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CASE REPORT

Anterior submandibular retropharyngeal odontoid osteotomy and posterior atlantoaxial fusion for irreducible atlantoaxial dislocation associated with odontoid fracture malunion

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Abstract
Purpose To report a case of complex odontoid fracture malunion accompanied by atlantoaxial dislocation which was treated with a new surgical approach.
Methods A 53-year-old female was admitted due to progressive symptoms with a stiff limp and unsteady gait. Preoperative examination, diagnostic radiography, computed tomography, and magnetic resonance imaging were performed following admission. The examinations showed odontoid fracture malunion, an old right axial zygapophyseal fracture, atlantoaxial dislocation, and spinal cord injury. Anterior submandibular retropharyngeal odontoid osteotomy and posterior atlantoaxial fusion were then performed.
Results Good reduction of the atlantoaxial dislocation was gained. The cervical spinal cord compression was significantly relieved and neurological function was also significantly improved.
Conclusion Anterior submandibular retropharyngeal odontoid osteotomy and posterior atlantoaxial fusion fixation is an effective method for treating IAAD associated with odontoid fracture malunion, it avoids the adverse effects of anterior transoral odontoid osteotomy and provides a new option for the treatment of odontoid fracture malunion associated with atlantoaxial dislocation.

Keywords Anterior · Odontoid osteotomy · Atlantoaxial dislocation · Odontoid fracture · Malunion

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Introduction

Odontoid fracture malunion is intractable. It can lead to irreducible atlantoaxial dislocation and can cause spinal cord irritation. Anterior surgery is the usual primary treatment or is combined with adjuvant therapy for complete reduction of atlantoaxial dislocation. Anterior transoral odontoid partial resection or atlas anterior arch resection is routinely used for anterior reduction; however, the disadvantages include tedious perioperative preparation, narrow operative field, and high incidence of complications [1–3]. A patient with a complex odontoid fracture malunion accompanied by atlantoaxial dislocation was treated with a new surgical approach. Anterior submandibular retropharyngeal odontoid osteotomy was used for anterior reduction. This method avoids the adverse effects of anterior transoral odontoid osteotomy and provides a new option for the treatment of odontoid fracture malunion associated with atlantoaxial dislocation.

Case report

A 53-year-old female farmer in previous good health fell from a farm vehicle 3 months prior to evaluation. She had neck pain, but did not go to the hospital at that time. One month prior to evaluation, she had progressive symptoms with a stiff limp and unsteady gait. One week prior to admission, an odontoid fracture malunion associated with atlantoaxial dislocation was diagnosed using computed tomography (CT). Preoperative examination, diagnostic radiography, CT, and magnetic resonance imaging (Fig. 1) were performed following admission. The examinations showed odontoid fracture malunion, an old right axial zygapophyseal fracture, atlantoaxial dislocation, and spinal

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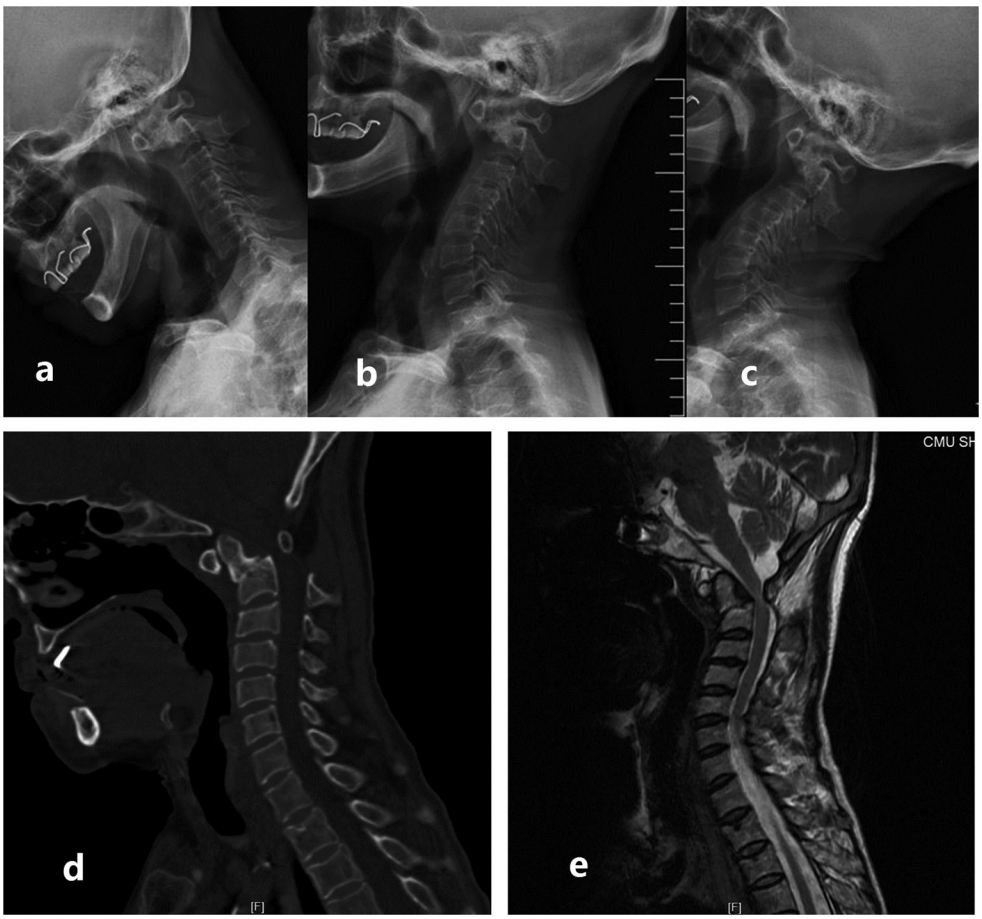

Fig. 1 A 53-year-old woman who had progressive symptoms with a stiff limp and unsteady gait was admitted. Odontoid fracture malunion on CT (**d**), irreducible atlantoaxial dislocation on dynamic DR (**a**–**c**) and spinal cord compression on MRI (**e**) were observed

cord injury. Physical examination showed increased limb muscle tone, decreased horizontality of the manubrium, right upper extremity, left upper extremity, and bilateral lower extremity muscle strength grades of 3, 4, and 4, respectively. Hoffmann sign: L(+), R(+); Babinski sign: L(+), R(+). The preoperative Japanese Orthopaedic Association (JOA) score was 8.

After induction of general anesthesia, skeletal traction was performed; traction weight was gradually increased to a maximum of 5 kg, with no change in dislocation. Anterior submandibular retropharyngeal odontoid osteotomy and posterior atlantoaxial fusion were then performed. The patient was placed in the supine position on a radiolucent table, with a pillow under the shoulder; her head was extended on a cranial ring that provided sustained traction (5 kg). A 4-cm oblique incision was made 2 cm inferior to the mandible and extended along the anterior border of the sternocleidomastoid muscle. The superficial fascia and platysma were then divided in line with the incision. The superficial layer of the deep cervical fascia was transected, exposing the sternocleidomastoid muscle and the superior belly of the omohyoid muscle. The superior thyroid artery along the superior laryngeal nerve could then be stretched for exposure. Care was taken not to injure or stretch the superior laryngeal nerve, which courses deep and parallel to the superior thyroid artery along the constrictor muscles of the pharynx and terminates at the level of the thyrohyoid membrane. Blunt dissection was then performed on the retropharyngeal space between the carotid sheath laterally and the larynx and pharynx medially. The mandible was retracted upward with an S-shaped handheld retractor. The alar fascia and prevertebral fascia were transected longitudinally, and the longus colli muscles were subperiosteally elevated to expose the C1–C2 lateral facet.

A long-head electrotome was used for incising the hyperplastic soft tissue, the contracted anterior longitudinal ligament, and the longus colli. Odontoid osteotomy was performed using a high-speed burr and ultrasonic bone curette (Fig. 2), in accordance with the preoperative planning. One assistant applied skeletal traction at the side of the head by slowly increasing cranial traction, with continuous monitoring of atlantoepistrophic joint reduction. The soft tissues around the caudal odontoid and bilateral atlantoaxial joint capsule were released because

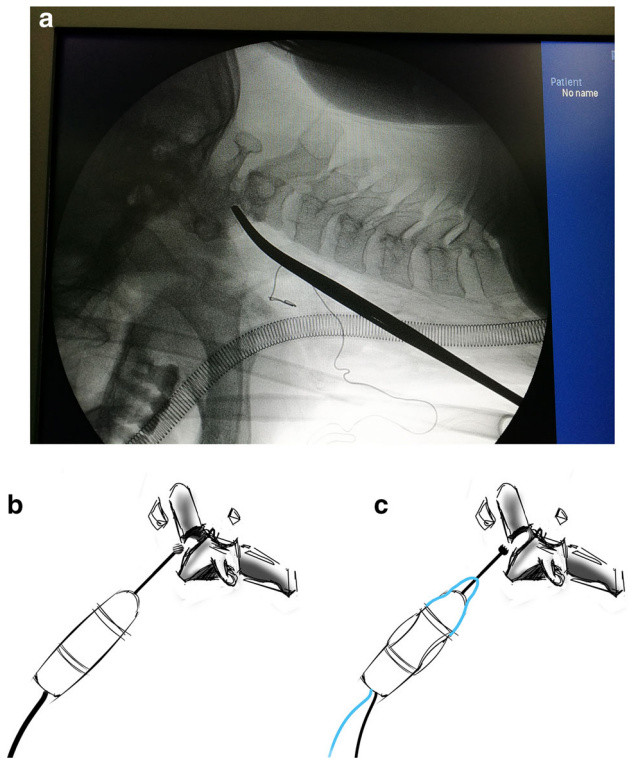

Fig. 2 Anterior submandibular retropharyngeal odontoid osteotomy and posterior atlantoaxial fusion were performed. The osteotomy starting point and direction were determined through intraoperative fluoroscopy (**a**), and a high-speed burr (**b**) and ultrasonic bone curettage (**c**) were used for osteotomy

atlantoaxial vertebral reduction was unsatisfactory. After the anterior incision was closed, cranial traction was removed, a neck collar was fitted, and the patient was placed in a prone position. With pressure on the C2 spinous process, X-ray showed good reduction of the atlantoaxial dislocation (Fig. 3). Cancellous bone was harvested from the posterior superior iliac spine for subsequent graft fusion. The surgery was successful and the operative time was 245 min, with about 200 ml of blood loss. CT reexamination showed that the ventral spinal cord was completely decompressed. The atlantoepistrophic slope angle increased from 126° to 141° (Fig. 4). The cervical spinal cord compression was significantly relieved and neurological function was also significantly improved. Reexamination 1 month after surgery showed a JOA score of 12, with a 50% improvement.

Discussion

The etiology of atlantoaxial dislocation includes trauma, inflammation, tumor, or congenital malformation. Decompression and internal fixation are usually performed [4, 5]. Yin et al. used clinical presentation to categorize reducible atlantoaxial dislocation (RAAD), irreducible atlantoaxial dislocation (IAAD), and fixed atlantoaxial dislocation (FAAD) [6]. Patients with IAAD can be repositioned through anterior transoral decompression, and those with FAAD undergo osseous atlantoaxial fusion. Patients with IAAD and FAAD are usually managed with combined anterior surgery [7, 8].

The atlantoaxial vertebra has a unique structure and its stability depends on the transverse and alar ligaments. An intact odontoid process is important for the stability of the atlantoaxial vertebra. The transverse and alar ligaments become ineffective when the odontoid process is fractured, leading to atlantoaxial instability and dislocation. Odontoid fractures are not generally associated with spinal cord injury, and only neck pain may be initially present. Thus, odontoid fractures are easily misdiagnosed and ultimately result in neurological impairment. Odontoid fracture malunion can lead to atlantoaxial dislocation and can be intractable.

Hao et al. [9] reported 22 IAAD cases treated with anterior atlantoaxial release through an anterior retropharyngeal approach, following traction reduction with C-arm fluoroscopy monitoring. Most cases were able to achieve

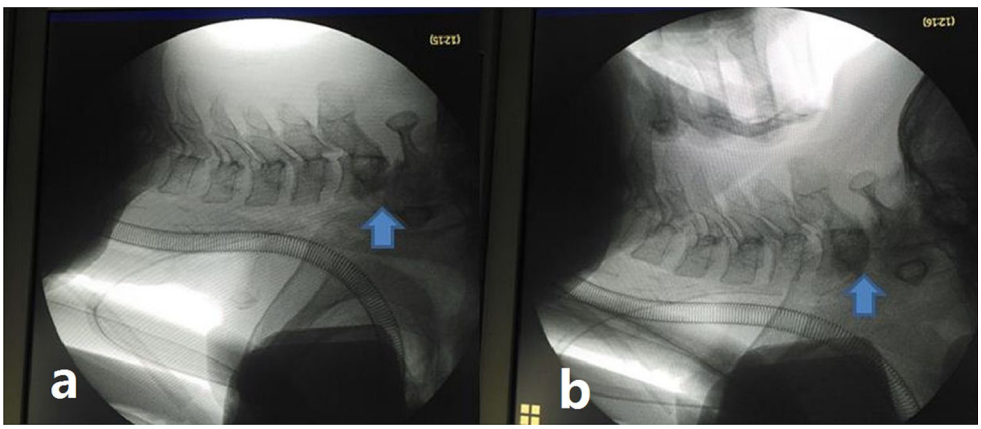

Fig. 3 The dislocation was obvious after the patient was placed in a prone position (**a**), and reduction of dislocation was seen with pressure on the C2 spinous process (**b**)

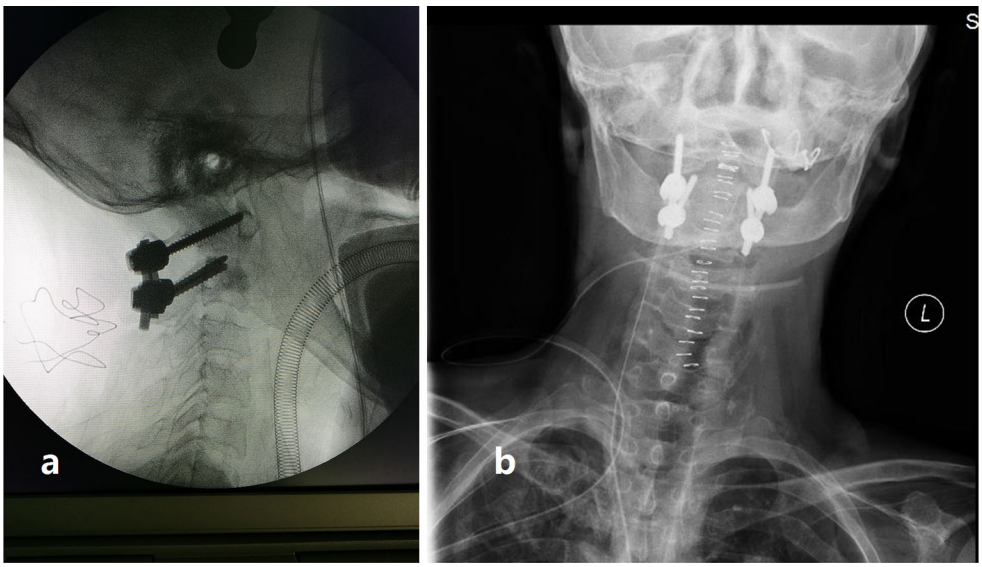

Fig. 4 Pedicle screws and lateral mass screw fixation were used in posterior surgery; good effect of fixed location and reduction were seen through lateral (**a**) and front (**b**) DR after the operation

satisfactory reduction by dissecting the bilateral longus colli and longus capitis muscles and the anterior longitudinal ligament caudal to the anterior ring of C1 with long-tip Bovie electrocautery, while a few cases achieved reduction with release of C1–2 articular contraction and resection of the anterior arch of C1.

Xu et al. [10] performed transoral anterior release, odontoid partial resection, and reduction with posterior fusion for the treatment of irreducible atlantoaxial dislocation caused by odontoid fracture malunion. After surgery, the rate of improvement of neurological symptom achieved 87.4% and the average blood loss was about 670 ml (range 510–930 ml).

Vender et al. [11] described seven patients requiring C-2 corpectomy who underwent retropharyngeal C1–3 fusion, followed by instrumentation with a Caspar plate. Despite good therapeutic effect, a tracheal cannula was required for 3–5 days to relieve pharyngeal and retropharyngeal edema, and supplemental intravenous or nasogastric nutritional support was required.

Anterior transoral odontoid partial resection is increasingly used because the anatomic structure of the cervical region is complex. However, because the operative space is narrow and the infection rate is high, serious complications can occur, including cerebrospinal fluid leakage, wound infections, or meningitis [1–3]. In addition, patients sometimes have extreme throat discomfort and difficulty feeding. Anterior submandibular retropharyngeal odontoid osteotomy can avoid or reduce such complications and can also reduce perioperative preparation.

The anticipation of osteotomy direction and safe gap measurement are of great significance to the surgery. A high-speed drill was used in our study to strip odontoid sclerostin for anterior transoral odontoid osteotomy, because the malunion of the odontoid base is relatively wide. Because the spinal cord moves forward in anterior atlas dislocation, there is great risk in stripping the back wall of the odontoid, as this may cause a tear in the endorhachis and injure the spinal cord. The procedure is difficult because of the deep position of the upper cervical spine and poor stability with a long-handled drill. Accordingly, ultrasonic bone curettage and a high-speed burr were used for osteotomy because of their high efficiency and speed, as well as the safety of piezosurgery. A high-speed burr was used to remove the odontoid base of the malunion, creating a 2-mm-wide groove; shaving of residual sclerostin was performed with piezosurgery until the back wall of the odontoid malunion was completely excised (Fig. 2).

The habitus of the patient is also an important factor in surgery; patients with obesity or a short neck are not suitable for this approach. The patient in our study had a height of 148 cm and weight of 36 kg; she had a long neck and high mandible. These provided appropriate conditions for the operation. In contrast with that of a rotating high-speed drill bit, piezosurgery significantly reduced the risks of mechanical injury to vascular and neurologic tissue and the dura mater. The self-irrigating cooling feature of piezosurgery can spray sufficient water to reduce the risk of local thermal and mechanical damage, and provide good protection for the spinal cord [12, 13].

With the aim of reduction and stable fixation, posterior atlantoaxial fusion was performed after anterior surgery. The right side of C1 was fixed with bilateral cortical

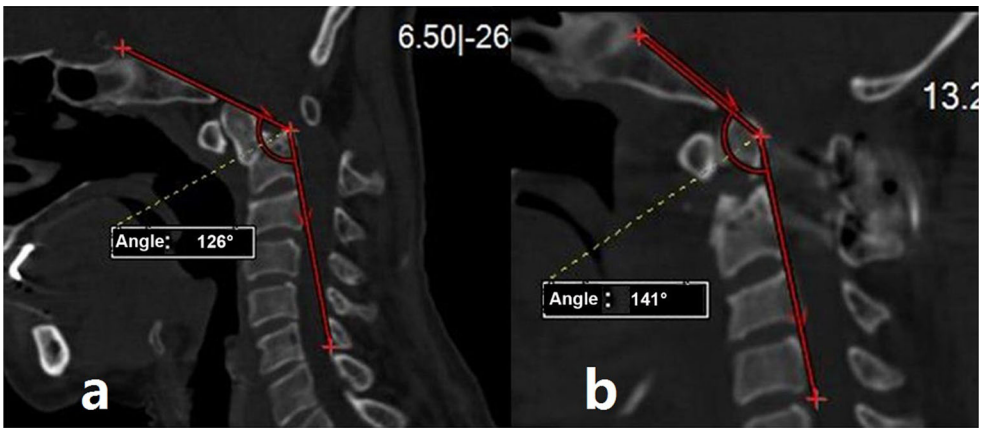

Fig. 5 CT reexamination showed that the atlantoepistrophic slope angle increased from 126° to 141° (**a**, **b**) and the ventral spinal cord was completely decompressed (**b**)

screws, because the right side pedicle was too thin. The left side of C1 and bilateral sides of C2 were fixed using pedicle screws (Fig. 5). The patient returned to a regular ward after surgery without hoarseness, swallowing difficulty, or cough. Soft food was tolerated the day after surgery and the drainage tube was removed after 48 h. The patient was able to get out of bed with a neck collar in place.

This operative approach has the advantages of minimal trauma, fast recovery, and simple perioperative preparation and nursing, which help to avoid serious oropharyngeal complications. In addition, this method significantly reduces the likelihood of pharyngeal discomfort or difficulty feeding. However, the anatomic structure of the anterior high cervical spine is complicated. As traction during surgery may injure the laryngeal nerve, extensive dissection of subfascial planes and neurovascular bundles is necessary to reduce stretch injury and provide greater surgical freedom [14, 15].

In our patient, spinal cord compression mainly resulted from fractures of the proximal odontoid and posterior arch of the atlas. Preservation of the anterior arch and release of surrounding soft tissue can avoid posterior occipitocervical fusion, not only relieving compression on the ventral spinal cord but also maintaining atlanto-occipital stability. A shorter anterior operative time and reduced traction and irritation of anterior neck tissues will reduce common postoperative complications. Pedicle screws and lateral mass screw fixation were used in posterior surgery to lift and reset the atlantoaxial vertebra through a lever principle with satisfactory results. The atlantoaxial pedicle screw system has good three-dimensional stability and biomechanical performance and can meet the needs of atlantoaxial fusion fixation.

Anterior submandibular retropharyngeal odontoid osteotomy and posterior atlantoaxial fusion fixation is an effective method for treating IAAD associated with odontoid fracture malunion. This approach can avoid the many drawbacks of anterior transoral surgery and can reduce the risk of injury to important tissues caused by increased operative time in anterior fusion fixation, maintain atlanto-occipital stability, and obtain satisfactory outcomes.

Compliance with ethical standards

Conflict of interest The authors declare that they have no actual or potential conflicts of interest.

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