Pediatric occipitocervical fusion: long-term radiographic changes in curvature, growth, and alignment

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OBJECTIVE The authors assessed the rate of vertebral growth, curvature, and alignment for multilevel constructs in the cervical spine after occipitocervical fixation (OCF) in pediatric patients and compared these results with those in published reports of growth in normal children.

METHODS The authors assessed cervical spine radiographs and CT images of 18 patients who underwent occipitocervical arthrodesis. Measurements were made using postoperative and follow-up images available for 16 patients to determine cervical alignment (cervical spine alignment [CSA], C1–7 sagittal vertical axis [SVA], and C2–7 SVA) and curvature (cervical spine curvature [CSC] and C2–7 lordosis angle). Seventeen patients had postoperative and follow-up images available with which to measure vertebral body height (VBH), vertebral body width (VBW), and vertical growth percentage (VG%—that is, percentage change from postoperative to follow-up). Results for cervical spine growth were compared with normal parameters of 456 patients previously reported on in 2 studies.

RESULTS Ten patients were girls and 8 were boys; their mean age was 6.7 ± 3.2 years. Constructs spanned occiput (Oc)–C2 ($n = 2$), Oc–C3 ($n = 7$), and Oc–C4 ($n = 9$). The mean duration of follow-up was 44.4 months (range 24–101 months). Comparison of postoperative to follow-up measures showed that the mean CSA increased by 1.8 ± 2.9 mm ($p < 0.01$); the mean C2–7 SVA and C1–7 SVA increased by 2.3 mm and 2.7 mm, respectively ($p = 0.3$); the mean CSC changed by -8.7° ($p < 0.01$) and the mean C2–7 lordosis angle changed by 2.6° ($p = 0.5$); and the cumulative mean VG% of the instrumented levels (C2–4) provided 51.5% of the total cervical growth (C2–7). The annual vertical growth rate was 4.4 mm/year. The VBW growth from C2–4 ranged from 13.9% to 16.6% ($p < 0.001$). The VBW of C-2 in instrumented patients appeared to be of a smaller diameter than that of normal patients, especially among those aged 5 to < 10 years and 10–15 years, with an increased diameter at the immediately inferior vertebral bodies compensating for the decreased width. No cervical deformation, malalignment, or detrimental clinical status was evident in any patient.

CONCLUSIONS The craniovertebral junction and the upper cervical spine continue to present normal growth, curvature, and alignment parameters in children with OCF constructs spanning a distance as long as Oc–C4.

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KEY WORDS alignment; arthrodesis; curvature; fixation; growth; occipitocervical; pediatric; radiographic; spine

CURVATURE, range of motion, and stability of the cervical spine are conferred by the bony structures, ligaments, discs, and muscles.^{10,20,23} After occipitocervical fixation (OCF), a child's cervical spine is believed to experience changes in growth and curvature that might be clinically relevant for the patient, but evidence from large series is scarce.^{3,4,7,11,12,14,19,24,30}

Single-level vertical growth after OCF has been reported previously,³ with the C-2 vertebra accounting for approximately 38% of the total growth of the cervical

spine in healthy children.^{3,29} However, the rate of growth of vertebrae within multilevel constructs has not yet been described, to the best of our knowledge.

In this study, we analyzed measurements taken from radiographs obtained in pediatric patients (age range 7 months to 12 years at time of surgery) following OCF. Careful evaluations of vertebral body growth and curvature and alignment in the cervical spine were performed on the basis of images taken immediately postoperatively and at the last follow-up (range 12–89 months).

ABBREVIATIONS AD = anterior displacement; CSA = cervical spine alignment; CSC = cervical spine curvature; Oc = occiput; OCF = occipitocervical fixation; PD = posterior displacement; SVA = sagittal vertical axis; VBH = vertebral body height; VBW = vertebral body width; VG% = vertical growth percentage.

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Methods

This study was approved by the Institutional Review Board of St. Joseph's Hospital and Medical Center, Phoenix, Arizona.

A retrospective review was performed of cervical spine anteroposterior and lateral plain radiographs and midsagittal CT images obtained in 40 pediatric patients who underwent OCF between 2004 and 2013. Images were obtained from the clinical database of a single surgeon (N.T.).

While adhering to policies regarding patient anonymity, we searched for the name and medical record number of each patient in the Dominator diagnostic reading station (DR Systems, Inc.) and made measurements on the resulting images using built-in features of the software. Inclusion criteria included the availability of at least 1 good-quality, neutral, preoperative, cervical spine lateral radiograph with clear visualization from C-2 to C-7, or a midsagittal CT study with visualization from the tip of the dens to C-7. Images were compared with the same type of image taken immediately after surgery and at the last follow-up. All cervical radiographs were acquired with the patient in the upright position, to better reproduce normal load distribution, curvature, and alignment along the spine. The minimum duration of follow-up required for inclusion was 12 months. Of 40 patients, 18 met the inclusion criteria and 22 were excluded because they did not. Twelve of the 22 excluded patients had undergone surgery within 12 months of this analysis, 7 had their first postoperative imaging performed after 8 weeks of follow-up,

and 3 had imaging that either did not include the tip of C-2 or did not include C-7 for measurement. None of the 22 excluded patients was lost to follow-up before 24 months.

All included patients (n = 18) were assigned to 2 analytical cohorts; patients with appropriate data for the analyses were included in both cohorts. Cohort 1 (n = 16) included patients with cervical alignment and curvature measurements, and Cohort 2 (n = 18) included patients with vertebral height, width, and growth measurements. Patients with inadequate images were excluded from cohort analyses.

Measurements

All measurements were made on images obtained immediately postoperatively and during follow-up examinations. All measurement comparisons were made between the last follow-up for each patient (Table 1) and the immediate postoperative imaging. Cervical spine alignment (CSA) was classified on the basis of the anterior displacement or the posterior displacement of the vertebral bodies on lateral radiographs or sagittal CT images from a line extending between the posterior borders of C-2 and C-7. CSA was further classified into lordotic (anterior displacement > 2 mm), straight (anterior or posterior displacement < 2 mm), kyphotic (posterior displacement ≥ 2 mm), or swan neck (anterior/posterior displacement ≥ 2 mm).^{3,28} The cervical sagittal vertical axis (SVA) was used to assess sagittal plane translation during follow-up by measuring the distance between a plumb line drawn from the anterior border of the tubercle of C-1 (C1–7 SVA)

TABLE 1. Summary of radiographic and clinical data obtained in pediatric patients after OCF*

Case No.	Age (yrs), Sex	Indication	FU (mos)	Instrumented Levels	Alignment		Curvature (°)		VG%
					PO	Difference (mm)†	CSC Difference†	C2–7 Lordosis Difference†	
1	0.58, F	Congenital	24	Oc–C2	NA	NA	NA	NA	37.0
2	2, F	Traumatic	31	Oc–C3	S	S, 0.4	–13	3	43.5
3	3, M	Congenital	32	Oc–C4	S	L, 2.1	–19	–17	73.9
4	4, F	Congenital	24	Oc–C2	L	L, 3.5	–26	–15	NA
5	4, M	Congenital	61	Oc–C4	L	L, –0.5	–8	–3	64.2
6	4, M	Congenital	36	Oc–C3	L	L, 1.4	–5	–2	30.9
7	5, F	Congenital	30	Oc–C4	L	L, –1.1	–7	–25	52.4
8	5, M	Traumatic	50	Oc–C3	L	L, –0.6	3	27	51.7
9	5, M	Traumatic	52	Oc–C4	K	L, 9.8	–15	12	52.0
10	7, F	Traumatic	48	Oc–C4	L	L, –0.5	2	4	27.3
11	8, F	Traumatic	55	Oc–C3	K	S, 4.6	–11	8	62.9
12	9, F	Congenital	36	Oc–C3	L	L, 3.2	–25	10	13.2
13	10, M	Traumatic	84	Oc–C4	Sw	L, 0.8	5	33	54.8
14	10, F	Traumatic	40	Oc–C4	K	L, 1.6	–19	–7	76.1
15	10, M	Congenital	33	Oc–C3	L	L, –1.3	8	3	50.0
16	11, F	Congenital	33	Oc–C4	L	L, 0.3	–7	–1	50.8
17	11, F	Congenital	101	Oc–C4	NA	NA	NA	NA	77.1
18	12, M	Traumatic	30	Oc–C3	K	L, 5.4	–4	13	57.3
Mean	6.7		44.4			1.8	–8.8	2.7	51.5

CSC = cervical spine curvature; FU = follow-up; K = kyphosis; L = lordosis; NA = not available; PO = postoperative; S = straight; Sw = swan neck.

* Lordotic angles are reported as negative values. Follow-up measurements were obtained at the last office visit of each patient.

† Determined by subtracting the postoperative value from the follow-up value.

and from the C-2 centroid (C2–7 SVA) to the posterior superior corner of C-7 (Fig. 1).

Cervical spine curvature (CSC) on neutral lateral radiographs was assessed using the Jackson physiological stress lines method (Fig. 2).^{2,15,22} The angle formed by the intersection of one line drawn tangentially to the posterior border of C-2 and a second line drawn tangentially to the posterior border of C-7 was measured. Furthermore, C2–7 lordosis^{13,27} was measured using sagittal Cobb angles at the intersection of 2 perpendicular lines, with each perpendicular line originating from a line drawn parallel to the inferior endplates of C-2 and C-7. Lordotic angles were given a negative value.

Vertebral body height (VBH) was measured on neutral lateral radiographs with a line extending from the upper to the lower border of the endplate of each vertebral body at the midline, and vertebral body width (VBW) was measured by a line drawn perpendicular to the midpoint of each VBH line, from the posterior to the anterior border of the anterior column (Fig. 3).²⁹ Furthermore, all measurements for VBH and VBW were compared with Wang and colleagues' previously published normative values of height and width of the cervical spine, which were based on data obtained in 96 children.²⁹

Vertical growth percentage (VG%), defined as the percentage of growth provided by the instrumented levels to the cervical spine, was calculated using the following formula: $[\text{construct growth/spinal growth}] \times 100$.³ The construct growth was defined as the change in VBH spanning the instrumented level(s) (excluding C-1) from postoper-

FIG. 2. Lateral cervical radiograph with screw-rod construct extending from the occiput to C-3. CSC is measured by determining the intersecting angle of a line drawn tangentially to the posterior border of C-2 with a line drawn tangentially to the posterior border of C-7.

FIG. 1. Sagittal plane translation is measured by determining the distance between cervical SVAs on lateral radiographs, which is assessed by drawing plumb lines from the anterior tubercle of C-1 (C1–7 cervical alignment [SVA], arrowhead) and from the axis centroid (C2–7 SVA, arrow) to the posterior superior corner of C-7.

FIG. 3. Lateral cervical radiograph with an Oc–C3 screw-rod construct. Dashed lines show VBH from C-2 to C-7, extending from the superior to the inferior border of each vertebral endplate. Perpendicular solid lines drawn from the midpoint of each VBH line show VBW from C-2 to C-7, extending from the anterior to the posterior borders of the anterior column.

ative to follow-up measurements, and spinal growth was defined as the change in spinal height from C-2 to C-7 from postoperative to follow-up measurements.

Statistical Analysis

Data were analyzed using paired Student t-tests (follow-up measurements vs immediately postoperative measurements). The level of significance was set at $p = 0.05$.

Results

Demographics and Indications for OCF

Of the 18 patients included in the study, 10 (55.6%) were girls and 8 (44.4%) were boys. Constructs spanned Oc–C2 in 2 patients (11%), Oc–C3 in 7 patients (39%), and Oc–C4 in 9 patients (50%). The mean age at surgery was 6.7 ± 3.2 years (range 7 months–12 years). The last available follow-up comparisons were performed in June 2014 (mean 32.4 ± 20.1 months; range 12–89 months); as patients continued to have follow-up and imaging studies, results were reviewed again and updated in May 2015, with a final mean follow-up of 44.7 ± 20.7 months (range 24–101 months). No changes between June 2014 and May 2015 were noted. Indications for arthrodesis were trauma in 8 patients (44%) and multiple congenital abnormalities in 10 (56%).

Data were gathered on the age and sex of patients, follow-up, indication for OCF, instrumented levels, analysis of alignment (C2–7 SVA and CSA [follow-up minus postoperative]), curvature (CSC and C2–7 lordosis [follow-up minus postoperative]), and vertical growth. Results are summarized in Table 1.

Cervical Alignment and Curvature

Cohort 1

On the basis of postoperative CSA measurements in 16 patients, 9 patients (56.3%) were classified as having a lordotic neck, 3 (18.7%) as having a straight neck, 3 (18.7%) as having a kyphotic neck, and 1 (6.2%) as having a swan neck. At the last follow-up for these 16 patients, 14 patients (87.5%) had a lordotic neck, 2 (12.5%) had a straight neck, and none had a kyphotic or swan neck. The mean postoperative CSA was 2.0 mm (range –3.6 to 4.6 mm), and the mean CSA at the last follow-up was 3.8 mm (range 1.3–7.3 mm), representing a statistically significant increase ($p < 0.01$) of 1.8 ± 2.9 mm. None of the 5 patients with a lordotic increase in CSA of more than 2 mm (Table 1) experienced any clinical or neurological deficits.

The mean sagittal alignment using C2–7 SVA was 13.8 ± 8 mm (range –1.4 to 26.6 mm) postoperatively and 16.2 ± 9.0 mm (range –3.5 to 30.2 mm) at last follow-up. The mean sagittal alignment using C1–7 SVA was 25.1 ± 9.8 mm (range 9.8–39.9 mm) postoperatively and 27.8 ± 10.2 mm (range 8.3–43.3 mm) at last follow-up. The differences between follow-up and postoperative measurements for both SVA calculations were not statistically significant (2.4 and 2.7 mm, respectively; $p = 0.3$).

CSC assessment showed a statistically significant increase in lordotic curvature of -8.8° ($p < 0.01$), with a postoperative mean of $-17^\circ \pm 13.7^\circ$ (range -46° to 0°) and

a last follow-up mean of $-25.8^\circ \pm 14.3^\circ$ (range, -4° to 53°). The mean C2–7 lordosis was $-14.7^\circ \pm 12.6^\circ$ (range, -39° to 6°) postoperatively and $-12.1^\circ \pm 20.1^\circ$ (range, -45° to 18°) at last follow-up ($p = 0.5$).

Cervical Spine Growth

Cohort 2

Table 2 summarizes VBH and VBW postoperatively and at last follow-up. For both instrumented and noninstrumented levels (i.e., C2–7), the cumulative VBH increased by a total of 13.3 mm ($p < 0.001$) from postoperative to last follow-up, with a mean single-level growth of 4.3 mm (32.2%) at C-2; 2.0 mm (15.2%) at C-3; 1.9 mm (14.2%) at C-4; 1.7 mm (12.7%) at C-5; 1.7 mm (12.4%) at C-6; and 1.8 mm (13.3%) at C-7. The cumulative mean growth of the instrumented levels (C2–3 and C3–4) provided 51.5% (VG%, range 13.2–77.1%) of the total cervical growth (C2–7).

Similarly, the VBW for C2–7 increased significantly ($p < 0.001$), with a mean growth of 1.5 mm (13.9%) at C-2, 1.8 mm (16.6%) at C-3, and 1.8 mm (16.2%) at C-4. Width growth from C-5 to C-7 ranged from 1.7 mm (16.0%) to 2.0 mm (18.7%) at each level.

Comparison of Growth in Normal Children and OCF Patients

Seventeen patients with OCF had radiographs acquired consistently between 1.5 and 2 years postoperatively and had their operative age adjusted to age at the time of follow-up (0 to < 5 years, 5 to < 10 years, and 10 to 15 years). The mean age was 8.5 ± 3.5 years (range 1.6–13 years).

TABLE 2. Growth in vertebral body height and width of patients following OCF*

Level	Mean PO, mm ($\pm$ SD)†	Mean FU, mm ($\pm$ SD)‡	Growth, mm (%)
VB height			
C-2	21.9 ($\pm$ 5.9)	26.2 ($\pm$ 6.8)	4.3 (32.2)
C-3	6.8 ($\pm$ 1.6)	8.8 ($\pm$ 2.4)	2.0 (15.2)
C-4	6.8 ($\pm$ 1.5)	8.7 ($\pm$ 2.2)	1.9 (14.2)
C-5	6.7 ($\pm$ 1.6)	8.4 ($\pm$ 2.2)	1.7 (12.7)
C-6	6.9 ($\pm$ 1.6)	8.6 ($\pm$ 2.0)	1.7 (12.4)
C-7	8.0 ($\pm$ 2.0)	9.8 ($\pm$ 2.5)	1.8 (13.3)
Total	57.3 ($\pm$ 14.1)	70.6 ($\pm$ 18.2)	13.3 (100)
VB width			
C-2	10.0 ($\pm$ 2.3)	11.4 ($\pm$ 1.4)	1.5 (13.9)
C-3	11.2 ($\pm$ 2.4)	12.9 ($\pm$ 1.7)	1.8 (16.6)
C-4	11.5 ($\pm$ 2.3)	13.2 ($\pm$ 1.6)	1.8 (16.2)
C-5	11.5 ($\pm$ 2.3)	13.5 ($\pm$ 2.0)	2.0 (18.7)
C-6	12.1 ($\pm$ 2.5)	13.7 ($\pm$ 1.6)	1.7 (16.0)
C-7	12.4 ($\pm$ 2.9)	14.5 ($\pm$ 2.0)	2.0 (18.7)
Total	68.9 ($\pm$ 14.7)	79.2 ($\pm$ 15.1)	10.3 (100)

VB = vertebral body.

* The differences between all reported values are statistically significant ($p < 0.01$).

† The mean patient age postoperatively was 86.1 months.

‡ The mean patient age at follow-up was 112.1 months.

TABLE 3. Comparison of vertebral body height between normal²⁹ and OCF patients in 3 age cohorts at last follow-up*

Level	VB Height					
	0 to <5 Yrs		5 to <10 Yrs		10 to 15 Yrs	
	Normal	OCF	Normal	OCF	Normal	OCF
C-2	18.86 ± 5.26	17.50 ± 5.3	27.51 ± 3.18	26.8 ± 4.8	33.97 ± 3.61	28.8 ± 7.6
C-3	5.68 ± 1.18	5.60 ± 0.6	7.96 ± 1.15	9.3 ± 2.1	11.54 ± 2.57	8.8 ± 1.8
C-4	5.56 ± 1.21	5.90 ± 1.0	7.89 ± 1.20	9.3 ± 1.8	11.13 ± 2.28	8.0 ± 1.5
C-5	5.61 ± 1.16	5.40 ± 1.1	7.61 ± 1.05	9.2 ± 1.6	10.65 ± 2.13	8.0 ± 1.4
C2-5	35.70 ± 8.80	34.40 ± 8.0	50.97 ± 6.56	54.6 ± 10.3	50.97 ± 10.58	53.6 ± 12.3

* Values are presented as the mean ± SD in millimeters. Normal sample size = 96 patients; OCF sample size = 15 patients. The mean age for OCF patients from 0 to < 5 years was 3.1 ± 1.3 years (range 1.6–4.7 years), from 5 to < 10 years was 7.2 ± 1.3 years (range 5–8.3 years), and from 10 to 15 years was 11.7 ± 1.1 years (range 10–13 years). The mean follow-up duration was 44.4 ± 20.7 months.

Two patients were older than 15 years at follow-up (16 and 18 years), and their growth was not compared with growth in normal children. The mean follow-up duration was 32.4 ± 20.1 months.

Comparisons were made with the results seen in 96 pediatric patients with normal VBH and VBW from C-2 to C-7 that were published by Wang et al.²⁹ The mean values and standard deviations of both sexes were calculated and presented for the same 3 age groups. Tables 3 and 4 and Figs. 4 and 5 summarize the comparisons of VBH and VBW, respectively, between patients undergoing OCF and normal patients.

An analysis of the cumulative annual growth (mm/year) in VBH and the average growth in VBW of the C3–7 vertebral bodies was performed using measurements obtained in 15 patients from our series after age adjustment. Results were compared with results reported by Kasai et al. for 360 normal pediatric cases.¹⁶ Table 5 summarizes this comparison.

Discussion

Concerns Following Occipitocervical Arthrodesis in Children

Long-term follow-up assessment of growth, curvature, alignment, and stability of the cervical spine after OCF in children is an unavoidable subject in most pediatric series. The clinical relevance of the potential effects of arthrodesis in the pediatric spine has been discussed extensively,^{3–7,9,11,19,21–26} but only a few studies have meticulously evaluated growth, curvature, and alignment^{3,22,25} at

the craniovertebral junction and the upper cervical spine in young patients in whom short constructs have been placed. So far, no study has reported significant changes in radiographic or clinical findings after a long follow-up period.

Anderson et al.,³ in an analysis of 17 children who underwent Oc–C2 fusion (mean follow-up 28 months, mean age at fusion 4.7 years), found an increase in lordosis of –12° and a mean vertical growth of 37% within the fusion segment, and considered both within acceptable parameters. No clinical detriment was noted in any patient. However, this issue has not been addressed in detail in patients in whom longer constructs have been placed (Oc–C3 and Oc–C4). Our series of 17 patients with constructs ranging from Oc–C2 to Oc–C4, with an average ongoing clinical and radiographic follow-up of 44.4 months, showed similar results. None of our patients had radiographic or clinical deterioration.

Cervical Curvature and Alignment

The purpose of this analysis was to assess cervical alignment and curvature changes at long-term follow-up. Surgical reduction of the cervical spine to reestablish normal curvature is always demanding, and the surgeon should consider the changes in curvature and alignment that normally take place in the developing spine after instrumentation. For any type of cervical arthrodesis, the main objective is to make the constructs as neutral and physiological as possible. Ongoing radiographic control can be achieved using the C-2 plumb line (C-2 SVA), CSA,

TABLE 4. Comparison of vertebral body width between normal²⁹ and OCF patients in 3 age cohorts at last follow-up*

Level	VB Width					
	0 to <5 Yrs		5 to <10 Yrs		10 to 15 Yrs	
	Normal	OCF	Normal	OCF	Normal	OCF
C-2	10.1 ± 0.65	8.7 ± 3.1	27.51 ± 0.83	11.48 ± 1.4	31.88 ± 0.86	11.8 ± 1.6
C-3	5.68 ± 0.64	9.3 ± 2.3	7.96 ± 0.88	13.6 ± 2.3	11.54 ± 0.96	13.6 ± 2.7
C-4	5.56 ± 0.66	9.1 ± 2.5	7.89 ± 1.05	13.7 ± 2.4	11.13 ± 1.13	14.2 ± 1.8
C-5	5.61 ± 0.70	9.4 ± 3.4	7.61 ± 1.10	13.9 ± 1.7	10.65 ± 1.13	14.4 ± 1.3
C2-5	26.94 ± 2.64	36.5 ± 11.3	50.97 ± 3.85	52.68 ± 7.8	65.19 ± 4.08	54.0 ± 7.4

* Values are presented as the mean ± SD in millimeters. Normal sample size = 96 patients; OCF sample size = 15 patients. The mean age for OCF patients from 0 to < 5 years was 3.1 ± 1.3 years (range 1.6–4.7 years), from 5 to < 10 years was 7.2 ± 1.3 years (range 5–8.3 years), and from 10 to 15 years was 11.7 ± 1.1 years (range 10–13 years). The mean follow-up duration was 44.4 ± 20.7 months.

FIG. 4. Comparison of VBH changes between the 96 normal patients reported by Wang et al.²⁹ and our 15 patients who underwent occipitocervical OCF at last follow-up by age group ([A] 0 to < 5 years; [B] 5 to < 10 years; and [C] 10 to 15 years).

posterior C2–7 angle (CSC), and C2–7 lordosis (Cobb) angle.^{1–3,27}

Our institution treats a considerable number of patients with complex spinal pathologies, and some patients included in this series presented preoperatively with severe cervical abnormalities from trauma and/or congenital deformities. Previous pediatric series have considered changes greater than 2 mm in lordotic alignment and more severe than -11° in curvature to be abnormal at final follow-up,²¹ whereas others have disregarded the actual clinical significance of these findings in the overall condition of patients.³ An SVA greater than 40–50 mm after placement of cervical instrumentation in adults indicates sagittal imbalance and is related to decreased quality-of-life scores and unfavorable neck disability indexes. After comparing our results with pediatric, adolescent, and adult parameters in healthy and fusion-treated patients,^{1,2,27} we found that, at long-term follow-up, the changes in curvature measured by C1–7 and C2–7 SVA and C2–7 Cobb angle were statistically insignificant and comparable with the physiological parameters reported in adult and pediatric populations.^{2,27}

Available data on cervical curvature show that it is not constant in children,¹ and while some authors have encountered predominant lordotic angles ($-4.8^\circ \pm 12^\circ$ and $-6.5^\circ \pm 11.7^\circ$),^{1,16,18} others have encountered neutral (< 11 years = $-6.5^\circ \pm 11.7^\circ$; ≥ 11 years = $-0.7^\circ \pm 13.8^\circ$)¹ and even kyphotic values and have catalogued them as normal.³¹ Therefore, we believe that comparing the curvature angles of our

patients with those of normal children would be unfruitful. However, the statistically significant lordotic increases of 1.8 mm in CSA and -8.7° in CSC in our patients are below both the previously mentioned 2.0 mm in CSA and -11° in CSC.^{3,17} The crankshaft phenomenon, as described by Dubousset et al. in 1989,⁸ is an increase in lordosis and rotation in young children with scoliosis treated by posterior thoracolumbar instrumentation prior to skeletal maturity.⁸ Other studies have defined hyperlordosis as an angulation $> 40^\circ$ in the cervical spine at last follow-up. Although 2 patients in the present series had a curvature exceeding this value at last follow-up, their initial curvature was $> 30^\circ$, and the change at last follow-up was $< 11^\circ$. None of our patients developed an evident crankshaft phenomenon or clinical deterioration or pain due to this change within a mean follow-up period of 44.4 months, which is comparable to findings in other studies with similar long-term follow-up.

Cervical Growth

A comparison of VBH, VBW, and growth was made between measurements obtained in our OCF-treated patients and the values from normal cervical spine radiographs of 96 healthy children reported by Wang et al.²⁹ and 360 healthy children reported by Kasai et al.¹⁶ When we compared our VBH and VBW values to those reported by Wang et al.,²⁹ we found similar VBH (C2–5) compared with the values reported in healthy children, without any

FIG. 5. Comparison of VBW changes between the 96 normal patients reported on by Wang et al.²⁹ and our 15 patients who underwent OCF at last follow-up by age group ([A] 0 to < 5 years; [B] 5 to < 10 years; and [C] 10 to 15 years).

difference in mean $\pm$ standard deviation (Fig. 4) for age groups 0 to < 5 years, 5 to < 10 years, and 10 to 15 years. We found an annual vertical growth rate of 4.4 mm/year, which not only is increased compared with the normal mean values (2.6 mm/year) reported by Wang et al.,²⁹ but also is similar to the rate they reported for children during growth spurts (4.4 mm/year).

In 2006, Anderson et al.³ reported that C-2 (in Oc-C2 constructs) provided 34% of the overall vertical growth of the cervical spine, which is similar to what was estimated as normal (38%) in the results reported by Wang et al.²⁹ The only patient in our series with an Oc-C2 construct is the only patient who did show a vertical growth of 37% at C2. However, the overall growth for all patients at C-2 was 32.2%. Patients with Oc-C3 and Oc-C4 constructs

had vertical growth provided by the instrumented levels of 44.2% and 58.6%, respectively.

Although no difference was evident between height and vertical growth, we noticed differences in the VBW of normal patients compared with the VBW in OCF-treated patients at last follow-up. In the majority of cervical levels (instrumented and noninstrumented), C-2 appeared to have a smaller diameter, especially evident in the age groups of 5 to < 10 years and 10 to 15 years, whereas the C2–5 levels of OCF patients had a significant increase in width compared with that of normal patients (Fig. 5). We hypothesize that the decreased weight bearing at C-2 might originate a deceleration in its width growth, with compensatory acceleration in width growth at the immediately inferior vertebral bodies.

TABLE 5. Comparison of cumulative annual vertebral body height growth and mean vertebral body width by sex between 360 normal pediatric patients¹⁶ and 15 OCF patients*

Patient Sex	Cumulative Annual VB Height Growth (mm/yr)†		Mean VB Width Growth (mm/yr)†	
	Normal	OCF	Normal	OCF
Male	2.8	5.7	0.52	1.3
Female	2.4	3.1	0.45	0.9
Total	2.6	4.4	0.5	1.1

* The mean age for OCF patients from 0 to < 5 years was 3.1 ± 1.3 years (range 1.6–4.7 years), from 5 to < 10 years was 7.2 ± 1.3 years (range 5–8.3 years), and from 10 to 15 years was 11.7 ± 1.1 years (range 10–13 years). The mean follow-up duration was 44.4 ± 20.7 months.
† Measured at C3–7.

Our results suggest that the vertical growth that occurs in fusion-treated patients is comparable to the growth observed in healthy children. Vertebral remodeling within and around the fused mass, as previously proposed by Anderson et al.,³ allows the spine to continue its normal development despite the presence of fixation instrumentation and arthrodesis. We did not encounter any cervical deformation or malalignment related to OCF or arthrodesis, or any detrimental clinical signs or symptoms in any patient. Surprisingly enough, even patients with connective tissue disease or chromosomal deletions with characteristic shorter-than-average height for age (Morquio syndrome, Down syndrome, Wolf-Hirschhorn syndrome, and spondyloepiphyseal dysplasia) continued to demonstrate statistically significant growth at the instrumented segments as well as VBH and VBW that were comparable to those of normal patients.

Limitations

This is a retrospective study with robust follow-up from a single surgeon in a tertiary care neurosurgical referral center. Results presented in this paper may not be reproducible in different clinical scenarios or for patients in whom longer constructs are placed.

Conclusions

The craniovertebral junction and the upper cervical spine continue to exhibit normal growth, curvature, and alignment parameters in children with constructs as long as Oc–C4. Although some patients experienced radiographic changes above the established normal parameters, no patient had neurological or physical deterioration compared with baseline, and none were in need of any revision for primary fixation failure or for adjacent-level disease related to their primary surgery. OCF in children remains a safe procedure, even in very young children with demonstrated long-term stability. Following a cohort of patients through adulthood will be important to assess the potential for long-term complications.

References

- Abelin-Genevois K, Idjerouidene A, Roussouly P, Vital JM, Garin C: Cervical spine alignment in the pediatric population: a radiographic normative study of 150 asymptomatic patients. *Eur Spine J* 23:1442–1448, 2014
- Ames CP, Blondel B, Scheer JK, Schwab FJ, Le Huec JC, Massicotte EM, et al: Cervical radiographical alignment: comprehensive assessment techniques and potential importance in cervical myelopathy. *Spine (Phila Pa 1976)* 38 (22 Suppl 1):S149–S160, 2013
- Anderson RC, Kan P, Gluf WM, Brockmeyer DL: Long-term maintenance of cervical alignment after occipitocervical and atlantoaxial screw fixation in young children. *J Neurosurg* 105 (1 Suppl):55–61, 2006
- Anderson RC, Ragel BT, Mocco J, Bohman LE, Brockmeyer DL: Selection of a rigid internal fixation construct for stabilization at the craniovertebral junction in pediatric patients. *J Neurosurg* 107 (1 Suppl):36–42, 2007
- Astur N, Klimo P Jr, Sawyer JR, Kelly DM, Muhlbauer MS, Warner WC Jr: Traumatic atlanto-occipital dislocation in children: evaluation, treatment, and outcomes. *J Bone Joint Surg Am* 95 (24 e194):e194, 1–8, 2013
- Brockmeyer DL, Brockmeyer MM, Bragg T: Atlantal hemirings and craniocervical instability: identification, clinical characteristics, and management. *J Neurosurg Pediatr* 8:357–362, 2011
- Couture D, Avery N, Brockmeyer DL: Occipitocervical instrumentation in the pediatric population using a custom loop construct: initial results and long-term follow-up experience. *J Neurosurg Pediatr* 5:285–291, 2010
- Dubousset J, Herring JA, Shuffeberger H: The crankshaft phenomenon. *J Pediatr Orthop* 9:541–550, 1989
- Fargen KM, Anderson RC, Harter DH, Angevine PD, Coon VC, Brockmeyer DL, et al: Occipitocervicothoracic stabilization in pediatric patients. *J Neurosurg Pediatr* 8:57–62, 2011
- Garrido BJ, Puschak TJ, Anderson PA, Sasso RC: Occipitocervical fusion using contoured rods and medial offset connectors: description of a new technique. *Orthopedics* 32:1–16, 2009
- Gluf WM, Brockmeyer DL: Atlantoaxial transarticular screw fixation: a review of surgical indications, fusion rate, complications, and lessons learned in 67 pediatric patients. *J Neurosurg Spine* 2:164–169, 2005
- Hankinson TC, Avellino AM, Harter D, Jea A, Lew S, Pincus D, et al: Equivalence of fusion rates after rigid internal fixation of the occiput to C-2 with or without C-1 instrumentation. *J Neurosurg Pediatr* 5:380–384, 2010
- Harrison DE, Harrison DD, Cailliet R, Troyanovich SJ, Janik TJ, Holland B: Cobb method or Harrison posterior tangent method: which to choose for lateral cervical radiographic analysis. *Spine (Phila Pa 1976)* 25:2072–2078, 2000
- Hwang SW, Gressot LV, Chern JJ, Relyea K, Jea A: Complications of occipital screw placement for occipitocervical fusion in children. *J Neurosurg Pediatr* 9:586–593, 2012
- Jackson R: The cervical syndrome. *Clin Orthop* 5:138–148, 1955
- Kasai T, Ikata T, Katoh S, Miyake R, Tsubo M: Growth of the cervical spine with special reference to its lordosis and mobility. *Spine (Phila Pa 1976)* 21:2067–2073, 1996
- Kennedy BC, D'Amico RS, Youngerman BE, McDowell MM, Hooten KG, Couture D, et al: Long-term growth and alignment after occipitocervical and atlantoaxial fusion with rigid internal fixation in young children. *J Neurosurg Pediatr* 17:94–102, 2016
- Lee CS, Noh H, Lee DH, Hwang CJ, Kim H, Cho SK: Analysis of sagittal spinal alignment in 181 asymptomatic children. *J Spinal Disord Tech* 25:E259–E263, 2012
- Menezes AH: Craniocervical fusions in children. *J Neurosurg Pediatr* 9:573–585, 2012
- Menezes AH, Traynelis VC: Anatomy and biomechanics of normal craniovertebral junction (a) and biomechanics of stabilization (b). *Childs Nerv Syst* 24:1091–1100, 2008
- Moorthy RK, Rajshekhar V: Changes in cervical spine curvature in pediatric patients following occipitocervical fusion. *Childs Nerv Syst* 25:961–967, 2009
- Nakagawa T, Yone K, Sakou T, Yanase M: Occipitocervical fusion with C1 laminectomy in children. *Spine (Phila Pa 1976)* 22:1209–1214, 1997
- Oppenlander ME, Clark JC, Sonntag VK, Theodore N: Pediatric craniovertebral junction trauma. *Adv Tech Stand Neurosurg* 40:333–353, 2014
- Oppenlander ME, Kalyvas J, Sonntag VK, Theodore N: Technical advances in pediatric craniovertebral junction surgery. *Adv Tech Stand Neurosurg* 40:201–213, 2014
- Parisini P, Di Silvestre M, Greggi T, Bianchi G: C1–C2 posterior fusion in growing patients: long-term follow-up. *Spine (Phila Pa 1976)* 28:566–572, 2003
- Rodgers WB, Coran DL, Kharrazi FD, Hall JE, Emans JB: Increasing lordosis of the occipitocervical junction after arthrodesis in young children: the occipitocervical crankshaft phenomenon. *J Pediatr Orthop* 17:762–765, 1997

27. Tang JA, Scheer JK, Smith JS, Deviren V, Bess S, Hart RA, et al: The impact of standing regional cervical sagittal alignment on outcomes in posterior cervical fusion surgery. **Neurosurgery** **71**:662–669, 2012
28. Toyama Y, Matsumoto M, Chiba K, Asazuma T, Suzuki N, Fujimura Y, et al: Realignment of postoperative cervical kyphosis in children by vertebral remodeling. **Spine (Phila Pa 1976)** **19**:2565–2570, 1994
29. Wang JC, Nuccion SL, Feighan JE, Cohen B, Dorey FJ, Scoles PV: Growth and development of the pediatric cervical spine documented radiographically. **J Bone Joint Surg Am** **83-A**:1212–1218, 2001
30. Yamazaki M, Akazawa T, Koda M, Okawa A: Surgical simulation of instrumented posterior occipitocervical fusion in a child with congenital skeletal anomaly: case report. **Spine (Phila Pa 1976)** **31**:E590–E594, 2006
31. Yukawa Y, Kato F, Suda K, Yamagata M, Ueta T: Age-related changes in osseous anatomy, alignment, and range of motion of the cervical spine. Part I: Radiographic data from over 1,200 asymptomatic subjects. **Eur Spine J** **21**:1492–1498, 2012

Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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Conception and design: Theodore, Martinez-del-Campo. Acquisition of data: Martinez-del-Campo. Analysis and interpretation of data: Martinez-del-Campo, Turner. Drafting the article: Martinez-del-Campo. Critically revising the article: Theodore, Martinez-del-Campo, Soriano-Baron, Newcomb, Kalb. Reviewed submitted version of manuscript: Theodore. Approved the final version of the manuscript on behalf of all authors: Theodore. Statistical analysis: Martinez-del-Campo. Study supervision: Theodore.

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