Posterior atlantoaxial subluxation due to os odontoideum combined with cervical spondylotic myelopathy : a case report

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Abstract In patients with os odontoideum and posterior atlantoaxial subluxation are extremely rare. No reports have described posterior atlantoaxial subluxation associated with os odontoideum combined with cervical spondylotic canal stenosis, both of which require surgical treatment. We report one case of a 75-year-old female who underwent arthrodesis between the occiput and C3 using a hook-and-rod system and also a double-door laminoplasty from levels C3 to C7. The claw mechanism was applied between the C2 lamina and the C3 inferior articular process. The posterior atlantoaxial subluxation was completely reduced by the method that the rod gradually pushed the posterior arch of C1 anteriorly during connection to the occiput. Twelve months after surgery, the patient showed improvement in preoperative clumsiness and gait disturbance, and the latest plain radiographs showed solid osseous fusion, with no loss of correction or instrumentation failure.

Keywords Posterior atlantoaxial subluxation ·
Os odontoideum · Cervical spondylotic myelopathy

Introduction

Although anterior atlantoaxial subluxation is a frequent complication of os odontoideum, posterior subluxation is extremely rare. No reports have described posterior atlantoaxial subluxation due to os odontoideum combined with spondylotic cervical myelopathy (CSM), both of which require surgical treatment. Here, we present a unique instance of a female who underwent occipito-cervical fusion using a hook-and-rod system for posterior atlantoaxial subluxation, and cervical laminoplasty for CSM.

Case report

A 75-year-old female experienced a gradual gait disturbance and began to use a T-cane in 2003. At the end of 2004, she noticed posterior neck pain and numbness in her right hand. The symptoms became worse in April 2005, and she also noticed numbness in her left hand. She experienced bilateral clumsiness and was diagnosed with posterior atlantoaxial subluxation due to os odontoideum combined with cervical spondylotic myelopathy in another hospital in October 2005. In November 2005, she visited our clinic and was admitted for surgical treatment.

The patient had undergone left mastectomy for breast cancer at the age of 58 years and right nephrectomy for a right pyelic malignant tumor at the age of 71 years. She had no past history of rheumatoid arthritis (RA) and no episode of traumatic injury of the head and/or neck. There were no particular disorders in her family.

Neurological examination at the time of admission revealed a spastic gait, and she required a T-cane for ambulation. She reported posterior neck pain with her neck in flexion-extension. Manual muscle testing revealed

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Fig. 1 **a** Preoperative plain radiograph in neutral position demonstrating os odontoideum and posterior atlantoaxial subluxation. **b** The subluxation is reduced with the neck in flexion

generalized muscle weakness (grade 4 of 5) throughout the upper extremities, but not in the lower extremities. Hypoesthesia was observed in the right C5-8 dermatome and the left C6 dermatome. The deep tendon reflexes in both the upper and lower extremities were increased, and scapulo-humeral reflex, Hoffman reflex, Tromner reflex, and Babinski reflex were positive. Bilateral clumsiness and difficulty in urinating were present, but dyschezia was not reported. She had no complaint of morning stiffness, and joint deformity was not observed. Laboratory data for both serum and urine, including serological test for rheumatoid factor, were within normal limits.

Serial plain radiographs and reconstruction computed tomography (CT) of the cervical spine showed os odontoideum and posterior atlantoaxial subluxation. The subluxation could be reduced with the neck in flexion (Figs. 1, 2). Magnetic resonance imaging (MRI) showed compression of the spinal cord at the C1/2 level between the anterior os odontoideum and the posterior C2 lamina and consecutive spinal canal stenosis from C3 to C7 levels (Fig. 3).

In December 2005, she underwent a decompression and fusion operation. Initially, a double-door laminoplasty from the C3 to C7 levels was performed using hydroxyapatite spacers inserted into each expanded lamina. Next, a reduction of the posterior atlantoaxial subluxation and arthrodesis between the occiput and C3 was performed using a hook-and-rod system. The claw mechanism was applied bilaterally between the hook of the C2 lamina and the hook of the C3 inferior articular process. During connection of the two rods to the occiput by using screws, the bilateral rods gradually pushed the posterior arch of C1 anteriorly to the anatomical position. We confirmed that the posterior surface of the dura mater was not compressed

Fig. 2 **a** Preoperative CT in flexion demonstrating reduction of the posterior subluxation (arrow). **b** A dislocated os odontoideum is observed in extension (arrow)

after this maneuver. Finally complete and adequate reduction of posterior atlantoaxial subluxation was obtained and confirmed by intraoperative X-ray picture. Autologous strut bone was harvested from the iliac crest and was grafted between the occiput and the C2 spinous

Fig. 3 MRI shows compression of the spinal cord at the C1/2 level between the anterior os odontoideum and the posterior C2 lamina, and consecutive spinal canal stenosis from C3 to C7 levels

process. The strut bone was securely fixed between the two rods using two sutures.

Two days after surgery, the patient was able to stand, and she attempted to ambulate using a walker. A soft neck collar was applied for 3 months. Twelve months after surgery, although slight muscle weakness was observed, the preoperative clumsiness and gait disturbance improved. Plain radiographs showed solid osseous fusion, with no loss of correction or instrumentation failure (Fig. 4).

Discussion

Most cases of atlantoaxial subluxation occur in patients with RA, and almost all are anterior subluxation. Posterior atlantoaxial subluxation is rather rare, but a small percent of patients with longstanding RA develop it [4, 9, 10, 12]. Atlantoaxial subluxation associated with os odontoideum is rare, and an associated posterior subluxation is rare [15]. Fielding et al. [3] demonstrated that posterior instability was observed in only 5 (14%) of 35 patients with os odontoideum. The main etiology of os odontoideum is the congenital anomalies of the odontoid process or trauma.

Instability secondary to hypermobile os odontoideum shows various clinical symptoms. Of the various symptoms, local neck pain is the most frequent and cervical myelopathy is the most serious. These symptoms are thought to be caused by the static compression of the spinal cord or repeated minor trauma to the spinal cord [15].

Operative stabilization is usually indicated when signs and symptoms of spinal cord compression and/or progressive myelopathy occur. However, the indication of operative treatment for patients without myelopathic deficits is controversial. Some authors have recommended an early operative fusion before any evidence of appreciable neural compression is found [13]. In planning the surgery, the area of decompression and fusion must be decided according to preoperative clinical symptoms and image findings. In the previous reports of posterior atlantoaxial subluxation due to os odontoideum, C1/2 fusion, occipito-C2 fusion, and occipito-C3 fusion had been performed [3, 15].

In our case, the patient had a positive scapulo-humeral reflex, and MRI demonstrated severe compression at the C1/2 level, indicating that decompression at this level was essential. Furthermore, since MRI also demonstrated compression at levels C3-7 and hypesthesia was observed in the dermatome of the lower cervical levels, we performed laminoplasty to decompress the spinal cord at those levels. To the best of our knowledge, this is the first case of occipito-C3 stabilization and laminoplasty for posterior atlantoaxial subluxation due to os odontoideum combined with cervical spondylotic myelopathy.

Shirasaki et al. [15] suggested the following surgical treatment for cervical myelopathy in os odontoideum with posterior instability. Atlantoaxial fusion in reduced position is indicated in the absence of canal stenosis at the C1/2 level, and laminectomy of the posterior arch of C1 followed by occipito-C2 arthrodesis is appropriate when associated with canal stenosis of the C1/2 level. However, in our case, spinal cord compression at the C1/2 level was caused by the os odontoideum and the C2 lamina, not by the posterior arch of C1. We considered that the posterior arch of C1 should be retained because the rods for occipito-C3 arthrodesis compress the posterior arch of C1 anteriorly to the anatomical position, so that posterior subluxation would be reduced. In fact, ideal reduction was achieved, and the retained posterior arch of C1 offered a favorable bony union bed for the grafted iliac bone. No previous reports have introduced this specific procedure, a reduction force of C1 lamina made posterior to anterior with bilateral rods. Recently, C1/2 transarticular screw fixation has been used for anterior atlantoaxial subluxation. However, in the procedure for posterior atlantoaxial subluxation, concomitant use of wires between C1/2 would pull the reduced posterior arch of C1 posteriorly again, therefore, we avoided these procedures.

Fig. 4 **a** Postoperative plain radiograph shows occipito-C3 fusion and double-door laminoplasty at C3–7 levels. **b** Lateral view of postoperative radiograph shows that subluxation was reduced and indicates solid osseous fusion

Occipito-cervical fusion with the hook-and-rod system is a safe and straightforward technique for posterior internal fixation of the cranio-cervical junction and the cervical spine. The lateral mass screw is associated with a risk of nerve root and vertebral artery damage [1, 8]. Heller et al. [6] reported immediate radicular symptoms in seven patients, cerebellar infarction in one and anterior horn infarction in one out of 78 patients that received posterior cervical plating with the attempted bicortical lateral mass screw procedure. In the pedicle screw system, the screws can be fixed securely to the vertebral body. However, there is also a possibility of injuring the nerve roots and the vertebral artery [16, 17]. The advantages of the hook-and-rod system, as compared to other techniques includes easy handling, a safe implantation, firm anchorage to the occipital bone, and an excellent postoperative stability. Several reports have described a high rate of successful osseous fusion [2, 5, 7, 11, 14]. On the other hand, one disadvantage of this system is the requirement of longer instrumentation because of the use of the claw configurations at two levels of fixation. In this case, the claw mechanism was applied bilaterally between the hook of the C2 lamina and the hook of the C3 inferior articular process. The spinal cord is sufficiently wide at C2 levels, indicating that there is less possibility of spinal cord compression with the use of hooks of the C2 lamina. We used pedicle hooks placed at the C3 inferior articular process. We consider this a safe procedure because the tips of the C3 hook are set outside of the spinal canal.

In this case, we preferred symmetrical double-door laminoplasty to asymmetrical open-door laminoplasty for decompression at C3–7 levels because bilateral rods for

occipito-cervical fusion can easily be set in the former procedure [7]. The C3 lamina was expanded and therefore, the claw mechanism was applied between the pedicle hook at the C3 inferior articular process and the C2 lamina hook. We considered that this procedure was safe because the tips of the C3 hook were set outside of the spinal canal. Instrumentation failure was not seen at the time of latest follow-up. However, careful follow up is required.

Conflict of interest statement None of the authors has any potential conflict of interest.

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