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Brown-Sequard syndrome caused by hyperextension in a patient with atlantoaxial subluxation due to an os odontoideum

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ABSTRACT

Brown-Sequard syndrome is an uncommon complication of atlantoaxial arthrodesis. A 50-year-old female visited our emergency department after falling from a ladder. Radiologic evaluations revealed chronic C1-2 instability with acute spinal cord injury. The day after atlantoaxial fusion was performed, she developed left-sided motor weakness and the loss of right-sided pain and temperature sensation. Based on physical examination and radiologic findings, we diagnosed her as having Brown-Sequard syndrome. Spine surgeons performing this procedure should therefore consider Brown-Sequard syndrome if a patient displays signs of postoperative hemiplegia.

Introduction

Posterior instrumentation of the C1–C2 vertebrae using screws has been used to treat various pathologies of the upper cervical spine since the 1980s. Due to the peculiar anatomy of the C1 vertebra, spine surgeons should be mindful of potentially severe and life-threatening complications, such as neurologic injury, hydrocephalus due to intraventricular hemorrhage, and vertebral artery injury (VAI).[1–3] In this case report, we describe a rare case of hemiplegia (Brown-Sequard syndrome) following atlantoaxial arthrodesis for the treatment of chronic atlantoaxial instability. Because this is such a rare complication, we have also included an auxiliary review of the literature.

Case report

A 50-year-old female visited our emergency department after falling from a ladder. She presented with neck pain and non-specific causalgia of both the upper extremities; however, neither motor weakness nor reduced sensation were observed during the initial exam. Before visiting our institution, she was fully independent with no history of medical problems or prior trauma.

Radiographs of the cervical spine revealed a type II odontoid process fracture (Fig. 1). There was no vertebral anomaly. She was initially managed conservatively with a Philadelphia neck collar. The CT scan revealed bone resorption with sclerotic changes at the odontoid fracture site; we diagnosed this as os odontoideum. Os odontoideum is an anatomic variant of the odontoid process of C2 that can be associated with atlantoaxial instability; it must be differentiated from persistent ossiculum terminale or an odontoid fracture. The etiology of os odontoideum in this case was unclear; however, we assumed it as a congenital lesion due to the lack of prior cervical trauma history. We also carefully conducted a flexion/extension dynamic CT scan. The space available for the spinal cord was measured and C1-2 instability was confirmed (Fig. 2). The Power's ratio of this patient was 0.9, which would indicate that there was no atlanto-occipital instability. In order to evaluate the patient's neurologic symptoms (causalgia), the patient underwent MRI, which revealed acute spinal cord injury (SCI) at the C1-2 level (Fig. 3). Thus, we finally diagnosed chronic C1-2 instability with acute SCI and planned to perform a C1-2 arthrodesis with screws and rods. Using Harms technique, which has been previously well

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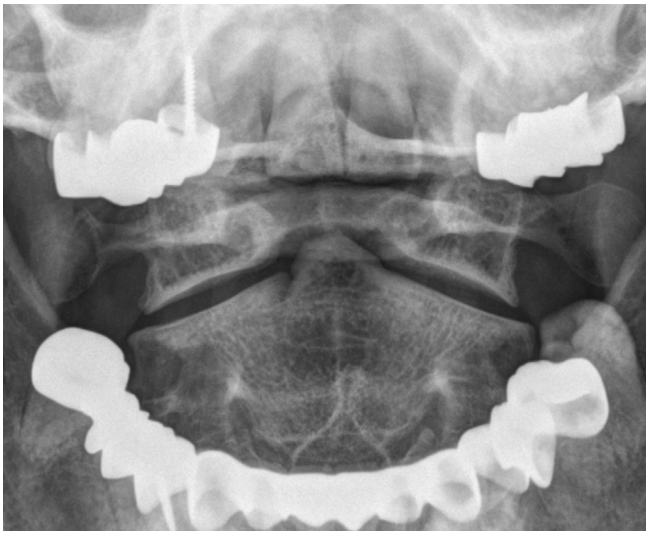

Fig. 1. Radiographs of the cervical spine revealing a type II odontoid process fracture.

documented,[4] bilateral C1 lateral mass (LM) screws and C2 pedicle screws were inserted under fluoroscopic guidance. Although there was no repetitive screw insertion, we had to push the hand drill and the screw insertion by force due to the hardness of the sclerotic bone. After screw fixation, under anteroposterior and lateral fluoroscopic guidance, we determined that the position of all 4 screws as acceptable. Next, to strengthen the C1-2 fusion and stability, sublaminar wiring with auto-iliac strut bone graft was applied to the posterior aspect of the C1-2 lamina (Gallie method).[5] No cerebrospinal fluid (CSF) leakage or active bleeding was noted.

On the postoperative day, the patient presented with left-sided motor weakness and the key muscles of the left upper and lower extremities were grade zero (hemiplegia); motor power on the right side was normal. We first assumed that her hemiplegia was due to either a postoperative cerebral infarction or VAI from the screw insertion. A brain MRI and cervical spine CT with vertebral artery angiography were emergently performed, and acute brain damage and VAI were ruled out; however, our initial observation was that the position of the left C1 LM screw was somewhat abnormal. The screw was located in the C1 LM and less than 50% of the diameter of the screw violated the surrounding cortex, therefore, the placement was deemed acceptable (Fig. 4). To rule out other diseases, we thoroughly repeated the physical examination and discovered findings that had been initially missed. During the first physical examination, we only checked touch sensation, which was normal bilaterally; however, the patient could not feel pain and temperature sensation on her right-sided extremities. Although the left C1 LM screw position was acceptable based on radiologic images, the patient underwent MRI of the cervical spine to evaluate the possibility of SCI. The MR images revealed a definite high signal change of the left hemisection of the spinal cord (Fig. 5), and we ultimately made the diagnosis of Brown-Sequard syndrome (BSS). There was no evidence of CSF leakage and all screw positions were acceptable, effectively ruling out SCI from screws. We postulated that applying a pushing force from the posterolateral direction while drilling for screw fixation caused hyperextension combined with lateral bending of the neck. On postoperative day 3, motor power of the large joints (shoulder, elbow, hip, and knee) had somewhat improved; one week after surgery, the patient's hand grip and toe movement had recovered to grade 3. Three weeks after surgery, she was ambulating on her own and was discharged. At her 8-month follow-up, motor power of the hip, knee, ankle, shoulder, elbow, and wrist had recovered to grade 5; however, motor power of the fingers and toes remained grade 4. Additionally, right-sided pain and temperature sensation remained unchanged from her initial state.

Discussion

Atlantoaxial arthrodesis widely used to treat C1-2 pathologies. Despite many reports that the procedure is safe with excellent rates of stabilization and arthrodesis,[6] technical errors during the procedure can lead to devastating results such as VAI, neurologic injury, or even death. In a meta-analysis concerning C1-2 fusion with screw-rod constructs including 24 studies with 1073 patients treated with this technique, Elliot et al[7] reported that the incidence of clinically significant screw malposition was 2.4%; neurologic injury occurred in two patients from screw malposition (0.2%), and the incidence of VAI was 2.0%. Although BSS is an extremely rare complication after screw insertion, here we describe a case of hemiplegia caused by forceful management of the injury site.

The safety and applicability of these techniques have been established in several series of patients with a variety of pathologic entities. Since technical errors at the level of the upper cervical spine can lead to catastrophic consequences, Bransford et al[8] investigated the effect of screw fixation on perioperative complications such as neurologic or vascular injury. They reported a 0% incidence of VAI and neurologic injury despite a 4% incidence of

Fig. 2. Sagittal CT scan of the cervical spine showing nonunion of the odontoid process (Os Odontoideum) (A). Flexion/extension dynamic CT scans showing chronic C1-2 instability (B, C). Figures of each image indicate the amount of space available for the spinal cord.

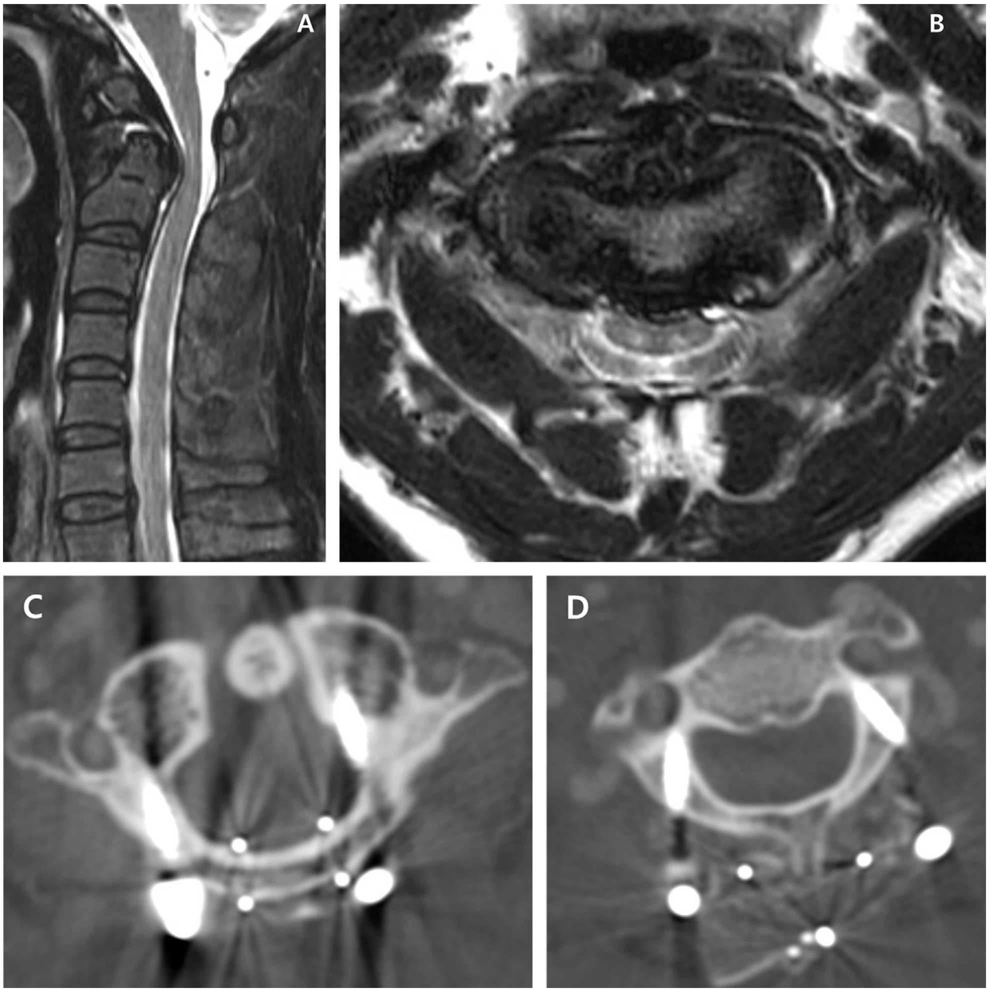

Fig. 3. Sagittal (A) and axial (B) MR images showing nonunion of the odontoid process fracture and spinal cord compression by the vertebral body due to chronic C1-2 instability. Spinal cord signal change indicates acute spinal cord injury.

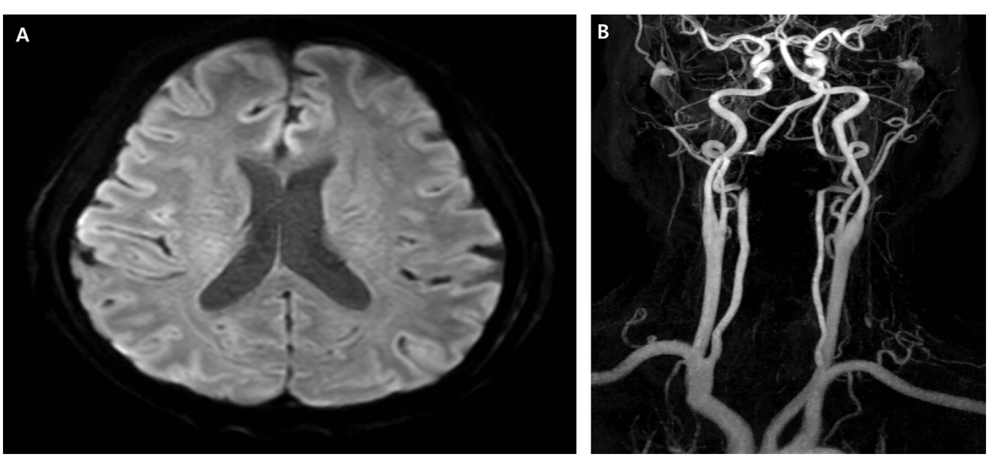

Fig. 4. Brain and vertebral artery were intact. (A) Diffusion weighted MR image, and (B) vertebral artery angiographic MR image. The left C1 lateral mass screw is observed within the C1 lateral mass where less than 50% of the diameter of the screw violates the surrounding cortex (C). Both C2 pedicle screws are located completely within the bony cortex (D).

unacceptable screw placement. In addition, Harms and Melcher,[4] Vilela et al,[9] and Gunnarsson et al[10] reported series of patients who underwent C1 LM screw placement in the setting of C1-2 fixation and reported no incidents of iatrogenic neurological or vascular injury. Despite the small size of these studies, these results suggest that C1-2 screw fixation can be safely performed with a low risk of perioperative complications.

Various reported complications of C1-2 screw insertion should be considered intraoperatively and postoperatively. The most common complication is C2 nerve dysfunction due to screw irritation, which

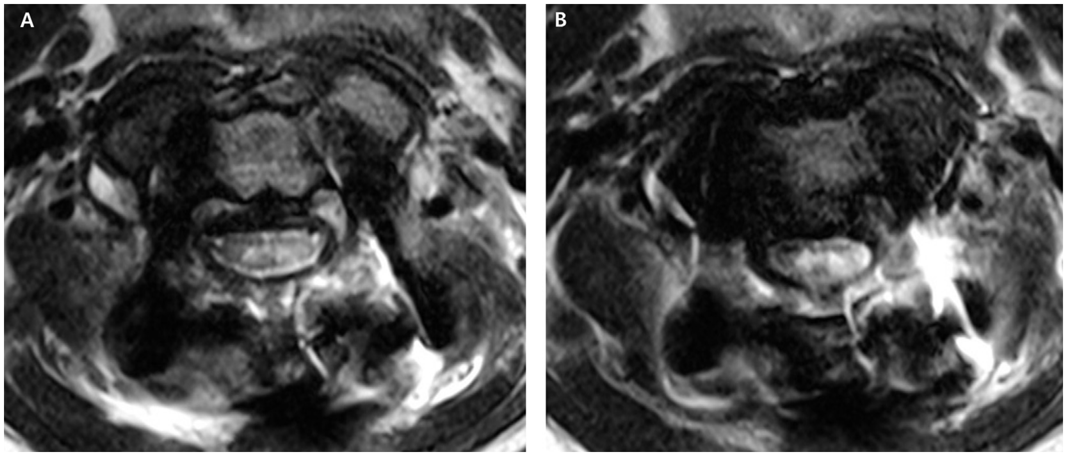

Fig. 5. MR performed 3 days after surgery: axial MR image showing definite high signal change in the left hemisection of the spinal cord (A: C1 level, B: C1-2 disc level).

causes occipital neuralgia, occipital numbness, and neuropathic pain.[2] In addition, excessively long screws may damage the internal carotid artery and hypoglossal nerve. Also a lateral trajectory of the screws may cause VAI and a medial trajectory can cause a dural tear, leading to CSF leakage or SCI. A very low incidence of acute hydrocephalus due to subarachnoid hemorrhage and venous embolism has also been reported as a complication of this procedure.[1,11] Although there have been many clinical trials evaluating complications of atlantoaxial arthrodesis, BSS remains very rare. Here we have described a case of this rare complication in order to increase awareness of BSS in patients who shows signs of postoperative hemiplegia.

Brown-Sequard syndrome presents with ipsilateral motor weakness and contralateral loss of pain and temperature sensation; however, spasticity and hyperactive reflexes may not be present with an acute lesion.[8] The incidence of BSS is rare and can be caused by cervical trauma such as lamina and pedicle fracture, disc herniation, penetrating or stab injury, and rotational injury resulting in a subluxation. Prognosis of BSS recovery is generally good, and Pollard and Apple[11] reported that a significant number of patients with BSS improved within 2 years of the injury. In the present study, we described BSS after C1-2 screw fixation. We assumed that SCI was not caused by direct trauma due to the screw, because there was no evidence of CSF leakage. Instead, we assumed that while drilling the lateral mass or pedicle, the pushing force caused neck hyperextension and the posterolateral force caused lateral bending of the neck, resulting in BSS.

After making the diagnosis of BSS, we considered both revision surgery and conservative treatment. A literature review revealed the dimensions were greatest at the atlas (C1); a screw position where less than 50% of the diameter of the screw violates the surrounding cortex and which protrudes less than 1 mm form the anterior cortex is considered acceptable.[6] Furthermore, in terms of SCI, prognosis is mainly associated with the timing of the decompression. Early intervention helps minimize the secondary damage caused by compression of the spinal cord after trauma. In our case, we concluded that it was not possible to obtain additional spinal cord decompression, therefore, we planned for conservative treatment. If screws compressed the spinal cord, urgent surgical decompression would likely play an important role in neurologic recovery and the overall outcome. This patient was followed-up continuously until 8 months after the injury; the recovery was noted to be satisfactory with normal motor function and a normal gait pattern. However, pain and temperature sensation on right side did not completely recover.

Based on the literature, atlantoaxial arthrodesis using C1 LM is a safe and effective treatment option for atlantoaxial pathology. Although BSS is a very rare complication after C1-2 screw fixation, this diagnosis should be considered if patients present with postoperative hemiplegia. Furthermore, when drilling the LM or pedicle for screw fixation, surgeons should avoid forceful maneuvers. If a patient presents with postoperative hemiplegia and there is no evidence of VAI, CSF leakage from a dural tear, or unacceptable screw placement, conservative treatment may be sufficient.

Disclosures

The manuscript submitted does not contain information about medical device(s)/drug(s).

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