Spondylolysis of C-2 in children 3 years of age or younger: clinical presentation, radiographic findings, management, and outcomes with a minimum 12-month follow-up

Clinical article

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Object. Cervical spondylolysis is a rare condition that results from a pars interarticularis defect. The C-6 level is the most frequently involved site in the cervical spine. Its clinical presentations range from incidental radiographic findings to neck pain and, rarely, neurological deficits. Although 150 patients with subaxial cervical spondylolysis have been reported, a mere 24 adult and pediatric patients with C-2 spondylolysis have been described. The long-term outcomes of very young children with bilateral C-2 spondylolysis are of great interest, yet only a few longitudinal studies exist.

Methods. The authors retrospectively reviewed 5 cases of bilateral C-2 spondylolysis at Texas Children's Hospital and Riley Children's Hospital; these were combined with 5 other cases in the literature, yielding a total of 10 patients. Data regarding the patients' age, sex, C2–3 angulation and displacement, associated spine anomalies, neurological deficits, treatment, and most recent follow-up were recorded.

Results. The patients' ages ranged from 3 to 36 months (mean 12.9 months). There were 6 boys and 4 girls. The C2–3 angulation, displacement, and width of pars defect were measured when available. The mean C2–3 angulation was 9.5° (range 1–34°), the mean C2–3 displacement was 4.78 mm (range 1.1–10.8 mm), and the mean width of the pars defect was 4.16 mm (range 0.9–7 mm). One patient developed myelopathy and spinal cord injury. All 10 of the patients were treated initially with conservative therapy: 3 with close observation alone, 1 with a rigid cervical collar, 4 with a Minerva jacket, 1 with a sternal-occipital-mandibular immobilizer, and 1 with a halo vest. Three patients ultimately underwent surgery for internal fixation due to progressive instability or development of neurological symptoms. All patients were neurologically intact at the last follow-up (mean 44.3 months, range 14–120 months).

Conclusions. Based on the literature and the authors' own experience, they conclude that most very young children with C-2 spondylolysis remain neurologically intact and maintain stability in long-term follow-up despite the bony defect. This defect is often an asymptomatic incidental finding and may be managed conservatively. More aggressive therapy including surgery is indicated for those patients with a neurological deficit from spinal cord compromise secondary to stenosis and local C-2 kyphosis, progressive deformity, or worsening C2–3 instability. (http://thejns.org/doi/abs/10.3171/2013.11.PEDS13422)

Key Words • bilateral pars defect • C-2 spondylolysis • pediatric spine • cervical spine • axis

Abbreviations used in this paper: NAT = nonaccidental trauma; SOMI = sternal-occipital-mandibular immobilizer.

* Drs. Gressot and Vadivelu contributed equally to this work.
Spondylosis of C-2 in very young children

There are limited reports of C-2 spondylolysis. Most cases are diagnosed in adulthood. There are even fewer longitudinal studies examining the long-term implications and natural history of this bony defect in the immature spine of very young children (3 years of age or younger). The C-2 vertebral body is anatomically and biomechanically distinct from other vertebral bodies; moreover, peculiarities of the spinal column in young children include inherent laxity and elasticity, and incomplete ossification; disproportionately larger heads with underdeveloped neck muscles; more horizontally oriented facet joints, resulting in greater mobility and less stability; and physiological wedging of vertebral bodies, facilitating forward movement of vertebrae. Therefore, the presentation, rate of instability, degree of associated neurological compromise, and subsequent treatment of C-2 spondylolysis in very young children may differ from those in adolescent or adult patients.

In this study we describe 5 cases of C-2 spondylolysis in children who were 3 years of age or younger. We followed them for at least 12 months, and show that in most cases spinal stability is maintained and may be treated conservatively. We also review the literature for an additional 5 cases, describing the presentation, stability of this lesion, risk of neurological compromise, and indications for conservative treatment versus surgery.

Methods

We retrospectively reviewed 4 consecutive cases of bilateral C-2 spondylolysis treated at Texas Children’s Hospital and 1 treated at Riley Children’s Hospital. We then performed a literature search and identified 5 other cases, which we combined with our patients, yielding a series of 10 patients (Table 1). Institutional review board approval was obtained from our respective institutions. We collected data including age, sex, associated spinal anomalies, presence of neurological deficits, motion on dynamic imaging, management, and follow-up for each patient. We measured the angulation by calculating the angle (α) formed by the inferior endplate of C-2 relative to the inferior endplate of C-3 (Fig. 1, α), and we measured the displacement of C-2 relative to C-3 by measuring the distance between the posterior aspect of the C-2 body and the posterior vertebral body of C-3 (Fig. 1, γ) as described by Levine and Edwards and later reviewed by Bono et al. We measured the width of the pars defect by the horizontal distance between bony elements (Fig. 1, β). Only patients with a minimum of 12 months of follow-up were included in the study.

Case 1

This 12-month-old girl without significant medical comorbidities presented in the emergency department after sustaining a low-impact fall at home. She was noted to have torticollis after the fall, which resolved after an “adjustment” from a chiropractor. Torticollis recurred and the patient was taken to an emergency department. The patient was neurologically intact and her parents noted no neck pain, but she did have tenderness to palpation of the cervical spine. The CT scans of the cervical spine demonstrated bilateral pars defects without displacement or involvement of the transverse foramina (Fig. 2A and B). There was 5° of C2–3 angulation and 3-mm C-2 anterior listhesis relative to C-3, but no evidence of acute soft-tissue injury or hematoma. At that time, she was placed in a rigid cervical collar, but was later transitioned to a Minerva jacket due to excess motion in the collar. She was followed closely with a repeat flexion-extension cervical spine radiograph prior to each follow-up visit (Fig. 2C and D). No instability was identified and there was also no evidence of healing, which was mounting evidence that the defect was congenital, and not acute fractures. This was further substantiated by clear smooth sclerotic margins along the defect. The patient wore the brace for 3 months, and follow-up flexion-extension radiographs showed no instability and stable defect. She is doing well at 48 months postinjury.

Case 2

This 14-month-old girl presented with concern for nonaccidental trauma (NAT). She was born at 33 weeks via cesarean section for premature rupture of membranes but without trauma, and later developed Kawasaki disease. She presented with multiple episodes of vomiting and periportal edema but without focal neurological deficit. Head CT scans demonstrated a nondisplaced orbital roof fracture, tonsillar descent of 4 mm, and cervical spine bilateral pars defects with smooth margins. The angulation of C-2 to C-3 was 1°, and there was 3-mm C-2 anterior listhesis with a 6.4-mm pars defect. An MRI sequence of the cervical spine was obtained, demonstrating mild posterior soft-tissue edema, a small synchondroses at C-6, and similar tonsillar ectopia. The patient was placed in a Minerva jacket for 3 months. Flexion-extension radiographs have demonstrated no instability and the patient was without pain or neurological deficit, giving reason to discontinue the orthosis. She continues to do well at 14-month follow-up.

Case 3

This 10-month-old boy presented with torticollis to the right and tenderness to palpation of his cervical spine. His CT scans were remarkable for several craniovertebral junction abnormalities including bilateral C-2 pars defects, platybasia, absence of anterior cartilaginous ring of C-1, rightward atlantoaxial rotation, and anterior subluxation of C-1 on C-2 and C-2 on C-3. The C2–3 angulation was 34°, with 6-mm C-2 anterior listhesis and a 4.5-mm pars defect. The MRI studies demonstrated reduced rotatory subluxation, posterior interspinous ligamentous signal intensity without evidence of disruption, and no evidence of acute bone edema. He was also noted to have bilateral clubfoot. He was initially without neurological deficit and was placed in a Minerva jacket with close follow-up.

After approximately 6 weeks in the Minerva brace he developed bilateral thenar wasting and hand weakness as well as diffuse hyperreflexia. Repeat cervical radiographs demonstrated increased widening of C-2 pars defect to 8.1 mm, worsening of angulation to 41°, and worsening C2–3

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<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>Case No.</th>
<th>Age (mos), Sex</th>
<th>Presentation</th>
<th>α (°)</th>
<th>β (mm)</th>
<th>γ (mm)</th>
<th>F/E Radiographs</th>
<th>Associated Spine Abnormalities</th>
<th>Neuro Deficit</th>
<th>Management</th>
<th>FU (mos)</th>
<th>Outcome at Last FU</th>
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<td>present study</td>
<td>1</td>
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<td>none</td>
<td>3 mos rigid collar, then Minerva</td>
<td>48</td>
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<td></td>
<td>2</td>
<td>14, F</td>
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<td>8 → 10†</td>
<td>5.5 → 7†</td>
<td>3.8 → 10.8†</td>
<td>14-mm subluxation of C-2 on C-3</td>
<td>assimilation of atlas by occiput, flattening of C1–condyle joint</td>
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<td>none</td>
<td>observation</td>
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</table>

* Ant = anterior; F/E = flexion/extension; FU = follow-up; neuro = neurological; NR = not reported.
† Arrow indicates progression from 2 to 5 years.
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Case 4

This 4-month-old girl presented with torticollis, which was noted shortly after birth. A CT scan of the cervical spine showed bilateral C-2 pars defects and fusion of the posterior arch of C-1 and the left lateral mass of C-1 to the occiput, possible platybasia, flattening of the C1–condyle joint, and a poorly formed odontoid (Fig. 3A and B). Results of her neurological examination at the time were unremarkable and she had 8° of C2–3 angulation, 3.8 mm of C-2 anterior listhesis relative to C-3, and a pars defect of 5.5 mm. She was initially followed with close observation. At 13 months she was noted to have mild progression of deformity and was placed in a Minerva jacket. At 2 years of age the patient remained asymptomatic and was meeting all motor milestones despite hypermobility on flexion-extension radiographs. The Minerva jacket was removed and the patient remained asymptomatic until 5 years of age when her parents noted pain on extension of the neck. Cervical radiographs showed 10° of C2–3 angulation, 10.8 mm of C-2 anterior listhesis, 14-mm C2–3 subluxation on dynamic radiographs, and a pars defect measuring 7 mm (Fig. 3C and D). An MRI study showed focal C2–3 myelomalacia (Fig. 3E), so the patient underwent occipitocervical fusion. The construct consisted of occipital screws, bilateral C-3 and C-4 lateral mass screws and a left C-5 lateral mass screw, with cancellous allograft and bone morphogenetic protein. She tolerated the procedure well and remained anterior listhesis of 10.7 mm. No preoperative dynamic radiographs were obtained. The patient then underwent internal fixation in consideration of worsening instability and development of neurological deficit. An occipitocervical fusion (O–C4) was performed. Due to the patient’s small size, only unilateral mass screws could be placed with fluoroscopy at C-3 (left side) and C-4 (right side), along with 4 occipital screws and parallel rods for the construct. Bone morphogenetic protein as well as bone matrix were used to augment autologous bone graft. Postoperative CT cervical spine scans and radiographs showed that the hardware was in an acceptable position and at an improved angulation of 8°, C-2 anterior listhesis was 2.6 mm, and there was no instability on dynamic imaging. He is grossly neurologically intact and was doing well at last follow-up at 36 months.
neurologically intact. Postoperative imaging including CT scans and radiographs showed that the hardware was in an acceptable position, with an angulation of 0° degrees, C-2 anterior listhesis of 3.9 mm, and no instability on dynamic imaging (Fig. 3F and G). She continues to do well 3 months after surgery, and 48 months from diagnosis.

**Case 5**

This 3-month-old girl was admitted for failure to thrive. She had a history of multiple congenital abnormalities including Pierre-Robin sequence, vesicoureteral reflux, micrognathia, cleft lip and palate, bicuspid aortic valve, and a patent foramen ovale. On examination, she kept her head in an extended position but moved all extremities without spasticity. Her imaging studies showed bilateral C-2 pars defects and C-2 subluxation. The angulation of C-2–3 was 8°, and there was 1.1-mm C-2 anterior listhesis with a 2-mm pars defect. Flexion-extension radiographs demonstrated C2–3 hypermobility with anterior listhesis. She was placed in a sternal-occipital-mandibular immobilizer (SOMI) brace. The patient had frequent hospitalizations due to her multiple comorbidities, ultimately requiring a tracheostomy. At 21 months of age she was without pain or neurological deficit and had stable imaging. She was transitioned to a hard cervical collar and is doing well at 23-month follow-up, although she has developed thoracic scoliosis.

**Results**

We reviewed 5 patients who were treated for C-2 spondylolysis at our institutions as well as 5 other cases identified in the literature. The range of patient ages was 3–36 months, with a mean age of 12.9 months. There were 4 girls and 6 boys in this series. The mean follow-up was at 44.3 months (range 14–120 months).

The C2–3 angulation, displacement, and width of pars defect were measured when available. The mean C2–3 angulation was 9.5° (range 1–34°), the mean C2–3 displacement (γ) was 4.78 mm (range 1.1–10.8 mm), and
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the mean width of the pars defect (β) was 4.16 mm (range 0.9–7 mm). Five patients had associated spinal anomalies, including 1 with a 2-mm C-6 syrinx and minimal tonsillar ectopia (Case 2); 1 with bilateral L-4 and L-5 pars defects; 2 with craniocervical junction abnormalities (Cases 3 and 4); and 1 with thoracic scoliosis (Case 5). Three patients had systemic diseases: Menke syndrome (Case 10); pyknodysostosis (Case 6); and Pierre Robin sequence with bicuspid aortic valve, vesicoureteral reflux, micrognathia, cleft lip and palate, and a patent foramen ovale (Case 5). All patients were neurologically intact at presentation; 1 patient developed myelopathy consistent with a spinal cord injury (Case 3), and another child developed asymptomatic T-2 cord signal change at C2–3 (Case 4).

All patients initially received conservative management; 3 with observation only and 7 with bracing. Three children ultimately underwent surgery. One patient (Case 3) developed myelopathy, bilateral hand weakness, and progression of deformity after 6 weeks of conservative therapy, and underwent occipitocervical fusion. One (Case 8) initially underwent observation only and had C1–3 fusion after 1 year, when he developed instability on follow-up dynamic imaging. Another patient (Case 4) had hypermobility that was persistent despite 12 months of bracing. This patient was followed for 4 years until she developed pain and asymptomatic MRI changes (myelomalacia of the spinal cord at the level of C-2), at which point she underwent an occipitocervical fusion. All patients whose disease was managed surgically in a delayed fashion are doing well at follow-up without neurological deficit.

Discussion

Bilateral C-2 spondylolysis in very young children is rare and presents diagnostic and management challenges. It is frequently identified as an incidental finding on radiographic studies, and no clear guidelines have been established in the literature to guide treatment decisions. This condition is caused by abnormal development of the posterior elements of C-2, and the radiographic findings of bilateral C-2 spondylolysis are confusing and can easily be misinterpreted as an acute fracture or dislocation. Concerns of an acute high cervical spine injury—especially in an immature spine—characterized by ligamentous laxity, large head with weak cervical musculature, and horizontally oriented facets may portend a poor natural history and poor potential for long-term healing, and thus indicate aggressive early therapy including surgical fixation. Instrumentation in very small children is difficult due to their small size, concerns of long-term growth potential, less ossified bone, and the inherent congenital anomaly in these patients. We believe that in contrast to traumatic bilateral spondylolysis, congenital bilateral C-2 spondylolysis is often an incidental finding and can be managed conservatively. We demonstrate in our combined series of 10 patients that most very young patients managed conservatively (70%) remain asymptomatic without instability at long-term follow-up. Of the patients who underwent surgery in a delayed fashion, all are doing well without evidence of morbidity incurred by delaying definitive fixation.

Congenital Origin

The origin of C-2 spondylolysis is considered to be congenital, which is supported by the absence of healing or evolution of the defect in follow-up; the presence of smooth, corticated margins of the defect; the frequent association with other spinal deformities; and the association with other systemic congenital syndromes and diseases. Spondylolysis of C-2 typically presents as a radiographic triad of the following: 1) absence of C-2 pars interarticularis with falsely enlarged appearing neural foramen; 2) dysplastic ipsilateral transverse process; and 3) posteriorly displaced dysplastic ipsilateral lamina and articular pillar.

Several findings in our combined series of 10 patients support the theory of a congenital origin for C-2 spondylolysis. It has been proposed that C-2 spondylolysis results from a primitive defect in the chondrification of vertebral body mesenchymal precursors. This hypothesis may explain the high incidence of associated spinal anomalies seen in patients with C-2 spondylolysis. Three (30%) of the 10 children had one or more additional spine anomalies including cervical or lumbar spondylolysis, segmentation anomalies, fused vertebral bodies or posterior elements, and failure of normal fusion of posterior elements. As seen in the 2 of 3 patients in our series who underwent instrumented fusion, the presence of other bony abnormalities at the craniocervical junction, including platybasia, basilar invagination, segmentation, and formation anomalies, seems to be a risk factor for developing instability requiring surgery.

Establishing whether the child has a history of recent or remote trauma is an important consideration. In our combined series, only 4 of the 10 children had a history of known or suspected trauma. Two patients were evaluated due to concerns about NAT. Even in patients with a history of trauma, the imaging characteristics of the C-2 spondylolysis were consistent with a congenital defect as opposed to acute fracture. All patients had smooth, well-corticated margins of defect on imaging, no signs of healing or remodeling, and no soft-tissue edema or evidence of acute trauma.

Patient Presentation

Spondylolysis of C-2 is frequently noted as an incidental finding on routine imaging performed for other reasons, including NAT evaluation (Cases 2 and 3), sinusitis (Case 8), or during evaluation of a systemic disorder (Cases 5, 6, and 10). Two patients were evaluated after minor trauma (Cases 1 and 7), but the imaging characteristics were consistent with a congenital origin and not acute traumatic fracture. The remaining 2 children presented symptomatically with torticollis (Cases 4 and 9).

Four children (40%) demonstrated instability at C2–3 on dynamic radiographs and 3 children (30%) underwent internal fixation. One patient (Case 8) had no evidence of instability at the time of presentation, and developed subluxation at C2–3 after 1 year of conservative therapy, requiring surgical intervention. Another patient (Case 4) had asymptomatic hypermobility at C2–3 as well as the craniocervical junction, which was treated conservatively,
and followed for 4 years until she developed pain. At this point repeat MRI studies showed a small region of asymptomatic myelomalacia, and she subsequently underwent an occipitocervical fusion. A third patient (Case 3) had atlantoaxial subluxation indicative of instability at presentation (which spontaneously reduced) as well as platybasia, absence of anterior cartilaginous ring of C-1. This patient initially received conservative management but developed myelopathy, tenar wasting, and progression of deformity, and thus underwent occipitocervical fusion. Another patient (Case 5) showed hypermobility at C2–3 on flexion-extension radiographs but remains neurologically intact without pain after a period of bracing with a SOMI, followed by a hard cervical collar. All patients are doing well without neurological deficit at last follow-up.

Treatment Options

Conservative. All patients initially underwent non-operative management. The treatment approaches included observation alone with close clinical and radiographic follow-up (3 patients) or bracing with a Minerva jacket (4 patients), halo vest (1 patient), SOMI (1 patient), or rigid cervical collar (1 patient). Duration of bracing ranged from 3 to 18 months. Adequately immobilizing very young children in an orthotic device is difficult due to their small size and lack of cooperation, and is fraught with the potential for complications. One child reported by Smith et al. was treated for a presumed fracture with halo immobilization. The patient fell during play, resulting in the stabilizing pins penetrating the calvaria, requiring replacement under general anesthesia. The child subsequently developed a pin site infection, at which point it was determined that the defect was probably a synchondrosis and not a fracture, and the halo vest was then removed without sequelae. In an asymptomatic child, even with abnormal motion at C2–3 on dynamic imaging, conservative treatment may still be appropriate initially. In our combined series, 7 (70%) of the 10 children remained neurologically intact with stable imaging, and were asymptomatic at follow-up with conservative treatment only.

Surgery. Three patients in our combined series of 10 patients underwent internal fixation. Conservative management failed in only 1 patient, at 6 weeks, with the development of worsening deformity and myelopathy necessitating surgery (Case 3). The patient attained reduction in deformity and resolution of instability postoperatively. At follow-up this patient is grossly neurologically intact and continues to do well. Two patients (20%) required fusion due to instability or the development of symptoms at 12 months and 4 years, respectively. Neither patient developed morbidity due to delay in definitive treatment, and this delay allowed for growth and development, mitigating some of the challenges in placing spinal instrumentation in very young children. In general, surgical intervention is indicated in very young children with C-2 spondylolysis who have a neurological deficit, significant instability resulting in canal compromise, or rapidly progressive deformity. Risk factors predisposing a patient to surgical intervention include the presence of bony abnormalities at the cranioce-
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consisting of stabilization is reserved for patients with C-2 spondyloysis associated with significant C2–3 instability or cervical canal stenosis with neurological deficits, or associated with significant structural bony abnormalities at the craniocervical junction.

Disclosure

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

Author contributions to the study and manuscript preparation include the following. Conception and design: Jea. Acquisition of data: Vadivelu, Fulkerson. Analysis and interpretation of data: Jea, Gressot, Vadivelu, Fulkerson. Drafting the article: Jea, Vadivelu. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Jea. Study supervision: Jea, Luerssen.

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