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Clinical outcomes of the surgical treatment of isolated unilateral facet fractures, subluxations, and dislocations in the pediatric cervical spine: report of eight cases and review of the literature

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Abstract

Purpose We present a small series consisting of eight children with unilateral facet injury of the cervical spine treated surgically.

Methods A retrospective review was performed. Injury data, radiographs, surgical data, and outcomes (Neck Disability Index (NDI), Short Form 36 (SF-36), and Visual Analog Scale for Neck Pain (VAS-NP)) were collected from seven patients. A literature review was performed for one additional case.

Results Motor vehicle accidents (62 %, $n=5$) and falls (38 %, $n=3$) accounted for all injuries. The C6–7 level accounted for most of the injuries (37.5 $\%$, $n=3$). The mean NDI score with at least 3 months follow-up was 5.3 ($n=6$, range, 1–12; standard deviation, 4.5), corresponding to mild disability. Of the norm-based SF-36 scale scores available $(n=6)$, the mean physical functioning (PF), role-physical (RP), and roleemotional (RE) scores were significantly less than the adult, age 18–24, norm-based means, with a mean difference of −6.4, −9.13, and −11.3, respectively (p value=0.03, 0.001, and 0.01, respectively). The mean general health (GH) and vitality (VT) scores, however, were significantly greater than the adult, age 18–24, norm-based mean, with a mean difference of 7.82 and 10.3 ($p=0.04$ and 0.02, respectively). VAS-NP showed a return to the "no pain" level at 3 months or more follow-up in all patients.

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Conclusions We suggest that surgical treatment of these injuries in the pediatric age group may lead to satisfactory clinical and radiographic outcomes, but HRQoL analysis suggests that patients remain physically and emotionally disabled to some degree after surgery.

Keywords Pediatric spine . Cervical spine . Facet injury . Spinal instrumentation . Health-related quality of life

Introduction

Unilateral cervical facet fractures, subluxations, and dislocations in adults generate considerable controversy regarding diagnosis [1–6]. The timing and manner of reduction [7–11] and the optimal surgical approach and technique are also debated if surgical treatment is pursued [2–4, 7, 12–22].

Unilateral cervical facet injuries typically involve translation of the vertebral body, up to 25 % of the anteroposterior diameter of the subjacent vertebral body [14, 23–26], and some degree of rotation, as the injured facet and lateral mass complex rotates along an axis centered on the intact contralateral facet [27]. Standardized measurement techniques to define vertebral translation, cervical kyphosis, and rotational displacement have been described (Fig. 1).

Cervical facet injury in the pediatric age group has rarely been reported in the literature, and management and treatment paradigms for children are even less clear [28–30]. To our knowledge, only one previous case report [28] described the surgical treatment of a unilateral facet injury in the cervical spine of a 22-month-old girl sustained in a motor vehicle accident.

We contribute seven more cases, collected from two institutions, of surgical treatment of children with isolated unilateral facet injury of the cervical spine after trauma and describe their health-related quality of life (HRQoL) outcomes.

Fig. 1 a The anterior displacement is calculated as the percentage of the inferior endplate diameter $(X/Y \times 100)$ where X (blue line) is the distance along the superior endplate between lines tangential to the posterior aspects of the adjacent vertebral bodies (red lines) and Y (yellow line) is the AP diameter of the inferior endplate. Measurements were taken at point of maximum AP translation. b Cervical kyphosis was measured by

the method of Cobb. Axial rotation was measured as the difference between two angles. One is the angle between the true perpendicular line and a line bisecting (c) the superiormost vertebral body. The second is the angle between the true perpendicular line and a line bisecting (d) the inferior vertebral body

HRQoL outcomes are becoming a staple of adult spine surgery outcomes assessment, but have yet to be adopted consistently in the pediatric spine population. The specific outcomes of interest were pain and disability at a minimum of 3 months postsurgery, as measured by the Neck Disability Index (NDI), Short Form 36 (SF-36), and Visual Analog Scale for Neck Pain (VAS-NP). We sought to determine if children with unilateral facet fractures, dislocations, and subluxations returned to normal general health status after surgical intervention.

Materials and methods

We retrospectively reviewed data on seven consecutive patients who underwent surgical treatment for a unilateral facet fracture, subluxation, or dislocation between C2 and T1, inclusive, at Texas Children's Hospital (TCH) and Riley Children's Hospital (RCH) between September 2007 and March 2013. Patient demographics, clinical history, indications for treatment, and operative data were recorded for these patients.

We collected and analyzed plain radiographs and CT scans at the time of injury as well as at the most recent follow-up, when available. The plain radiographs and CT scans at the time of injury were used to classify the injury as fracture, subluxation, dislocation, or some combination of the three and to measure the degree and pattern of subluxation or dislocation.

All four patients treated at TCH underwent a posterior instrumented fusion utilizing lateral mass screws $(n=3)$ or a combined C2 pars/lateral mass screw construct $(n=1)$ via a standard posterior midline approach. One patient also underwent interspinous wiring with braided titanium cables to supplement the fusion. All three patients at RCH underwent anterior cervical discectomy and fusion with one of the three also undergoing a posterior instrumented fusion at the same level. The patient described in the literature by Chen et al. underwent a posterior instrumented fusion using lateral mass miniplates [28].

Outcomes collected at follow-up included the NDI, SF-36, and VAS-NP. Patients completed the questionnaires alone or with parental assistance.

The NDI is a ten-item questionnaire that measures a patient's self-reported neck pain-related disability. Questions include activities of daily living such as personal care, lifting, reading, working, driving, sleeping, and recreational activities; pain intensity; concentration; and headache. Each question was measured on a scale from 0 (no disability) to 5, and an overall score (percentage) out of 100 was calculated by adding each item score and then multiplying the sum by 2. The overall score of 0–8 % represents no disability; 10–28 %, mild disability; 30–48 %, moderate disability; 50–68 %, severe disability; and >68 %, complete disability.

The SF-36 was also collected and is a well-known generic health status measure. It consists of 36 questions related to eight health concepts. These concepts include physical functioning, physical role, bodily pain, general health, vitality, social functioning, emotional role, and mental health. Adult population norms for each of the key concepts (scales) are available for comparison. There are no published pediatric normative values. Thus, for analysis, we chose to compare our data with norm-based scale scores from the adult population, ages 18–24, intending to compare our pediatric cohort to a normal population that approached their phenotype as much as possible.

A VAS-NP is a measurement instrument that measures pain, ranging across a continuum from none to extreme pain. This outcomes tool consists of a horizontal line, 100 mm in length, anchored by word descriptors at each end. The patient marks a point on the line that he feels represents his current perception of pain. The VAS score is determined by measuring in millimeters from the left end of the line to the point that the patient marks.

The Child Health Questionnaire (CHQ)—an 87-question exam which was developed for children and has available population norms—was felt to be impractical for use in the clinic setting, where visits typically last only 10–15 min.

Statistical analysis

Clinical, operative, and radiographic parameters were collected. Frequency distributions and summary statistics were calculated for these data. The mean of norm-based SF-36 scales from our series of patients was compared with their respective normative adult population mean using an unpaired two-sided t-test. There are no published pediatric normative values.

Literature review

We performed a PubMed search using the key words and phrases "pediatric," "pediatric spine," "pediatric cervical spine," and "cervical facet injury," and "cervical facet fracture," "cervical facet subluxation," and "cervical facet dislocation." Inclusion criteria for our study consisted of articles from peer-reviewed journals that described an isolated unilateral facet injury of the cervical spine in pediatric patients (<18 years of age). Three published reports [28–30] met our inclusion criteria; however, only one report [28] described surgical treatment of the facet injury, and the other two articles [29, 30] were subsequently excluded. Data collected included patient age, patient gender, etiology of cervical instability, pattern of cervical spine injury, procedure performed, levels fused, construct materials, follow-up duration, presence of a bony fusion, and complication type and frequency.

Results

Demographics

There are a total of eight patients in the aggregate series (Table 1), including four patients from our own institutional review, one from the review of the literature, and three from our collaborating institution, of which one was male and seven were female. The mean age at presentation was 12.4 years old (range, 1.8–18 years).

Clinical and operative data

Motor vehicle accidents were responsible for five (62.5 %) of the eight injuries, and falls accounted for three (37.5 %) injuries. The C2–3 level accounted for 12.5 % (one patient); C3–4, 25.0 % (two patients); C4–5, 0.0 % (zero patient); C5–

6, 25.0 % (two patients); C6–7, 37.5 % (three patients); and C7–T1, 0.0 % (zero patients). The mean length of follow-up was 20.2 months (range, 7.5–51 months) (Table 1).

Two cases were associated with a neurological deficit. Five patients had a facet fracture; four patients had facet subluxation; and one patient had facet dislocation (Fig. 2). The mean AP translation was 21 % $(n=7, \text{ range}, 0-45)$; axial rotation, 5.3° $(n=7, \text{ range}, 0-26)$; and local kyphosis, 7.1° $(n=7, \text{ range})$ range, −8–35°) (Table 1).

The patients had a mean of 1.4 levels fused $(n=8, \text{range}, 1-$ 2 levels). Of the six patients who had postoperative CT cervical imaging available, five had achieved Lenke fusion grade B, and one had achieved Lenke fusion grade C at 3 months early follow-up post-op (Table 1).

Complications

One patient developed anterior soft tissue swelling which resolved spontaneously. No other complications were noted.

HRQoL outcomes

HRQoL measures were available for six of seven of our patients on last follow-up. In general, surveys were handed out on initial follow-up visits, but some patients were lost to follow-up or significantly delayed in follow-up, and the process was not standardized.

The mean NDI score was 5.3 ($n=6$; range, 1-12; standard deviation, 4.5). Three patients reported no disability (NDI, 0– 8 %); three patients, mild disability (NDI, 10–28 %); no patients, moderate disability (NDI, 30–48 %); no patients, severe disability (NDI, 50–68 %); and no patients, complete disability (NDI>68 %) (Table 2). The mean VAS-NP was 0 $(n=6; \text{range}, 0=0; \text{standard deviation}, 0)$ (Table 2).

The mean norm-based SF-36 scale scores for those available (6/7 patients) was 47.0 for PF ($n=6$; range, 21.5–57.1; standard deviation, 13.2), 43.5 for role-physical $(n=5; \text{range})$, 28–56.2; standard deviation, 14.5), 51.7 for bodily pain (BP) $(n=6; \text{ range}, 37.5-62.7; \text{ standard deviation}, 10.6)$, 57.3 for general health (GH) ($n=6$; range, 53.2–64.0; standard deviation, 5.2), 57.3 for vitality (VT) $(n=5; \text{ range}, 45.1-68.0;$ standard deviation, 9.7), 49.0 for SF ($n=6$; range, 35.4–57.1; standard deviation, 8.2), 38.5 for role-emotional (RE) $(n=5;$ range, 34.3–55.3; standard deviation, 9.4), and 50.1 for mental health (MH) $(n=6; \text{ range}, 27.7-64.1; \text{ standard deviation},$ 12.6) (Table 2).

Of the norm-based SF-36 scale scores available $(n=6)$, the mean PF, role-physical (RP), and RE scores were significantly less than the adult, age 18–24, norm-based means, with a mean difference of -6.4 , -9.13 , and -11.3 , respectively (p value=0.03, 0.001, and 0.01, respectively). The mean GH and VT scores, however, were significantly greater than the adult,

Table 1 Clinical and operative data Table 1 Clinical and operative data

Fig. 2 a 3-D reconstructed CT of the cervical spine shows a left C2–3 facet subluxation (red arrow). **b** and **c** CT of the cervical spine with sagittal reconstructions demonstrate a left C3 facet fracture with

age 18–24, norm-based mean, with a mean difference of 7.82 and 10.3 ($p=0.04$ and 0.02, respectively) (Table 3).

The correlation coefficients between NDI and SF-36 physical component score (PCS) and mental component score (MCS)—two summary scores representing physical and mental dysfunction—were 0.1 and −0.9, respectively. The poor or inverse correlation accurately represents the different scoring between the scales, with higher SF-36 scores representing higher function and higher NDI scores representing higher dysfunction. While the MCS score was highly correlated, the PCS representing physical dysfunction did not correlate strongly with NDI scores, highlighting the mixed nature of postoperative HRQoL outcomes in our patient cohort. It was not possible to calculate correlation between VAS and SF-36 or NDI components as respondents uniformly reported 0/10 pain (Table 2).

Discussion

Unilateral facet injury is relatively common in adults and represents approximately 6 % of all cervical spine injuries [31]. When treating unilateral facet injuries in adult patients, most spine surgeons will suggest an operation, such as anterior cervical discectomy and fusion or posterior instrumented fusion, to avoid delayed instability and prolonged rigid external immobilization [32, 33]. However, there has been limited experience in children and adolescents [28–30]. The treatment approach in the pediatric age group is complicated; due to the immature skeleton and its potential for further growth, fusion is considered less desirable. However, because sophisticated biomechanical and clinical studies have demonstrated persistent instability resulting from unilateral cervical facet injuries in adults, and because recurrent dislocation or progression to dislocation has been observed in conjunction with

associated C3 on C4 anterolisthesis (C3 anterolisthesis from normal alignment over subadjacent vertebra emphasized via a red line)

nonoperative treatment including orthosis in the adult population [32, 34–36], we chose to pursue aggressive operative treatment in our pediatric cohort rather than closed reduction and prolonged use of rigid external immobilization.

Furthermore, the pediatric spine has unique biomechanical properties, distinct from the adult cervical spine, which also argues in favor of aggressive surgical stabilization. The facet joints are more horizontal in a child; the more horizontally oriented facet joints allow more motion in the sagittal plane, which increases the incidence of neurological and ligamentous injury without bony fracture [37]. Even in the absence of trauma, C2–3 and C3–4 pseudosubluxation is a common variant among young children [38]. Furthermore, uncovertebral joints are not yet developed in younger children, therefore allowing increased motion in the coronal plane and a predisposition to similar ligamentous injury. Finally, the head of a child is disproportionately large, which results in an axis of rotation centered at C2–3 rather than C5–6, as is the case in an adult. This high axis of rotation, combined with poor head control and general ligamentous laxity, results in a higher incidence of upper cervical spine injuries and a lower incidence of middle and lower cervical spine injuries in the pediatric age group. As ligamentous structures are the dominant stabilizers in the upper cervical spine, and high cervical injuries in children resulting from exaggerated sagittal and coronal plane motion are likely to involve ligamentous injury, pediatric cervical spine injuries are more likely to produce unstable injuries that require instrumented fusion.

There is only one published case [28] of surgical treatment of a unilateral facet injury at the C5–6 level in a 22-month-old girl with associated spinal cord injury after a motor vehicle accident. The girl was initially immobilized in a Philadelphia collar, and at 1 month after injury, closed reduction of a unilateral facet dislocation was attempted but failed. The child was taken to the operating room for a posterior approach for

open reduction and lateral mass fusion with titanium miniplates. At 1 year follow-up, the patient was reported to be doing well. There are two other pediatric cases published in the literature where a 10-month-old boy [30] and 9-year-old boy [29] were treated nonoperatively with closed reduction and Minerva jacket and rigid cervical collar, respectively. Both patients were reported to be doing well at last followup at 18 months and 2 years, respectively.

The most common injury in our series was unilateral facet fracture (five patients), followed by unilateral facet subluxation (four patients), then unilateral facet dislocation (one patient). Some patients' injuries involved a combination of fracture/subluxation or fracture/dislocation. The majority of injuries occurred at the C6 –7 level (three patients, ages 16, 18, and 18). The advanced age of these patients, with the resultant proportional head size and improved head control, may explain the low axis of rotation in these injuries. Five injuries (62.5 %) occurred as a result of a motor vehicle accident, and three injuries (37.5 %) resulted from falls. All patients from TCH were managed with posterior instrumented fusion. Patients at RCH underwent anterior $(n=2)$ or combined anteriorposterior fusion $(n=1)$. Meaningful comparisons are hard to draw between anterior, posterior, and combined operative techniques, given the small sample size.

To the best of our knowledge, this review is the first comprehensive series to report on reliable and valid HRQoL outcomes on a cohort of children with unilateral facet injuries, specifically those managed with surgery. The goals of treatment for cervical spine injuries are to preserve neurological function, to preserve spinal stability, to prevent postinjury pain or delayed neurological deterioration, and to return the patient to preinjury health status [7]. Unfortunately, the traumatic mechanism of these injuries made preinjury HRQoL assessment impossible. Furthermore, there are no normative values of an otherwise healthy North American pediatric population to be used as a surrogate for our patients ' preinjury health state. The CHQ, which is intended for the pediatric population and has population norms available, was felt to be too long for practical use in the clinic setting, where visits typically last only 10 –15 min. Although not ideal either, we adapted outcome data from a healthy adult patient population to use as a benchmark for comparison. In particular, we used normative means from an age 18 –24 adult population for our SF-36 analysis. As a group, those patients who were able to participate in HRQoL surveys reported satisfactory NDI and VAS-NP outcomes. Of those patients completing surveys, mean VAS-NP scores of 0 demonstrated no pain on follow-up; mean NDI scores were consistent with "no disability" or only "mild disability. " Norm-based SF36 scores, however, were significantly decreased for PF, RP, and RE, with means well below the 25th percentile for each scale. GH and VT scores, conversely, were significantly increased, with a mean well above the 75th percentile in both cases. It remains hard to explain the

Table 3 Norm-based SF-36 outcomes compared to adult 18-24 normative means Norm-based SF-36 outcomes compared to adult 18–24 normative means

lack of concordance between GH and VT scores and PF, RP, and RE scores, but it is fair to conclude that at least in aggregate, the data suggest our cohort did not return to population norms, suggesting some persistent physical and emotional disability. Of course, as stated previously, no preinjury HRQoL measures are available for direct comparison, so definitive conclusions cannot be drawn. Nevertheless, these findings suggest that our course of aggressive surgical management of these injuries requires further study.

In a recent adult study by Dvorak et al. [32] using standardized outcomes tools, the SF-36 bodily pain and physical component score—a summary of the various physical concept/scale scores—specifically, the authors found that unilateral facet injuries of the subaxial cervical spine led to reported levels of pain that were significantly worse than those of the healthy population. They also suggested that nonoperatively treated patients, even in long-term follow-up, and despite having an initially "benign" fracture pattern, reported worse bodily pain and physical disability outcomes than operatively treated patients. In their series, patients managed operatively had better BP scores than those managed nonoperatively, but both groups reported outcomes that were significantly worse than those in the normative population. In our series, no significant difference was seen between bodily pain in our operative cohort and the adult, ages 18–24, normative population. This discrepancy in postsurgical pain outcomes between children and adults (better bodily pain outcomes in our pediatric cohort) may be due to better premorbid health status in children, lack of "secondary gain" from injury in children, and plasticity that may augment surgical tolerance and recovery in children. The Dvorak et al. series also showed significant differences in the physical component score between operative patients with greater than 18 months follow-up and nonoperative patients, but did not show a significant difference between the mean PCS of either operative or nonoperative patients and the normative population at large, suggesting no physical disability in either group compared to the norm. Calculation of PCS scores in our cohort, when available, resulted in a mean of 51.4 ($n=5$; range, 43.0– 59.1), which was not significantly worse than the mean PCS score for the adult normative population aged 18–24, with a mean difference of -2.1 ($p=0.52$) (Table 3).

Drawing definitive conclusions from this finding is problematic, however, as neither the operative nor the nonoperative adult cohorts reviewed by Dvorak et al. showed a significantly worse PCS score than the normative mean. Also, our series lack a nonoperative cohort to contrast with our operative cohort. Further meaningful comparison between SF-36 outcomes from their adult operative cohort and our pediatric cohort is difficult, as they did not report on individual SF 36 scale scores. In future studies, obtaining outcomes at several time points from the immediate aftermath of injury to late

follow-up would provide an interesting temporal profile of the process of recovery.

Limitations

There are several weaknesses in this study. Our study is small with a cohort consisting of only eight patients. It must be said, however, that the literature is sparse on this topic—consisting only of isolated case reports—and our series the first and largest to date. The retrospective nature of this study precludes a more inclusive, and meaningful, analysis of operatively treated children with unilateral facet injuries.

Given our findings, which suggest some level of postoperative physical and emotional disability, the absence of a nonoperative group for comparison with our operative cohort is a major limitation. Our study demonstrates a transparent but clear surgical bias. Literature review revealed two case reports of pediatric patients with unilateral facet injuries who were managed conservatively, as mentioned in our discussion. These patients were reported to be doing well on long-term follow-up, but were not evaluated using HRQoL measures that invite statistical comparison with our patient population. Given our findings—the presence of postoperative HRQoL impairment—it must be emphasized that nonoperative treatment with external immobilization remains an acceptable management strategy for these injuries. Further head-to-head comparison may reveal its relative inferiority or superiority.

Furthermore, our small cohort makes it difficult to comment on the advantages of a posterior approach $(n=5)$ versus an anterior approach $(n=2)$ or a combined approach $(n=1)$. Indeed, the surgical approach employed was heterogenous patients treated at TCH underwent posterior instrumented fusions, while the RCH cohort underwent anterior or combined anterior-posterior fusions—an institutional bias that might further confound meaningful conclusions about the suitability of either.

The pathology included was heterogeneous as well, involving the full spectrum of ligamentous and bony injury. The injuries ranged from a pure ligamentous disruption to a comminuted, displaced facet and lateral mass fracture, to a facet fracture as part of a three-column injury.

Because preinjury HRQoL data were not available, we were forced to compare our outcomes, assessed at a single point in time, to normative population outcomes. As there are no published normative pediatric values available, adoption of adult norms was necessary, though possibly inappropriate, for comparison. We extrapolated normative values from a healthy adult population, ages 18 to 24, a statistical assumption that weakens our results and conclusions.

The HRQoL outcomes tools that we used were not specifically validated for the pediatric population. As stated in the "Introduction" and "Discussion," the CHQ was deemed impractical for use in the functional clinic setting. Even though HRQoL outcomes not specifically validated for children were used, we maintain that the introduction of such outcome measures to a pediatric spine study, where such measures are rarely used, is valuable. In the future, we hope to work with pediatric spine societies, such as the Pediatric Craniocervical Society, to develop a common and practical HRQoL measure for the pediatric spine population.

Conclusions

To the best of our knowledge, this is the most comprehensive effort to analyze a series of surgically managed unilateral cervical facet injuries in the pediatric age group. Moreover, it is the only one using HRQoL tools. Unilateral facet injuries of the subaxial cervical spine comprise a complex and heterogenous array of bony and ligamentous injury. The management and treatment for unilateral facet injuries in the adult patient population may be controversial, but management and treatment recommendations for the pediatric patient population are nonexistent. In our small series, patients showed satisfactory clinical and radiographic outcomes, but reported varied HRQoL outcomes after undergoing surgical treatment that suggest some degree of persistent emotional and physical disability compared to a normative sample of young adult patients. These mixed HRQoL outcomes, weighed against the potential for continued instability and neurologic injury with nonsurgical treatment, necessitate further study with comparisons between operative and nonoperative cohorts for unilateral facet injuries in the pediatric age group.

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