Traumatic, high-cervical, coronal-plane spondyloptosis with unilateral vertebral artery occlusion: treatment using a prophylactic arterial bypass graft, open reduction, and instrumented segmental fusion

Case report

SUNIL MANJILA, M.D., SHAKEEL A. CHOWDHRY, M.D., NICHOLAS C. BAMBAKIDIS, M.D., AND DAVID J. HART, M.D.

Department of Neurological Surgery, University Hospitals Case Medical Center, Cleveland, Ohio

The authors present a case of traumatic, complete, high cervical spine injury in a patient with gradual worsening deformity and neck pain while in rigid cervical collar immobilization, ultimately resulting in coronal-plane spondyloptosis. Due to the extent of lateral displacement of the spinal elements, preoperative evaluation included catheter angiography, which revealed complete right vertebral artery (VA) occlusion. A prophylactic arterial bypass graft from the right occipital artery to the extradural right VA was fashioned to augment posterior circulation blood supply prior to reduction and circumferential instrumented fusion. Following surgery, the patient was able to participate in an aggressive rehabilitation program allowing early mobilization, and he ceased to be ventilator-dependent following implantation of a diaphragmatic pacer. The authors review factors leading to progression of this type of injury and suggest technical pearls as well as highlight specific management pitfalls, including operative risks.

Key Words • cervical spondyloptosis • arterial bypass grafting • cervical spine fusion • hypoplastic vertebral artery • fracture dislocation of cervical spine

Abbreviation used in this paper: VA = vertebral artery.

LATERAL, or coronal plane, spondyloptosis occurring in the high cervical spine is rarely reported in the neurosurgical literature. The clinical symptoms resulting from this pathological process can vary, but often comprise a myeloradiculopathy with or without vertebral artery (VA) compromise and respiratory distress from phrenic neuropathy. Spondyloptosis in the cervical spine occurs rather infrequently compared with the thoracolumbar spine, with the latter occurring more often in the sagittal plane. Spondyloptosis most often results from trauma but may also occur in the setting of spondyloarthropathies or degenerative disease. Coronal spondyloptosis of the thoracolumbar spine, unlike sagittal spondyloptosis, may be associated with an incomplete neurological injury.

The authors believe that management of high-cervical, coronal-plane spondyloptosis presents a unique challenge in comparison with the thoracolumbar variant for 3 key reasons: 1) the limited number of spinal levels available above the injury for purchase during reduction and fixation; 2) the physiological lordosis of the cervical spine and the importance of its restoration for a positive long-term outcome; and 3) the potential for harm to the VA, both at the time of initial injury and during operative reduction and stabilization. Both the biomechanics of the cervical spine and the relationship of the VA to the ptotic spinal segment contribute to the technical challenge of surgical treatment. We report a case of upper cervical spine, coronal-plane spondyloptosis managed with the use of an arterial graft followed by open reduction and circumferential instrumented fusion.

Case Report

History and Examination. This patient was a 39-year-old man who suffered a severe, high cervical spinal cord injury following a motorcycle accident. He was found unresponsive, without a helmet, and initially in asystole. He was intubated at the scene, resuscitated, and then transferred to an outside hospital, where bilateral thoracostomy tubes were placed and blood transfusions were administered. He was hypotensive with agonal respirations and
flickering movement of only his left arm prior to transfer to a Level I trauma center. Hemoperitoneum prompted an emergency exploratory laparotomy. No frank visceral injury was identified. In the subsequent 4-week intensive care unit stay, the patient developed refractory bradycardia and occasional asystole due to his high spinal cord lesion despite therapeutic levels of theophylline. A cardiac pacemaker was then implanted. As detailed below, he had sustained multiple cervical spine fractures, which were nonsurgically managed with a Philadelphia cervical collar (Philadelphia Cervical Collar Co.), as well as occlusion of the right VA. When the patient was eventually transferred to a neurorehabilitation facility, he remained a ventilator-dependent tetraparetic patient (American Spinal Injury Association [ASIA] scale Grade A), with only minimal movement of his left arm.

The initial CT scan of his cervical spine revealed a fracture of the right C-2 pedicle with a fracture along the spinolaminar line of C-2 extending into the right transverse foramen. A fracture of the superior articular process of C-3 extending into the left C-3 transverse foramen was also noted. Additional fractures along the lamina of C-6 on the left extending along the spinolaminar line and bilateral C-7 lamina were identified. The first rib on the right was fractured, along with multiple left rib fractures, and the thoracic T-3 vertebral body fracture was noted without significant retropulsion. Computed tomography angiography depicted right VA occlusion and a patent left VA. Magnetic resonance imaging of the cervical spine (Fig. 1) showed evidence of an acute hemorrhagic contusion within the cervical spinal cord at the C2–3 level, measuring approximately 1.5 cm in length. Extensive cord edema spanned from the cervicomedullary junction to the level of the C4–5 interspace. Edema within the C2–3 intervertebral disc was observed along with ventral edema associated with disruption of the anterior longitudinal ligament. Edema or disruption of the posterior longitudinal ligament was not identified. There was at least partial disruption of the interspinous ligament at the C2–3 level.

Over the next several months, the patient developed a progressive deformity of his neck with a painful head tilt, rendering him unable to participate in rehabilitation. Imaging studies obtained 6 months after the injury revealed a complete coronal-plane spondylolisthesis of C-2 over C-3, with radiographic suggestion of some degree of autofusion of the vertebral bodies and facet joints at C2–3. Accordingly, the patient’s spine failed to reduce in traction (Figs. 2 and 3). A combined anterior and posterior approach with ventral and dorsal osteotomies, realignment, and circumferential spinal stabilization was planned to correct this deformity, with the goal of reducing the patient’s severe pain and restoring his normal alignment to maximize this highly motivated patient’s potential for improvement with intensive neurorehabilitation.

Operative Planning. Repeat CT angiography (Fig. 4) and catheter angiography confirmed right VA occlusion with retrograde filling of the right VA segment and posterior inferior cerebellar artery. Posterior communicating arteries were not visualized. The patent left VA was believed to be at risk for injury during the planned procedure for 2 reasons: proximity to the partially autofused C2–3 vertebral bodies, and surgical realignment. The proximity to a large, irregular fusion mass in the setting of spondylolisthesis increased the risk of injury during ventral and dorsal osteotomies. The open reduction introduced a risk of shear injury or kinking if the vessel was tethered along its course. Unroofing of the transverse foramen has been previously described to prevent VA kinking during correction of cervical deformity. In our case, we believed that the significant anatomical aberration and partial fusion contributed to a relatively high risk of iatrogenic VA injury with unroofing of the VA. Artery-to-artery bypass procedures are generally well tolerated, but are not without risk. A prophylactic right occipital artery to extradural right VA bypass surgery was planned. If the left VA was significantly compromised during the procedure, the
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risk of devastating posterior circulation infarction would be remarkably high. In this scenario, a prophylactic right VA bypass would provide critical flow to the posterior circulation. If the left VA remained intact, then absence of demand coupled with robust retrograde filling of the right VA would contribute to stasis within what would now be a noncritical bypass. The surgery was staged over 2 days.

Operative Course

Stage I. The patient was brought to the operating room for Stage I of the operation and positioned prone with a halo ring secured to a Jackson table. A traditional hockey-stick incision was fashioned extending down to the level of C-5. The occipital artery was identified and dissected free in routine fashion with a periadventitial cuff of tissue. The right VA was identified above the C-1 lamina on the right side. Temporary clips were placed on the VA, and an extradural vertebral arteriotomy was performed with a no. 11 blade; excellent back-bleeding under high pressure was observed. The occipital artery was divided, prepared in routine fashion, and rotated into the field. An end-to-side anastomosis to the right VA was fashioned with 9-0 Prolene polypropylene sutures (Ethicon Inc.). Graft patency was confirmed with microdoppler ultrasonography and indocyanine green angiography.

The lateral masses of C-4 and C-5 were identified and instrumented using standard lateral mass screws bilaterally. During the dissection, the posterior elements of C-2 had subluxed to the left and telescoped caudally over C-3. The result was complete “unroofing” of the right C-3 lateral mass and a “naked” right C-3 superior articular process, which afforded easy placement of a lateral mass screw into the right C-3 lateral mass. However, the vertebral translation resulted in the lamina and lateral mass of C-2 being overlapped and obscured nearly entirely by the C-3 lateral mass on the left. Obtaining visualization to allow for pars screw placement at C-2 on the right necessitated

Fig. 2. Preoperative posteroanterior (left) and lateral (right) plain radiographs following attempted traction.

Fig. 3. Reconstructed CT scans showing cervical spondyloptosis. A: Coronal image depicting the coronal-plane spondyloptosis at C2–3. The associated leftward neck angulation and soft tissue displacement are also appreciated. B–D: Sagittal CT reconstructed views of the right side of the injury (B), the midline (C), and the left side (D) demonstrating the lateral displacement and posterior column fractures.

Fig. 4. Computed tomography angiography (A) demonstrating C2–3 coronal-plane spondyloptosis with patency of the left VA and lack of opacification in the transverse foramen on the right. On the right VA injection angiogram with anteroposterior projection over the neck (B), the right VA fills slowly to the level of C-3 and supplies numerous muscular branches. No filling is observed beyond C2–3. On the left VA injection angiogram with anteroposterior projection over the neck and head (C), the left VA is widely patent and supplies the posterior circulation. Flow across the vertebrobasilar junction is appreciated with retrograde filling of the distal right VA and opacification of the right posterior inferior cerebellar artery.
resection of the right lateral mass of C-3. Before drilling was performed, a midline laminectomy of C2–3 was conducted. There was obvious gross evidence of prior injury to the spinal cord, with areas of scarred arachnoid mater visible through a thin, translucent pseudodura membrane. No CSF leakage was observed; the pseudodura was pulsatile and stained with hemosiderin. After the laminectomy, complete bilateral osteotomies were performed through the C2–3 facet joints. Bilateral C-2 pars screws were then placed. The position of all 7 posterior screws was verified using a 3D isocentric fluoroscopic unit (Siremobil Iso-C 3D; Siemens Medical Solutions). The portion of the surgical wound overlaying the right posterior fossa was closed in layers, and the midline wound was closed with a running Prolene stitch over the unconnected hardware. A drain was left in the wound, and the patient was returned to the neuroscience intensive care unit and maintained in low-weight cervical halo traction until the following morning.

Stage II. In the second stage, the patient was positioned supine on a flat Jackson table. A standard anterior cervical approach was performed through a horizontal incision. The C-2 vertebral body, C-3 vertebral body, and fusion mass between these vertebral bodies were exposed. The superior endplate of C-3 and the inferior endplate of C-2 were adequately visualized. The left VA was identified, carefully protected, and dissected away from the fusion mass. Caspar distraction posts were fixed into the C-2 and C-3 vertebral bodies. A Midas Rex Legend Gold high-speed pneumatic drill (Medtronic Inc.) was used to drill through the fusion mass, essentially disconnecting the 2 vertebrae from each other. Distractive force was applied by manual traction on the patient’s halo ring and on the Caspar distraction posts. Over 30 minutes of incrementally increasing force (distraction and lateral pressure) was applied, and the deformity was partially reduced. The authors noted that a relatively higher amount of force than anticipated was required, possibly due to chronic ligament and muscle contractures. It was believed that an approximately 70% realignment of the inferior C-2 endplate relative to the superior C-3 endplate was obtained. These endplates were then prepared in routine fashion. An iliac crest tricortical wedge spacer (9 mm in height) was then placed into the interspace. During the realignment, as the vertebral bodies separated and began to translate coronally, copious CSF leakage and bleeding from the epidural venous plexus were simultaneously encountered. No arterial bleeding was appreciated, and the left VA appeared intact. Once the epidural bleeding was controlled, the CSF leak was managed with application of Duragen (Integra LifeSciences Corp.) and Tisseel fibrin glue (Baxter) as well as immediate postoperative lumbar subarachnoid drain placement. Reduction forces were maintained using Caspar posts while an anterior cervical plate (Aesculap Inc.) was applied to C-2 and C-3. Due to the residual dislocation, only a single screw could be placed in solid bone at C-3, which proved to be sufficient as neither migration nor loss of reduction was observed once the force on the Caspar distraction posts was released. Adequate placement of the graft and plate was confirmed with intraoperative fluoroscopic imaging.

Following closure of the anterior cervical wound, the patient was rotated prone. On visual inspection, the sagittal and coronal alignment of the patient’s head and neck had remarkably improved. The midline incision from the Stage I surgery was reopened, and all 7 screws were identified. The posterior elements were cleared of soft tissue and decorticated using a Midas Rex drill. The alignment of the C-2 pars screws was improved in the sagittal, coronal, and axial planes, facilitating connection of the rods. The salvaged bone from the patient’s previous laminectomy was rinsed and laid as morcellized autograft across C-2 and C-3. A Vitoss bone graft substitute (Orthovita) was packed over the remaining lamina, lateral masses, and facet joints bilaterally from C-2 to C-5. Final fluoroscopic imaging depicted satisfactory placement of hardware and marked interval correction of the deformity (Fig. 5).

Postoperative Course. At the most recent follow-up 22 months after the procedure, the patient reported excellent pain control, rating his pain no higher than 2 out of 10 on a visual analog scale (compared with 9 out of 10 preoperatively). He remains tetraparetic with minimal movement of his upper extremities and patchy sensation below his level of injury at C-3 (Nurick Grade 5), but he is now able to actively participate in his rehabilitation program, which was impossible for him before surgery due to severe pain. He received a diaphragmatic pacemaker, which greatly affected his quality of life; he is now able to sit up with support. A CT scan of the cervical spine at discharge and at the 1-year follow-up revealed evidence of progressive circumferential fusion at the C2–3 level (Fig. 6).

Discussion

The authors present the first reported case of traumatic, high-cervical, delayed coronal-plane spondyloptosis with unilateral VA occlusion. Generally, in the case of complete neurological injuries, the role of instrumented spinal fusion is to restore stability and maintain normal spinal alignment, allowing early mobilization and improved pulmonary toilet. In patients with an incomplete neurological injury, decompression and stabilization may contribute to earlier neurological recovery. The authors present a case in which the patient had developed a severe painful neck deformity with a tilt that negatively affected his quality of life and limited his ability to participate in rehabilitation. Beyond the technical aspects of realignment of high-cervical spondyloptosis, this case report highlights the significance of adequate acute stage immobilization in cervical spine trauma in preventing delayed deformities. Proper recognition of the extent of injury and anticipation of detrimental aftereffects may have prompted surgical instrumentation or halo vest orthosis in the early period, thereby preventing the need for the extensive surgery that ensued later. The delayed recognition of the deformity allowed for formation of a pseudarthrosis, similar to that noted in Charcot’s arthropathy.

This unique clinical picture draws a close parallel to Charcot’s arthropathy, also called spinal neuropathic or neurogenic arthropathy. Occurring several years after spinal trauma, Charcot’s arthropathy is a destructive condition that affects the intervertebral disc and adjacent vertebral bodies and is treated surgically by decompression.
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Fig. 5. An immediate postoperative lateral plain radiograph (A) demonstrating improvement in alignment and restoration of height following reduction and circumferential instrumentation and fusion. Posteroanterior (B) and lateral (C) plain radiographs obtained 2.5 months later revealing maintenance of correction with persistent but much improved head deviation to the left.

Fig. 6. Reconstructed sagittal (A and B) and coronal (C and D) CT scans of the cervical spine obtained immediately (left column) and 1 year (right column) after the surgery showing evidence of a developing fusion mass at the operated levels.

and circumferential fusion. In our case, the clinical and radiological profile of deformity suggests a faster evolution compared with what is described in typical Charcot’s joints. The indication for surgery remained the same as in Charcot’s joints: severity of functional limitation, disabling pain, and lack of response to conservative treatment. The present case report differed widely in pathology from that of ankylosing spondylitis (seronegative, progressive, systemic, inflammatory, rheumatic spondyloarthropathy) in which the deformity is more rigid, mostly kyphotic, and has a different natural history of progressive osteoporosis and fractures that occur typically at the level of intervertebral discs. Charcot’s arthropathy with a coronal spondyloptotic deformity or vascular compression has not been reported since Neugebauer first described spondyloptosis in 1882, the term spondyloptosis is routinely used to describe subluxation greater than 100% at any spinal level. Cervical spondyloptosis is relatively rare, and most of the relevant literature is in the form of case reports or short case series. Of note, cervical spondyloptosis has been reported in patients without neurological deficits. Nontraumatic cervical spondyloptosis has been reported in the setting of aneurysmal bone cyst, neurofibromatosis, ankylosing spondylitis, or congenital absence of posterior elements of the cervical spine. The first documented case of traumatic cervical spondyloptosis was of an 8-year-old girl treated with C5–7 corpectomies with in situ fusion using a tricorticate iliac bone graft from C4–T1. Amacher reported the case of a 7-year-old boy with lower cervical spondyloptosis (C-7 on T-1) treated with C7–T2 corpectomies and posterior C6–T3 fusion using bone and wires, without an anterior bone graft. Jayakumar and colleagues described late presentation (4 years after initial injury) of a Type III axis fracture with a painful spondyloptosis that was treated with a single-stage in situ posterior fixation. Shah and Rajshekhar reported successful management of a posttraumatic C7–T1 spondyloptosis with an uninstrumented ventral surgery. However, none of these reports were of coronal spondyloptosis or with concomitant unilateral VA occlusion.

In our case, the original injury to the spinal elements was extensive, compromising multiple bone and ligamentous structures, despite initially adequate alignment. Mechanically, a coronal spondyloptosis, or complete lateral displacement of the cervical vertebral body, is likely to result from a shear-type mechanism with a coronally or laterally directed vector force. A rotatory component is rare but is more likely to be found in cervical spine trauma as opposed to the thoracolumbar region, where movement is restricted by the ribcage itself. Direct or indirect involvement of the VA adds to the complexity of treatment, especially in the upper cervical spine, where fixation points are fewer and the anatomy is more complex.

Role of Prophylactic Arterial Grafting

Tumialán and Theodore have recently published a re-
Vertebral artery injury is a rare but well recognized catastrophic iatrogenic complication of cervical spine surgery with a reported incidence of 0.3%–0.5%. In the case presented in this report, the left VA was at risk during the exposure, osteotomies, and bone reduction for 2 reasons: 1) proximity of the artery to the partially autofused C2–3 vertebral bodies with potential for injury during osteotomy; and 2) anticipated significant alteration in its course that would occur with successful realignment, placing the vessel at risk for shear injury or kinking during translation. Unroofing of the transverse foramen has been previously described to prevent VA kinking during correction of cervical deformity. In our case, we believed that anatomical malalignment and partial fusion would contribute to a relatively high risk of arterial injury if unroofing of the transverse foramen was attempted. Thus, a prophylactic right occipital artery bypass was performed to the right VA, which was occluded in the V2 segment secondary to the patient's injury. Iatrogenic VA injury during cervical spine surgery has been documented to produce fistulas, late hemorrhages, pseudoaneurysm, thrombosis, and death. Common techniques to reduce the risk of VA injury include the following: 1) partial excision of the longus colli muscle, allowing exposure of the uncinate processes and transverse processes; 2) careful uncectomy and removal of lateral osteophytes followed by uncovertebral joint resection up to the depth of the floor of the transverse foramen cephalad and caudal to the transverse process; 3) exposure large enough for proximal and distal control of the VA should inadvertent arterial injury be encountered while working on the fusion mass; and 4) use of neuronavigation and Doppler probe ultrasonography to more clearly identify the vessel near the fusion mass.

The patient's cerebrovascular anatomy should be fully assessed in high cervical spine trauma, especially with compromised flow in the VA. If a hypoplastic VA is compromised, the risk of posterior fossa ischemia is less than a situation in which a dominant VA is compromised. If the patient lacks sufficient anterior to posterior circulation collaterals to allow for retrograde filling of the vertebrobasilar system (fetal posterior cerebral arteries), the risk of catastrophic injury with VA injury increases. One potential nonbypass option is cervical osteotomy and circumferential fusion with bilateral transverse foraminoctomy in chin-on-chest deformity (sagittal spondyloptosis) to prevent VA kinking. However, in our patient the right VA was already completely occluded, and the biomechanics and shear forces involved in correcting a coronal spondyloptosis are not the same as those for correction of a sagittal chin-on-chest deformity. Vascular grafting should be strongly considered in situations in which the contralateral VA is hypoplastic and the ipsilateral VA is occluded or injured. In the trauma literature, there are several reported cases of symptomatic vertebral and basilar artery thrombosis, with and without surgical reduction of cervical spine injury. Once a patient develops a symptomatic basilar artery occlusion, the prognosis is generally ominous, although a case report of successful mechanical and pharmacological thrombolysis does exist in the literature.

**Surgical Timing**

The indication and timing of surgical intervention in a quadriparetic patient with partial fusion but radiological progression of disease is certainly open to some debate. Appropriate cervical immobilization after trauma can prevent further neurological deterioration and, in this case, might have prevented the development of such a complex chronic deformity mandating an extensive corrective surgical procedure as described. Equally important is the caution that needs to be exercised while operating on an injured cervical spine with a unilateral VA occlusion. We hypothesize that initial stabilization with a halo vest orthosis might have prevented this outcome, and even if the halo had failed, at that early stage the patient likely would have been adequately stabilized with a simple posterior instrumented fusion.

Other potential issues of concern regarding our surgical treatment include the choice of fusion levels from C-2 to C-5. Upon observing the failure of the patient’s spine to reduce in traction, we anticipated potentially high forces resisting correction and maintenance of correction and believed that long-segment posterior stabilization would be desirable. However, we believed that, in this quadriplegic patient, maintaining his occiput–C1 and C1–2 mobility would have a significant impact on his remaining ability to interact with his environment and, therefore, should be preserved. Contrarily, fusing the C4–5 level as opposed to stopping at C-4 would cost him little in terms of range of motion. The decision to perform decompression, an osteotomy, and place posterior screws during the first day’s procedure was merely an attempt to provide comparable, optimized surgical and anesthesia times for both days as it was anticipated that performing only the arterial bypass on Day 1 and all of the deformity correction, decompression, and anterior/posterior hardware placement and fusion on Day 2 might be excessively strenuous for the patient during a second anesthesia induction.

**Conclusions**

Posttraumatic delayed coronal-plane cervical spondyloptosis is an extremely rare phenomenon, and surgical correction may be mandated depending on clinical presentation. The proximity of the VA to the anatomical distortion in coronal-plane cervical spondyloptosis makes this condition uniquely hazardous to treat. An understanding of the complex biomechanical forces that produce this injury is imperative to determining the steps required to reduce and stabilize it. We recommend a vascular study of the vertebrobasilar circulation as part of the preoperative evaluation because unrecognized compromise of 1 VA may lead to fatal and/or debilitating deficits if the single patent VA is damaged. This case brings to light the tem-
poral progression of high-cervical, coronal-plane spondyloptosis and highlights the importance of adequate early rigid immobilization.

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Disclosure

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 Address correspondence to: David Hart, M.D., University Hospitals Case Medical Center, 11100 Euclid Ave., Cleveland, OH 44106.
 email: david.hart@uhhospitals.org.