Progressively Unstable C2 Spondylolysis Requiring Spinal Fusion: Case Report

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Abstract

Cervical spondylolysis is a rare condition defined as a corticated cleft at the pars interarticularis in the cervical spine. This is the case of C2 spondylolysis demonstrating progressive significant instability, which was successfully treated by anterior cervical discectomy and fusion (ACDF) with cervical anterior plate. We describe a 20-year-old female with C2 spondylolysis presenting with progressive worsening of neck pain associated with progressive instability at the C2/3 segment. The progression of instability was well-documented on flexion-extension cervical spine x-rays. She was successfully treated by C2/3 ACDF with anterior cervical plate. Her preoperative significant neck pain resolved immediately after the surgical intervention. She was completely free from neurological symptoms at 1-year postoperative follow-up. We also review the literature and discuss 24 reported cases with C2 spondylolysis. When planning treatment, we should make sure to differentiate this pathology from acute traumatic fracture, which is a hangman's fracture. Assessment of C2/3 instability associated with neurological deficits is extremely important to consider management properly. C2/3 ACDF with cervical plate is biomechanically viable, less invasive, and provides adequate surgical stabilization for unstable C2 spondylolysis.

Key words: C2 spondylolysis, hangman's fracture, anterior cervical discectomy and fusion, instability

Introduction

Cervical spondylolysis is a condition defined as a corticated cleft at the pars interarticularis[1–3] in the cervical spine and most often diagnosed incidentally after minor injury. It is commonly asymptomatic. This pathology is a rare condition, and to date there have been 150 cases reported in the literature.[4] C6 is the site most commonly involved and accounts for about 70% of cases described in the literature,[3] followed far less frequently by the C4 and C5 levels, but cervical spondylolysis may occur at any level from C2 to C6.[1,3,4] The C2 vertebra is anatomically distinct from other cervical vertebrae; and thus, the presentation, rate of instability and neurological compromise, and treatment of C2 spondylolysis may also differ.[4,5] Furthermore, the location is anatomically similar to a hangman's fracture and differentiation of these two pathologies is essential for surgical decision making.

In this article, we describe a case of symptomatic C2 spondylolysis in a healthy, athletically active young female who demonstrated progressive C2/3 instability

and was successfully treated with C2/3 anterior cervical discectomy and fusion (ACDF) with anterior cervical plate. This case illustrates several important points that must be considered when we plan the surgical strategy. We also review the literature and discuss the proper management of this rare lesion.

Case Report

I. Clinical history

This 20-year-old female was initially assessed at a children's hospital 7 years before, when she was incidentally discovered to have unspecified abnormal lesion involving the craniovertebral junction, on cervical spine x-ray and computerized tomography (CT) as part of investigation of two syncopal episodes. This imaging work-up revealed C2 spondylolysis associated with a hypoplastic posterior C1 arch. The patient was asymptomatic at this time and flexion-extension cervical spine x-rays did not show any instability. Afterward, she had kept participating in many sports including soccer and hockey where she had sustained multiple injuries as usual. Four years later, she began to develop mild neck pain. Repeat lateral cervical
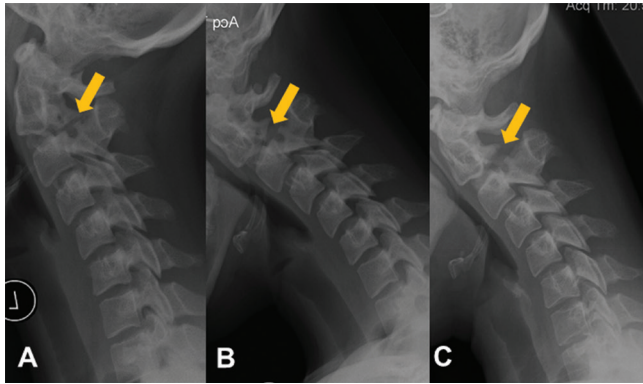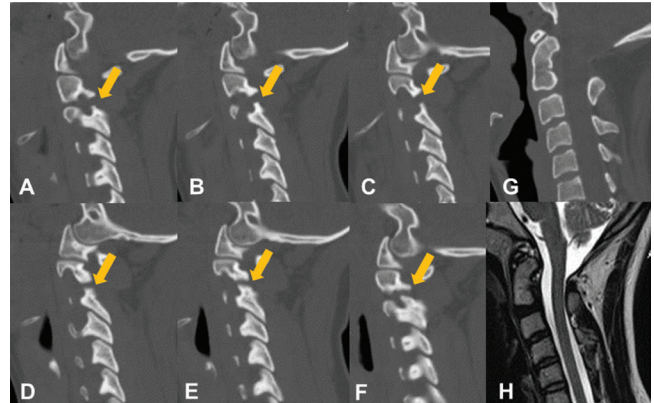
Fig. 1 A: Cervical spine x-ray obrtained three years bofore her first visit to us shows C2 spondylolysis (*arrow*) and kyphotic deformity of the whole spine. B: The flexion and extension x-ray at this point shows the gap in the C2 pars defect (*arrow*) widens from 0 mm in extension to 4 mm in flexion. C: The flexion and extension x-ray at her first visit to us shows the gap of C2 pars defect (*arrow*) widened 6 mm in flexion, which suggests progressive cervical spinal instability.

Fig. 2 Sagittal cervical spine computed tomography (para-median plane on the left side; A–C, paramedian plane on the right side; D–F, midline plane; G) reveals a deformity of the odontoid with posterior angulation of the tip, a partial absence of the posterior arch C1 and a fusion of the posterior arch of C2 and C3 with a bilateral spondylolysis (*arrow*) of the C2 pars interarticularis. Cervical spine magnetic resonance imaging (H) demonstrates no evidence of spinal cord compression, ligament injury, hematoma and soft tissue injury, which help us rule out hangman fracture.

spine x-ray demonstrated C2 spondylolysis (Fig. 1A, *arrow*) and hyperlordosis at C1/2 segment as well as an angular deformity at the C2/3 disc space. Repeat flexion and extension x-ray showed the gap in the C2 spondylolysis widened from 0 mm in extension to 4 mm in flexion (Fig. 1B, *arrow*). No surgical intervention was offered to her back then because her neck pain was not that severe and she was otherwise neurologically intact. She started to restrict her activities following this episode. Shortly thereafter she began to experience worsening neck pain and headache, prompting referral to us.

II. Examination

The patient was alert, slender in her body morphology, and looked appropriate for her age. Physical examination showed normal cervical range of motion and no neurological deficit other than significant neck pain. Cervical spine x-ray demonstrated kyphotic deformity of the whole spine with compensatory hyperlordosis at C1/2. The flexion and extension x-ray showed C2 spondylolysis with a gap that widened from 0 mm in extension to 6 mm in flexion (Fig. 1C). The gap at the defect of C2 pars interarticularis had widened over time compared to the imaging obtained 3 years beforehand (Fig. 1B), suggestive of progressive cervical spinal instability. Cervical spine CT sagittal image (Fig. 2A–G) revealed a deformity of the odontoid with posterior angulation of the tip, a partial absence of the posterior arch of C1, and a fusion of the posterior arch of C2 and C3 with a bilateral C2 spondylolysis. CT axial image (Fig. 3A–J) demonstrated well-corticated clefts on both sides of the spondylolytic defects. Cervical spine magnetic resonance imaging (MRI) demonstrated no evidence of spinal cord compression,

spinal cord signal change, ligament injury, disc disruption, soft tissue injury, or hematoma (Fig. 2H). We were able to rule out acute spinal cord injury and acute hangman fracture based on these radiographic imaging findings. As such, we believed that her symptoms of severely worsening neck pain were related to her progressive spinal instability. Consequently, we decided to perform surgical intervention to correct the instability and alleviate the risk of severe neurological deficit from minor trauma.

III. Clinical course

Surgical management of this patient included C2/3 ACDF with interbody fusion polyetherether ketone (PEEK) cage (Solis PEEK; Stryker Spine, Allendale, New Jersey, USA) and anterior cervical plate (Reflex Hybrid Anterior Cervical Plate System, Stryker Spine, Allendale, New Jersey, USA). We considered the combination of the interbody fusion cage and anterior cervical plate was necessary for progressive instability over years. The patient was placed supine with her neck slightly extended by placing a small pillow under her shoulders. We made the transverse neck incision in a natural skin crease at about the level of the hyoid bone on the right side. Limited flaps were raised inferiorly and superior in the subplatysmal plane with identification of the marginal mandibular nerve as it transverse the submandibular gland. Care was taken to remain below the submandibular triangle and marginal mandibular nerve pathway as well as hypoglossal nerve at the depth of the digastric muscle. Marginal mandibular nerve was tested by a facial nerve stimulator, and shown

to stimulate normally then moved superiorly with gentle blunt retraction to remain clear of the surgical approach. The ansa hypoglossal nerve bundle was identified crossing from superior to inferiorly toward the lateral strap muscles and retracted superiorly. The superior laryngeal nerve was not encountered in the operative field. The plane between the sternomastoid muscle medial border and the lateral border of the straps was widely opened vertically. Finger blunt dissection was done in the investing fascia plane posterior to the pharynx, anterior to the spine vertically, which led us to sufficient access to the C2/C3 vertebrae. We used Caspar Cervical Distractor System (Aesculap Implant Systems; Center Valley, Pennsylvania, USA) for discectomy and instrumented fusion. After the discectomy was completed, the hollow Solis PEEK cage was filled with synthetic bone graft (Vitoss Bone Graft Substitute; Stryker Spine, Allendale, New Jersey, USA) and put into the disc space with disctraction force applied. Subsequently, anterior cervical plate was implanted with compression force applied under fluoroscopic guidance. We confirmed the cervical spine is well aligned after all procedures. There were no adverse reactions to the surgical procedure and blood loss was 50 ml. On immediate postoperative cervical spine x-rays, satisfactory hardware placement was achieved. Her neck pain disappeared immediately after the surgery. At 1-year follow-up, the patient was completely neurologically intact and cervical spine x-rays showed rigid fixation and excellent spine alignment (Fig. 4).

Discussion

Spondylolysis of C2 is a rare condition with limited previous reports in the literature. A review of the litera-
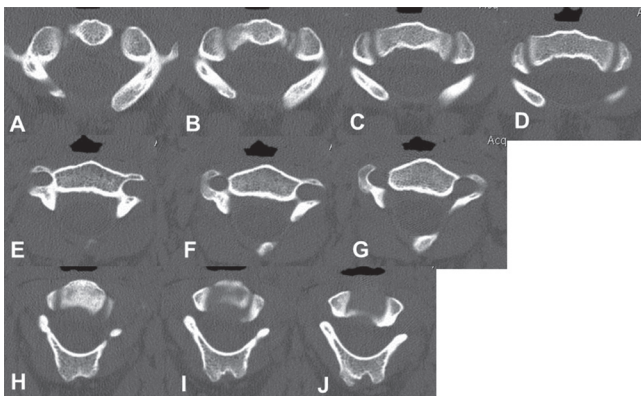
ture reveals characteristics of this rare entity, which is listed in Tables 1 and 2. Table 1 shows 7 cases with C2/3 instability including present case[5–10] and Table 2 shows 18 cases without C2/3 instability,[4–6,10–18] Three out of six reported cases with spinal instability went through surgical intervention.[5,7,8] We describe the case who demonstrated progressive significant spinal instability and was successfully managed by ACDF. This case illustrates several important points that must be considered when approaching surgical management of this rare condition.

I. Diagnosis

When planning treatment, either with a conservative approach or surgery, it is important to differentiate C2 spondylolysis from acute traumatic fracture, such as a hangman's fracture.[4,17] For this purpose, cervical spine CT and MRI are imperative (Fig. 4). Previous reports[3,4,17,19,20–22] outlined several points in this regard, namely, we should find, based on CT, (1) a well-marginated cleft or a sclerotic margin with a triangular configuration of the pillar fragments on either side of the spondylolytic defect, (2) compensatory hyper- or hypoplasia of the ipsilateral articular pillars at the level above and/or below the defect, and (3) the presence of associated dysplastic changes at the involved level. We should also make sure of absence of edema in the fractured bones, soft tissue swelling, disruption of the intervertebral disc, ligamentous injury, hematoma based on MRI. Furthermore, absence of neurological symptoms associated with acute trauma is

Fig. 3 Axial images of cervical spine computed tomography (A–J) show a partial absence of the posterior arch of C1 (A–D) and triangular pillar fragments with well-corticated and well-defined margins on both sides of the spondylolytic defects of C2 (E–J). These imaging characteristics indicate congenital dysgenesis of pars interarticularis, which is completely distinct from acute hangman's fracture.

Fig. 4 Cervical spine x-ray at 1-year postoperative follow-up revealed rigid fixation and excellent spinal alignment.

Table 1 Cases with C2/3 instability

	Age/Sex	Reason for assessment	Instability	Spinal cord compression	Management	FU	Presentation at final FU
Farwdon and Fielding (1981)[7]	5 yrs/ M	Clicking of neck, neck pain	Flexion-extension instability	Not mentioned	ACDF followed by neck collar for 9 months	36 mos	Complete resolution
Nordström (1986)[10]	9 yrs/ F	Incidentally found after neck trauma	Flexion-extension instability	Not mentioned	Skull traction followed by neck collar for 6 weeks	19 mos	Asymptomatic
Currarino (1989)[6]	4 mos/ M	Incidentally found in the work-up for skeletal dysplasia	Flexion-extension instability	Not mentioned	Cervical collar for 3 months and FU with serial imaging	52 mos	Asymptimatic imaging unchanged
Hinton (1993)[8]	22 yrs/ M	Transient four limbs numbness, weakness	Intraoperative hypermobility of C2 laminae as opposed to negative radiographs	Yes, due to medialization of C2 laminae into the spinal canal	C1/3 posterior fusion with C2 laminectomy	3 mos	Complete resolution
Howard and Letts (2000)[5]	3 yrs/ M	Incidentally found after sinus infection	Progressive flexion-extension instability	Not mentioned	C1/3 posterior fusion	12 mos	Asymptomatic
Kubota (2003)[9]	57 yrs/ F	Myelopahty	Rotational instability	Intermittent compression caused by neck rotation	Limitation of activity and FU with serial imaging	6 mos	Unchanged
Present case	20 yrs/ F	Severe neck pain	Progressive flexion-extension instability	No	ACDF	12 mos	Complete resolution

ACDF: anterior cervical discectomy and fusion, F: female, FU: follow-up, M: male, mos: months, yrs: years.

Table 2 Cases without C2/3 instability

	Age/Sex	Reason for assessment	Instability	Spinal cord compression	Management	FU	Presentation at final FU
Gehweiler (1977)[11]	34 yrs/F	Neck pain, paresthesia of arm	No	Not mentioned	No	No	NA
Matthews et al. (1982)[14]	11 yrs/F	Incidentally found after neck trauma	No	Not mentioned	No	No	NA
Kish and Wilner (1983)[13]	30 yrs/F	Neck pain, paresthesia of arm	No	No	Cervical collar	No	NA
Hasue et al. (1983)[12]	21 yrs/F	Incidentally found after neck trauma	No	Not mentioned	Bedrest for 5 days	12 mos	Asymptomatic
	7 yrs/M	Neck pain	No	Not mentioned	FU with serial imaging	36 mos	Improved
	41 yrs/F	Incidentally found after neck trauma	No	Not mentioned	No	No	NA
	35 yrs/F	Incidentally found after neck trauma	No	Not mentioned	No	No	NA
Nordström (1986)[10]	37 yrs/M	Family evaluation	No	Not mentioned	No	No	NA
Currarino (1989)[6]	2 mos/M	Incidentally found in the work-up for Crouzon disease	No	Not mentioned	No	No	NA
	2 yrs/M	Incidentally found in the work-up for epiglottitis	No	Not mentioned	No	No	NA
Riebel and Bayley (1991)[15]	5 yrs/M	Incidentally found after neck trauma	No	No	No	No	NA
Smith et al. (1993)[16]	18 mos/M	Incidentally found after neck trauma	No	No	Halo vest for 5 mos because of misdiagnosis as an acute fracture	66 mos	Asymptomatic
Williams et al. (1999)[18]	2 yrs/F	Incidentally found after neck trauma	No	No	No	No	NA
Howard and Letts (2000)[5]	9 mos/M	Torticollis	No	Not mentioned	FU with serial imaging	120 mos	Unchanged
	3 yrs/M	Incidentally found after aspiration of food	No	No	No	No	NA
Rijn et al. (2005)[17]	5 mos/F	Incidentally found after neck trauma	No	No	FU with serial imaging	1 mo	Asymptomatic
Gottfried et al. (2010)[4]	16 yrs/M	Subjective hypermobility of neck	No	No	FU with serial imaging	24 mos	Asymptomatic
	23 yrs/F	Incidentally found after neck trauma	No	No	FU with serial imaging	12 mos	Asymptomatic

F: female, FU: follow-up, M: male, mo(s): month(s), NA: not available, yrs: years.

significantly important as well.

II. Instability

Only 6 out of 24 patients with C2 spondylolysis were reported to exhibit significant instability at the C2/3 segment[5–10] on preoperative imaging or intraoperative findings. All three surgically-treated cases were included in these cases and the rationale for surgical intervention was spinal instability with/without neurological manifestations. Some authors judged the presence of C2/3 instability based on only static x-rays, however, we believe dynamic imaging including flexion-extension x-rays would provide more accurate information. In the present case, progression of instability was first detected on flexion-extension x-rays although static x-rays were unchanged. Four out of six cases with spinal instability, all of which were children, were clearly discovered to have unstable spine on flexion-extension x-rays. Kubota et al.[9] showed an interesting case with rotatory spinal instability, whose neck pain and progressive myelopathy had deteriorated after neck rotation. Flexion-extension instability was not detected, but spinal cord was squeezed by dysplastic isthmus when neck was rotated on dynamic CT myelography. Hinton et al.[8] reported one case with transient upper extremity weakness associated with sudden neck motion, where spinal cord was compressed by congenital dysplastic C2 laminae. Surgical findings disclosed C2 laminae was hypermobile in spite of no evidence of instability on preoperative imaging. No cases without spinal instability required surgical treatment and had remained neurologically stable for a long time of follow-up. From these reports, deterioration of this congenital pathology is highly suspected to be involved with some sort of instability. We should always try to search for the evidence of instability in patients with neurological deficit including neck pain. Even if there is no obvious radiographic instability, we cannot deny the presence of instability, which could be first detected during the surgery just like the case reported by Hinton et al.[8] Present case had been athletically active and joined vigorous daily sports activities, where she had sustained some minor trauma, but no defining event of major trauma that precipitated a sudden worsening. Some authors have asserted that initially dysplastic vertebra is more predisposed to microtrauma.[3,23] In addition, her congenital kyphotic C2/3 posterior element synostosis may have altered kinematics, have exerted extra stress on the adjacent mobile segment, which was C2 spondylolysis, and have resulted in progressive segmental instability.

III. Treatment

Our surgical indications for C2 spondylolysis include instability associated with neurological deficits and progressive instability irrespective of symptoms. If a patient is asymptomatic and demonstrates no abnormal movement on flexion-extension images, then there may not be an indication for aggressive treatment. Immobilization is only an effective treatment in such cases.[4] There are three reported cases with instability who were managed with skull traction, neck collar, or restriction of activity.[6,9,10] One case declined the surgery and two other cases were asymptomatic 4 month-old and 9 year-old children, both of which were fortunately stable at the last follow-up. However, we cannot expect spontaneous fusion of congenital spondylolysis, therefore, patients with intractable neck pain or neurological deficits associated with instability or spinal cord compression should be considered as surgical candidates.[1,3,20] Three patients with unstable spine were reportedly operated for stabilization at C2/3, all of which resulted in excellent postoperative outcome.

IV. Surgical strategy

Three reported surgeries for C2 spondylolysis were: one C2/3 ACDF, and two C1/3 posterior instrumented fusions.[4] Recently, direct transpedicular screw fixation of acute hangman fractures with compression using lag screws has been reported under computer-assisted image guidance, such as Iso-C[3D]-based computer navigation[24] or three-dimensional image (O-arm)-based navigation.[25] The benefit of this novel technique is its ability to spare the motion segments from fusion. However, this procedure is deemed unfeasible for present case because of the presence of huge gap as well as well-defined corticated margins of the pillar fragments at the spondylolytic defect, both of which would hinder screw placement and bony fusion. The reasons why we decided to perform a C2/3 ACDF were: exclusion of C1 from the fusion would allow the preservation of rotational movement at the atlanto-axial joint, dysgenesis of C2 pedicles and C3 lateral masses would prevent us from achieving rigid C1/3 fusion, mild C2/3 spondylolisthesis, and absence of soft tissue swelling associated with acute trauma in the anterior neck encouraged us to adopt anterior approach. Furthermore, biomechanical study showed C2/3 ACDF for C2 spondylolysis was adequate in restoring the stiffness of specimens in the anterior-posterior movement and they concluded that C2/3 ACDF was a biomechanically viable surgical option for unstable C2 spondylolysis.[2] However, this high cervical approach carries the inherent risk of injury to the marginal mandibular, hypoglossal, and superior laryngeal nerves.[26] The blunt dissection through soft tissues, staying more than 2 cm away below the mandibular rim and the submandibular salivary gland, and the use of facial nerve monitoring can reduce the incidence of complications related to the marginal branch of the facial nerve. Retraction should be loosened intermittently and surgical time should be shortened to protect from hypoglossal nerve injury. We should realize that the superior laryngeal nerves lie directly deep to the loop of superior

thyroid artery and the courses are almost identical and constant.[26)]

Conclusion

C2 spondylolysis is very uncommon with limited cases. First, we should tell this pathology from hangman's fracture based on radiographic studies as well as adequate clinical history. Second, spinal instability should be noted and assessed on flexion-extension x-rays after ruling out acute trauma. Third, symptomatic patients with instability and/or spinal cord compression as well as patients with progressive instability should be considered as surgical candidates. Patients of C2 spondylolysis even without instability have to be followed up carefully keeping close attention to progression of instability due to vulnerability to minor trauma.

Acknowledgments

Conflicts of Interest Disclosure

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