarding development of the cervical spinal canal, the interpedicular distance appeared to increase over time, the anteroposterior canal diameter remained constant during growth, and the percentage of adult growth achieved was more than 80% by age 5 years and 90% by age 10 years[16]. These findings suggest that the use of screw-rod instrumentation might be applicable to the cervical spine in pediatric patients younger than 10 years.

Fusion rates and complications related to fusion surgery in pediatric patients

In recent years, the use of modern screw constructs has provided high fusion rates, even for pediatric cervical spines; these improvements have been facilitated by refinements in spinal instrumentation and surgery-supporting technologies[5-11]. In the systematic review of instrumented fusion in the pediatric cervical spine[17], the use of the screw construct provided higher fusion (99%) and lower complication (14%) rates compared with the use of wiring constructs (fusion rate, 89%; complication rate, 52%) for both O-C fusions and cervical fusions not involving the occipito-cervical junction. The complications in the screw cohorts, including transient neurological deficit, cerebrospinal fluid leak, kyphotic deformity, infection, and nonunion, appeared to be less severe than those in the wire cohorts, which had complications including death, respiratory compromise, and quadriparesis. However, a few studies reported iatrogenic vertebral artery injury or dural venous sinus injury by applying cervical or occipital screw constructs to the smaller and thinner bone structure[18-20], which might result in mortality. Regarding vertebral artery variations, only 1 patient with bilateral vertebral arteries entering the transverse foramen at the C4 level was observed in this series. Because a high frequency of anomalous vertebral arteries in the V3 segment associated with atlanto-axial instability and congenital or acquired conditions (e.g., congenital deformity, Down syndrome) was reported[21,22], careful preoperative study using thin-slice CT angiography is recommended to confirm sufficient bone structure and the existence of the anomalous vertebral artery or osseous anomaly[13], especially before screw-rod instrumented fusion. Unintended fusion in an adjacent segment has been reported after O-C fusion in children in studies involving more than 7 years of follow-up[23]. Development of unintended fusion at adjacent levels was seen in 5 patients in the present study. Meticulous exposure should be performed so that the adjacent uninvolved levels are not exposed, which could eliminate the unintended fusion.

Regarding cervical alignment, a previous study demonstrated that most young children undergoing upper cervical fusion continue to have good alignment and continued growth within the fused levels after surgery[24]. In the present study, O-C2 and C2-7 angles were maintained and C2-7 ROM was surprisingly increased at final follow-up, which might be a result of the specific population of syndromic patients with ligamentous laxity. Although spontaneous realignment and remodeling of the subaxial kyphosis after upper cervical fusion are expected in growing patients[25,26], we have followed the principle of the adult population of not fusing kyphotic alignment in the preoperative neutral position[27]. This principle could be adapted to the pediatric population to prevent dysphagia and dyspnea from airway obstruction after O-C fusion[28]. Although osseous fusion is critical, the instrumentation selection must also meet the need for biomechanical stability while minimizing surgical morbidity[29].

External immobilization in pediatric patients

Although use of the rigid screw-rod system has become widespread, we believe that the use of rigid external immobilization including the halo jacket is critical, especially for O-C fusions and for younger patients (younger than 3.0 years) with difficulty in applying screw-rod instrumentation. However, the use of external immobilization for younger children entailed significant morbidity and still does[30,31]. Limpaphayom et al reported 68 pediatric patients (mean age, 10 years; range, 1-20 years) who were treated with a halo to correct spinal deformity followed by immobilization and who sustained a high complication rate (53%), including pin-site complications and neurologic complications related to halo traction[31]. Regarding pin-site complications, 13 patients with infections were successfully treated with oral antibiotics or removal of the pin and 2 patients encountered skull penetration with no sequelae. In this series, there were 3 halo pin configurations, 4 pins with 6 pounds per square inch (lb/inch^2) of torque in 31 patients, 6 pins with 5 lb/inch^2 of torque in 27 patients, and 8 pins with 3 to 4 lb/inch^2 of torque in 11 patients. In the present study, 1 patient experienced skull penetration with no sequelae, which might have otherwise caused dural laceration, cerebrospinal fluid leakage, and subdural or epidural hematoma formation. Hence, our pin configurations were applied using 4 to 10 pins with torque less than 1 lb/inch^2 to 4 lb/inch^2, and we recommended checking the distance between the halo ring and the skin of all pins every day if the patient was admitted. Although alternatives to the halo jacket are the use of a pinless halo jacket or a total contact brace to prevent pin-site complications, there is a possibility of skin-related and pressure-related complications, such as pressure sores, due to the device; therefore, more careful and frequent examinations of the pressure points of the halo and jacket are necessary.

This study has some limitations. First, because of the small number of cases recruited from a single institution over a long duration, there were various causes of AAI and the surgical procedures including use of instrumentation were not uniform. Second, this study lacks powerful statistical analysis of the cervical alignment.

Conclusion

With careful preoperative evaluations to check the bone structure and the vertebral artery, using rigid screw-rod instrumentation in the upper cervical spine is amenable for pediatric patients younger than 10 years. However, due to the lack of long-term follow-up data, concern still exists regarding the effects of rigid instrumentation; conventional procedures including a simple monocortical onlay bone graft and sublaminar wiring or tapping fixation are still useful options, especially for younger children, to prevent serious neurological or vascular complications. Although surgical instrumentation systems as well as surgery-assisting technology have allowed for remarkable advances, the use of rigid external immobilization such as a halo jacket has an important role in enhancing rapid osseous union and reducing the load on bone-implant interfaces.

Conflict of Interest: The authors declare that there are no conflicts of interest.

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