Corrective surgery for deformity of the upper cervical spine due to ankylosing spondylitis

Bin Lin, Bi Zhang, Zhu-mei Li, Qiu-sheng Li

Abstract

Rotational and flexion deformity of C1-C2 due to ankylosing spondylitis is rare. We did surgical correction in one such case by lateral release, resection of the posterior arch of C1 and mobilization of the vertebral arteries, wedge osteotomy of the lateral masses of C1 and internal fixation under general anesthesia. There were no vascular and neurological complications during the surgery. After operation the atlantoaxial rotational deformity was corrected and the normal cervical lordosis was restored. At 1 year followup his visual field and feeding became normal and internal fixation was stable.

Key words: Ankylosing spondylitis, cervical deformity, internal fixation, osteotomy

Introduction

Ankylosing spondylitis is a chronic inflammatory rheumatic disorder that primarily affects the axial skeleton.[1] The rotational and flexion deformity of the upper cervical spine due to ankylosing spondylitis is a rare disease. The cervical deformity has great influence on patients' daily life. However, the clinical treatment of it is still a challenge as the corrective surgery of the upper cervical spine has great risk. So the corrective surgery of the severe rotational deformity of the upper cervical spine is rare.

We successfully performed such a corrective surgery on a patient with rotational and flexion deformity of the upper cervical spine by lateral release, resection of the posterior arch of C1 and mobilization of the vertebral arteries, wedge osteotomy of the lateral masses of C1, and internal fixation under general anesthesia. After the surgery, his clinical symptoms had disappeared, his appearance had been improved significantly.

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Access this article online	
Quick Response Code:	Website: www.ijoonline.com
	DOI: 10.4103/0019-5413.128771

Case Report

A 32 year old patient a known case of ankylosing spondylitis[2] presented to us with upper limb pain and morning stiffness for 13 years. With the progression of the disease the rotational and flexion deformity of the upper cervical spine gradually increased, leading to a severe rotational deformity of the cervical spine [Figure 1A and B] and difficulty in feeding. This rotational cervical spine deformity limited the activity of the neck and the visual field. The muscles of neck were in spasm and bilateral

Figure 1A: (a) Clinical photograph front view demonstrating the degree of the flexion deformity with rotation to the right, limiting the activity of cervical spine and the visual field (b) Clinical photograph side view demonstrating the degree of the rotational deformity of the cervical spine shoulder levels were not equal. The muscle strength of both of lower limbs and muscle tone were normal. However, the muscle strength of the patient's upper limbs was in medical research council (MRC) grade IV. Fine touch sensation of skin of the right forearm, thumb, forefinger, and the middle finger was reduced. His HLA-B27 test was positive, the erythrocyte sedimentation rate was 50 mm/h (Westergren method) and C-reactive protein was 26 mg/L (speed scattering turbidimetric method). His pelvic X-ray [Figure 1B] showed the fusion of bilateral Sacroiliac joints, three-dimensional CT revealed a rotational deformity of the upper cervical spine [Figure 1B]. This patient was given conservative treatment with skull traction for four weeks, the clinical symptoms of the cervical spine and the upper limbs did not improve. A surgery was thus planned.

The patient was positioned prone under general anesthesia, with the head in the horseshoe-shaped head fixator and with maintained skull traction [Figure 2x]. The "U" pad was under his chest and abdomen. Transcranial electrical stimulated motor evoked potential monitoring (TES-MEP) was done to insure the safety of the procedure. A 10-cm incision was made from the right mastoid to the posterior edge of the sternocleidomastoid [Figure 2y]. Dissection was done through the subcutaneous tissue, sternocleidomastoid was retracted forwards, trapezius was cut along with the splenius and the first half of spinal muscles. This revealed the posterior arch of atlas and the transverse process. Then we released the right side atlantoaxial joint and the right anterior arch from atlantoaxial gap to the front side of the atlas. A midline incision was made from occipital protuberance down to the cervical spinous process of C3 [Figure 2y]. The subcutaneous tissue, the nuchal ligament were then dissected. Near the spinous process and vertebral plate, pushed both sides of the trapezius muscle, capitis, head semispinalis and vertebral occipital muscle outward to the outer edge of the small joints. As the midline incision was not full to reveal, a transverse incision was made to connect the two incisions [Figure 2y]. Then we stripped the right side of small rhomboid muscle, capitis, splenius neck, head dorsi muscle and the levator scapulae. Opened both sides of muscle tissue by self retaining retractor, dissected

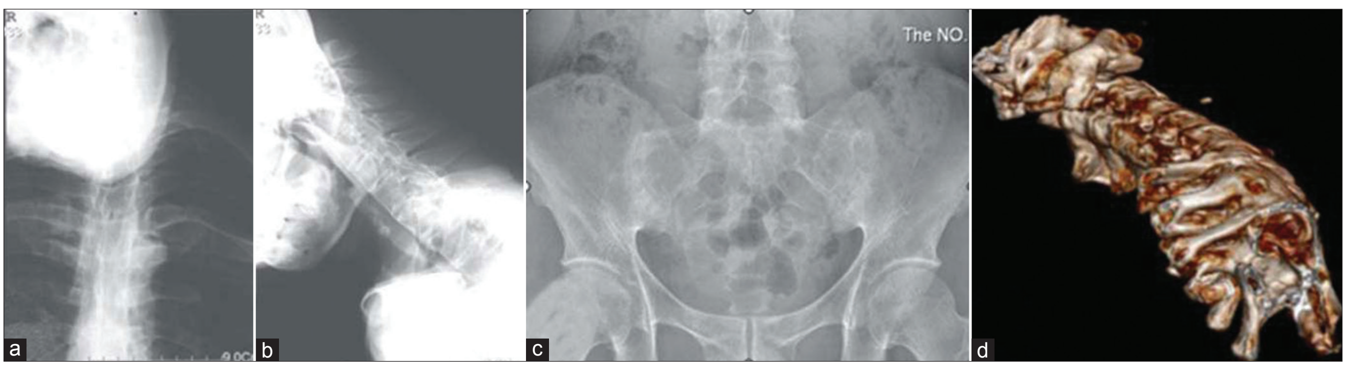

Figure 1B: Preoperative anteroposterior (a) and lateral view (b) cervical radiographs revealing the flexion deformity between the cranial base and C1 and C2. The position of the mandible in relation to the cervical spine can be clearly seen (c) Pelvic X-ray of both hips (anteroposterior view) showing the fusion of bilateral sacroiliac joints (d) Three-dimensional CT demonstrating the rotational deformity of the upper cervical spine

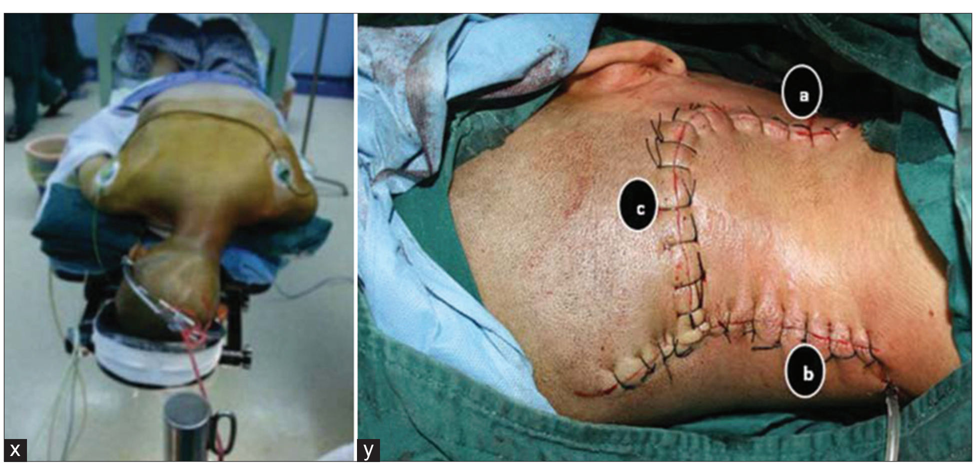

Figure 2: Peroperative photographs showing (x) the patient positioned prone, with his head in the horseshoe-shaped head fixator, maintained in skull skeletal traction (y) The explanation of the three incisions. (a) A 10-cm incision was made from the right mastoid to the posterior edge of the sternocleidomastoid. (b) A midline incision was made from occipital protuberance down to the cervical spinous process of C3. (c) Because the midline incision was not full to exposure, a transverse incision was made to connect the two incisions to expose fully

attached muscle on the occipital bone, the posterior arch of the atlas, axis, behind the axis, the lamina, the lateral mass of C2-3 by periosteal stripping and sharp knife. This revealed lateral mass of C2-3, spinous process, lamina, atlanto-occipital joint. We stripped the adhesive organization of the atlantoaxial joint. Then we used burr drill to break cortex openings in the cervical pedicle and probed the walls of the bony hole, and measured the depth of the bone hole. The appropriate cervical pedicle screws were inserted into the bony holes respectively. Two appropriate pedicle screws were inserted into the bilateral pedicle of C2 in the same way. Holes were drilled in the bilateral lateral mass of C3 respectively, and two lateral mass screws were inserted into C3. At the side of the occipital protuberance, we drilled the cortex by grinding drill, drilled holes by pyramid, inserted two appropriate screws on both sides of the occipital protuberance, respectively. Nerves and blood vessels were mobilized with nerve retractors, then 30-degree wedge osteotomy was performed by the high-speed grinding drill, with the intercepted structure of lower edge of the right posterior arch of C1 cut [Figure 3]. Finally, the intercepted structure of the left side of the upper edge part of C2 was cut [Figure 3], then rotational deformity was resetted under skull skeletal traction. After resetting the cervical sagittal sequence, the bended rod was placed [Figure 4]. Then it was made sure that the rotational deformity had been corrected and the position of the internal fixation was good by C-arm fluoroscopy. During the surgery, there were no vertebral artery, spinal cord and nerve root complications. After the surgery the atlantoaxial rotational deformity was basically corrected. The X-ray and CT scan showed the position of internal fixation was good [Figure 5a-c]. Followed up for 1 year, his clinical symptoms had disappeared, his appearance had been improved significantly, his visual field and feeding became normal [Figure 6]. The followup X-ray [Figure 7a and b] and three dimensional CT [Figure 7c] showed that the position of internal fixation was good, without loosening and implant breakage.

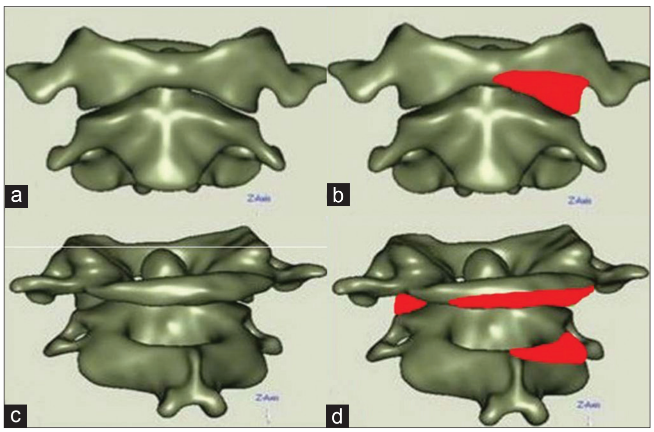

Figure 3: Diagrammatic illustrations of the stages of the operative procedure. (a) preoperative diagram of the upper cervical spine (b) 30-degree wedge osteotomy, cut the intercepted structure of lower edge of the right posterior arch of C1 (c) After 30-degree wedge osteotomy through the lateral mass of C1, the odontoid process can be visualized (d) After wedge osteotomy, the intercepted structure of the left side of the upper edge part of C2 was cut

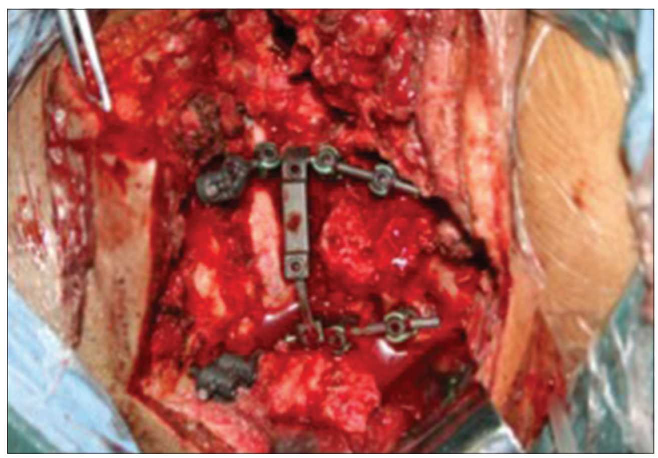

Figure 4: After resetting the cervical sagittal sequence, the bended rod was placed, and the installation of the internal fixation was completed

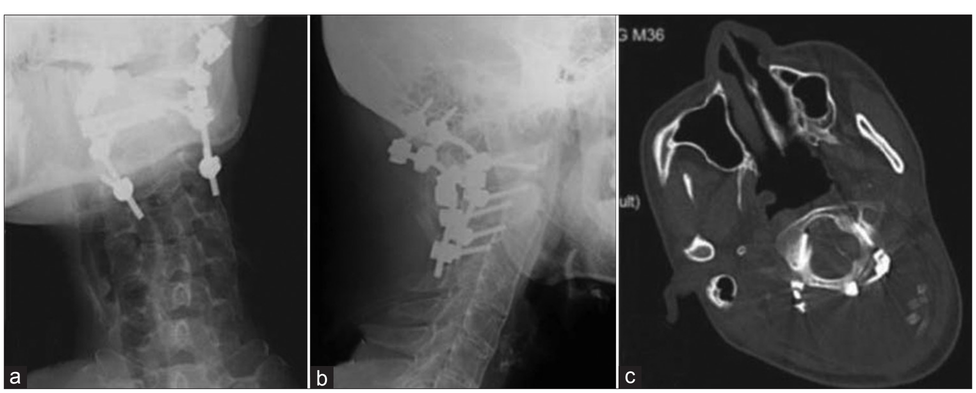

Figure 5: (a) Postoperative anteroposterior radiograph showing the atlantoaxial rotational and inflexional deformity was basically corrected (b) Postoperative lateral cervical radiograph showing the atlantoaxial flexion deformity was corrected, the position of internal fixation was good (c) CT scan showing the position of pedicle screw was good

Discussion

The pathogenesis of the rotational and in flexional deformity of the upper cervical spine (C1 C2) due to ankylosing spondylitis is still not clear. Gore *et al.*[2] observed the degree of lordosis from C2 to C7 in patients with osteoarthritis ranging from 16° to 22° in men and from 15° to 25° in women. However, the normal range of this degree varies according to the standard measurement. In normal lordosis of cervical spine, the cervical spine load-bearing axis lies to the posterior vertebral bodies of C2-C7. Vertebral withstands a pressure from the central axis about 36%, and the posterior column withstands a pressure approximately 64%. This balance maintains the normal cervical curvature and maintains the head in the normal position. Any disruption of this balance will lead to a sagittal abnormality, and bring about a cervical deformity.[3,4]

Urist *et al.*[5] firstly reported cervical osteotomy for cervical kyphosis due to ankylosing spondylitis in 1958. As the failed endotracheal intubation due to the cervical flexion,

Figure 6: Clinical photograph at 1 year followup showing that the rotational deformity of the cervical spine was basically corrected, his clinical symptoms had disappeared, his visual field became normal

the patient was placed in a sitting position under local anesthesia. However, other surgeons tended to position the patient prone with general anesthesia to ensure the safety of the surgery.

There are some traditional approaches for the upper cervical spine surgery. That is the transoral approach, anterior and posterior combined approach and posterior approach. The indications for anterior approach are considered as follows: (1) disease from the anterior elements of the upper cervical vertebrae; (2) anterior compression of the medulla and spinal cord which cannot be decompressed by a posterior approach; (3) recurrent affection of the medulla and spinal cord which cannot relieved by posterior depression; (4) instability which cannot be treated by posterior operation because of disease or absence or inaccessibility of posterior elements;[6] The transoral atlantoaxial reduction plate (TARP) designed by Yin QS[7] is adapted to atlas and the axis of irreducible anterior dislocation caused by disorders, such as basilar invaginationt, Arnold-Chiari, Congenital odontoid dysplasia, Odontoid free, Rheumatoid arthritis and odontoid old fracture and transverse ligament of atlas fracture scar formation. Range of application can be extended to instable Jefferson fracture, tumor, tuberculosis, and eosinophilic granuloma. But the atlantoaxial structure must be integrated, including the lateral mass of atlas and axis vertebrae. Yin *et al.*[7] took the transoral approach to release and reduce dislocation of atlantoaxial, and perform fixation at the same time, effectively avoid possible risk of spinal cord injury. However, there is a defect which the atlantoaxial screw fixed on the vertebral lacks of holding force, so there is a risk of atlantoaxial vertebral screw loosening and fixation failure.[8] Another defect is that the reduction plate does not meet up with the atlantoaxial physiological curvature in the sagittal, so postoperative X-ray shows atlantoaxial physiological curvature has not been restored well.[9]

There are many clinical reports about simple posterior

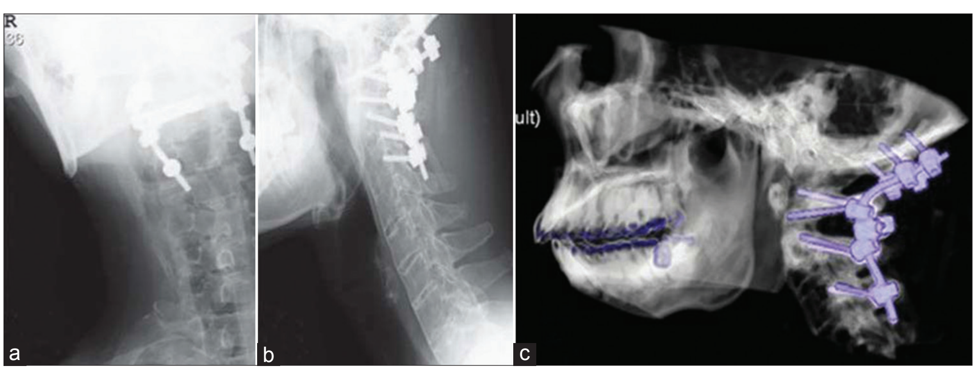

Figure 7: (a) X-ray cervical spine anteroposterior view at 1 year followup showing a rotational deformity of the cervical spine had been corrected and the position of internal fixation was good (b) lateral view of cervical radiograph showing the atlantoaxial deformity had been corrected, and the position of internal fixation was good, without loosing and breakage (c) Three-dimensional CT at one year followup showing the position of internal fixations was good

fixation, for example, Hong Bo *et al.*[10] had compared the biomechanical stability of pedicle screws with various established posterior atlantoaxial fixations used to manage atlantoaxial instability. The C1-C2 transarticular screw and Gallie fixation procedures provid the highest stability. The Gallie fixation alone may not be adequate for atlantoaxial arthrodesis, because it does not provide sufficient stability in lateral bending and rotation modes. In the C1 lateral mass-C2 pedicle screw fixation, the use of a short pedicle screw may be an alternative when other screw fixation techniques are not feasible. Sonntag's Gallie wire fixation technique was worse than other reconstruction techniques. In recent, however, the posterior fixation techniques have been improved. Related techniques such as pedicle screws, lateral mass screws, and laminae within the screw have significantly improved the fusion rate of the upper cervical spine, and improved the stability and safety. The patients generally need persistent halo ring fixation.[11] The technique of C1, C2 zygopophysis screw, greatly increases the rate of atlantoaxial fusion. The zygopophysis screws used to fix the three columns of C1, C2 firmly combined with posterior wire and spinous process of the screws form a three points of fixation mode, which limits atlantoaxial flexion and extension and rotation, provides a stability of the upper cervical spine.[12] However, the majority of upper cervical deformities are not corrected directly, mainly through correcting the lower cervical spine or thoracic spine as a compensatory surgery. There is a high risk of upper cervical spine osteotomy, so this technique is still not be applied widely. Grundy *et al.*[13] reported one case of osteotomy of the upper cervical spine deformity due to ankylosing spondylitis. It was reported the upper cervical vertebraes were exposed via a midline posterior incision, the posterior arch of C1 was excised, and the vertebral arteries were mobilized, a wedge osteotomy was performed through the lateral masses of C1 and subsequently through the odontoid. They believed that it was possible to gain sufficient surgical access to the odontoid process via a posterior approach. The described technique is beneficial when the alternative anterior approaches to the upper cervical spine are technically difficult or impossible.

Simple transoral anterior operation can achieve completely decompression for spinal cord, but it has higher rate of infection after surgery and needs to rely on external fixation for a long time after surgery, so it is difficult for patients to accept. A simple posterior procedure can not complete fully decompression, releasing, and good reduction.Considering the defects of a simple anterior or posterior procedure, Chao Wang *et al.*[14] took the anterior release combined with posterior fixation to treat irreducible atlantoaxial dislocation in 2006. They reported 33 patients with irreducible atlantoaxial dislocation underwent one stage anterior release and posterior fixation and fusion, gained the safety and efficacy. But the severe rotational and flexion deformity of the cervical spine due to ankylosing spondylitis do not allow us to take the transoral anterior operation.

We gained satisfactory result from the patient with the rotational and flexion deformity of the upper cervical spine, but we still need more cases to evaluate the surgical procedures and the safety of the corrective surgery.

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How to cite this article: Lin B, Zhang B, Li Z, Li Q. Corrective surgery for deformity of the upper cervical spine due to ankylosing spondylitis. Indian J Orthop 2014;48:211-5.

Source of Support: Nil, **Conflict of Interest:** We declare that we do not have any commercial or associative interest that represents a conflict of interest in connection with the work submitted.