CASE REPORT

Cervical rotational osteotomy for correction of axial deformity in a patient with ankylosing spondylitis

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Abstract

Purpose Severe cervical axial deformity associated with ankylosing spondylitis (AS) is rare in clinic, and there are little concerns about surgical treatment of axial deformity associated with AS. The case study aims to show the surgical technique to perform cervical rotational osteotomy.

Methods We present the case of a young AS patient whose neck was fixed in a left-rotational posture at 18°, requiring his trunk to be turned to the right to look forward visually. This made his gait appear to be limping, inconveniencing him with great difficulty. In order to correct this deformity, we performed a novel cervical rotational osteotomy through a one-stage posterior–anterior–posterior approach. Firstly, we performed laminectomies of C7 and T1, followed by a C7/T1 facetectomy with release of the bilateral C8 nerve roots. Next, we performed C7/T1 discectomy, bony resection of the lateral body and uncovertebral joints. The head of the patient was then rotated manually, so that both his face and torso were simultaneously facing frontward. Finally, rods spanning the screws from C6 to T2 were fixed.

Results Postoperatively, the patient's axial malalignment was significantly improved, and he was able to walk normally. Surgical outcomes were well maintained at a 3-year follow-up.

Conclusion Through this case, we hope to draw the attention to spinal axial deformity and provide a reference point in the surgical treatment of spinal axial deformity.

Keywords Axial deformity · Ankylosing spondylitis · Cervical · Osteotomy

Introduction

Ankylosing spondylitis (AS) is a chronic immune-mediated inflammatory disease, which primarily affects the spine and sacroiliac joints. In the late stages of AS, patients can develop significant fixed spinal three-dimensional deformity. Sagittal deformity is most often secondary to AS, such as thoracolumbar kyphosis, chin-on-chest deformity, or ear-on-shoulder deformity. This results in difficulty being able to look forward or lie down horizontally, displaying problems with ambulation, dysphagia, and dyspnea. Currently, there is extensive literature on osteotomy techniques for AS patients with sagittal deformity [1, 2]. However, there is little concern regarding axial deformity associated with AS.

Additionally, there is a lack of reports concerning the surgical treatment of AS patients with severe axial deformity.

We present the case of a young patient with AS, who had previously complained about severe neck stiffness. There was axial rotational deformity that resulted in his trunk having to be turned to the right, in order to look forward visually. This caused him to walk lopsidedly, as if limping, with the patient reporting of tiring easily. We performed a novel 360° cervical rotational osteotomy through a one-stage posterior-anterior-posterior approach to correct his cervical axial deformity, allowing his face and torso to simultaneously face forwards. The operation was successful, for postoperatively, the patient no longer had to walk with his trunk turned to the right and there was no difficulty in his gait. Overall, the surgery significantly improved his quality of life. Through this case, we hope to draw attention to spinal axial deformity and provide a reference in the surgical treatment of spinal axial deformity.

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Patient information

A 24-year-old male patient, who had a 12-year history of AS, complained about significant neck stiffness recurring over 5 years, with no neurological symptoms. He was unable to rotate or flex his neck. His horizontal gaze was well-maintained, and his neck was fixed at 18° of left rotation (Fig. 1). To look forwards while walking, he had to turn his body to the right (Fig. 2a), causing him to walk with difficulty, as though limping (see supplementary video 1). Due to the asymmetric manner of walking, he easily becomes tired and suffers from right hip pain.

Instead, if he had walked with his trunk facing frontwards, his head would be constantly turned to the left (Fig. 2b). As a result, he must walk slowly and exercise extreme caution to avoid falling (see supplementary video 2). He had received bilateral total hip replacements 1 year ago.

He reported that symptoms initiated insidiously over recent years, with mild cervical pain. Present reports stated, he was incapable of rotating his head, which was now fixed in a left-rotational position.

Upper and lower limb strengths were grade V (normal); sensitivity was normal and symmetric in both the torso and limbs. No pathologic reflexes or signs of myelopathy were found.
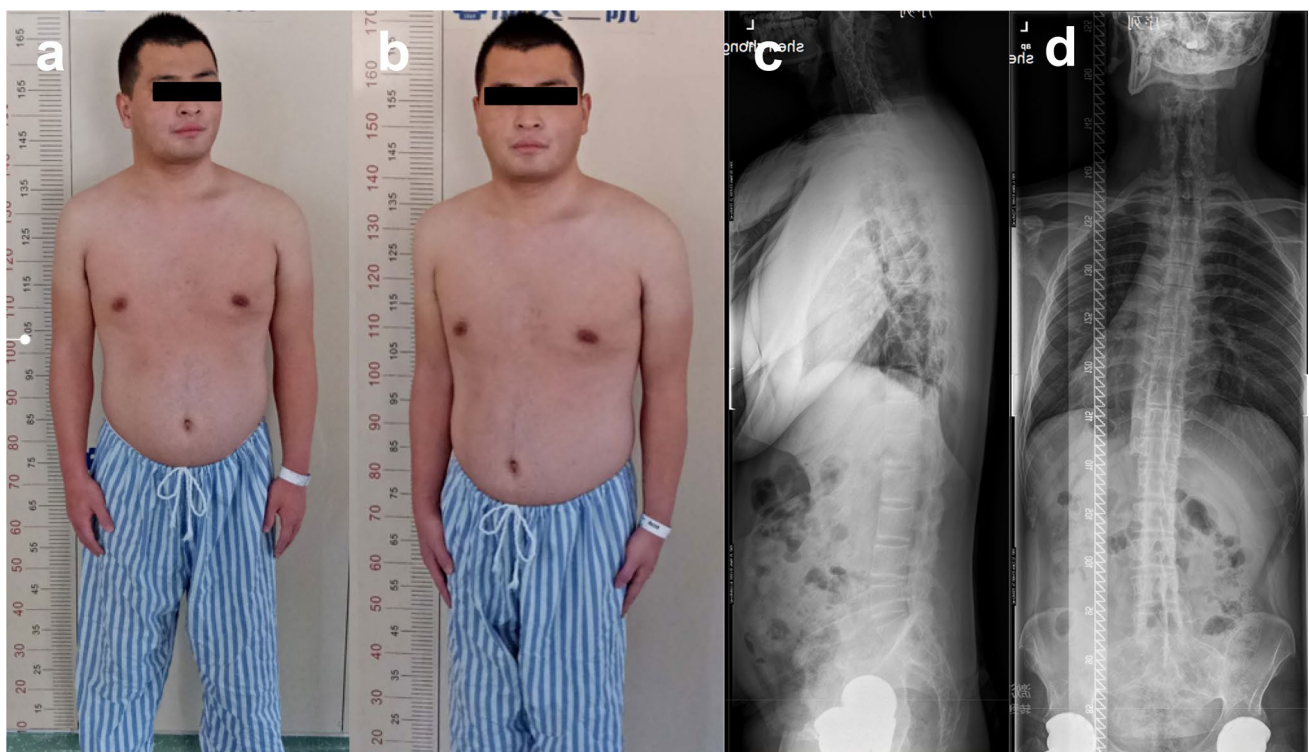

Fig. 1 AS patient with cervical axial malalignment. His neck was fixed in a left-rotational position at 18° (**a**). To look forward, he had to turn his body to the right (**b**). Preoperative full-length spinal plain radiographs with lateral position (**c**) and anteroposterior position (**d**)

Fig. 2 Schematic diagram showing the patient's manner of walking. **a**: walking with his face facing forwards; **b**: Walking with the trunk facing frontwards
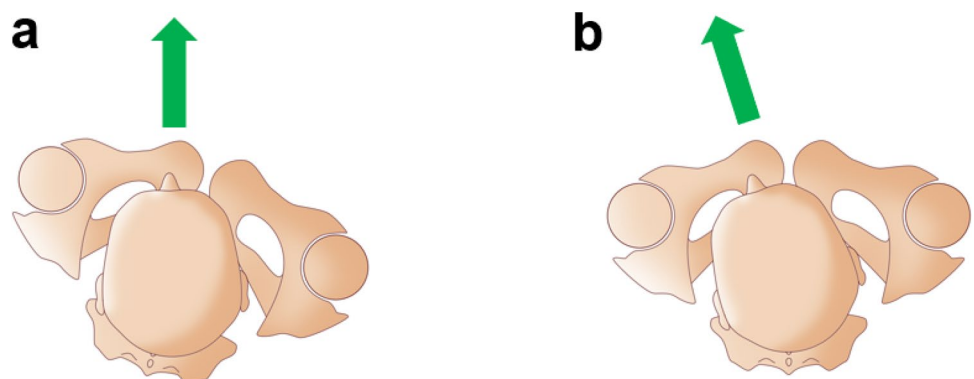
The patient had no significant cervical sagittal deformity. His global sagittal balance was well-maintained (Fig. 1c). His trunk shift was measured as the horizontal distance between the central sacral vertical line and the C7 plumb line. His C7 plumb line shift to the left was 2.7 cm. Radiographic shoulder height was measured as + 1.2 cm (Fig. 1d). The entire lumbar segments and most of the thoracic segments were observed to have been fused posteriorly (Fig. 3a, b). The entire cervical spine was fused anteriorly and/or posteriorly, including the atlanto-axial joint (Fig. 3c, d, e). From the CT scan, we observed that the rotation mainly occurred in the upper cervical segments, namely from the occiput to C2 (Fig. 4). The angle between the axial midline of the foramen magnum (FM) and C1 was noted as 2°. The angle between the axial
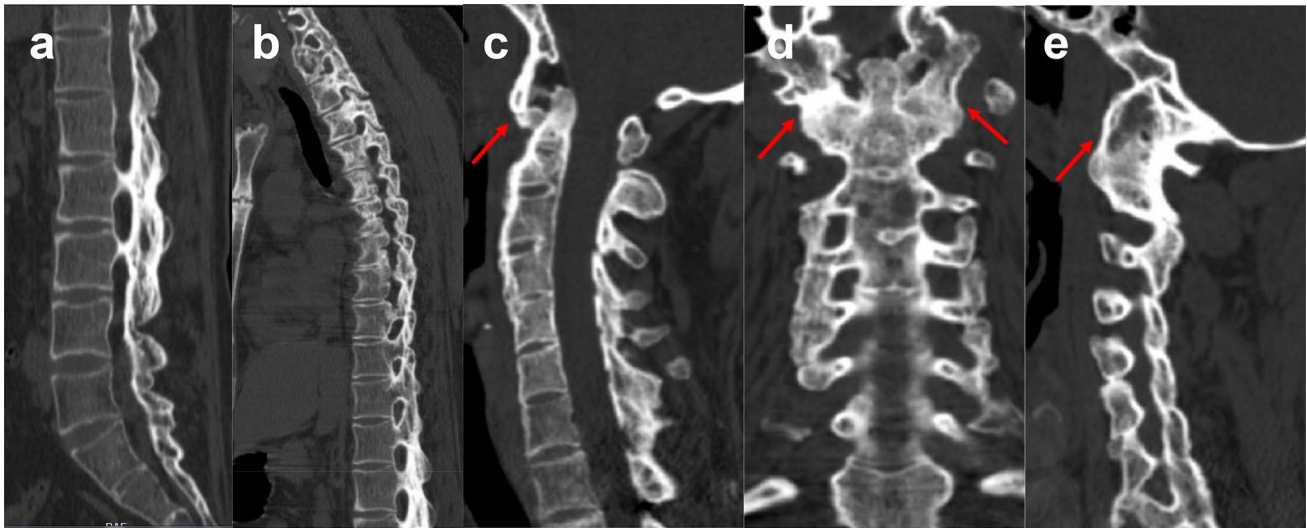
Fig. 3 Preoperative spinal CT sagittal reconstruction. **a**: lumbar sagittal CT; **b**: thoracic sagittal CT. The cervical segments were fused anteriorly and/or posteriorly, including the atlanto-occipital joint and atlanto-axial joint. It was circumferentially fused from occiput to axis (**c**, **d**, **e**)
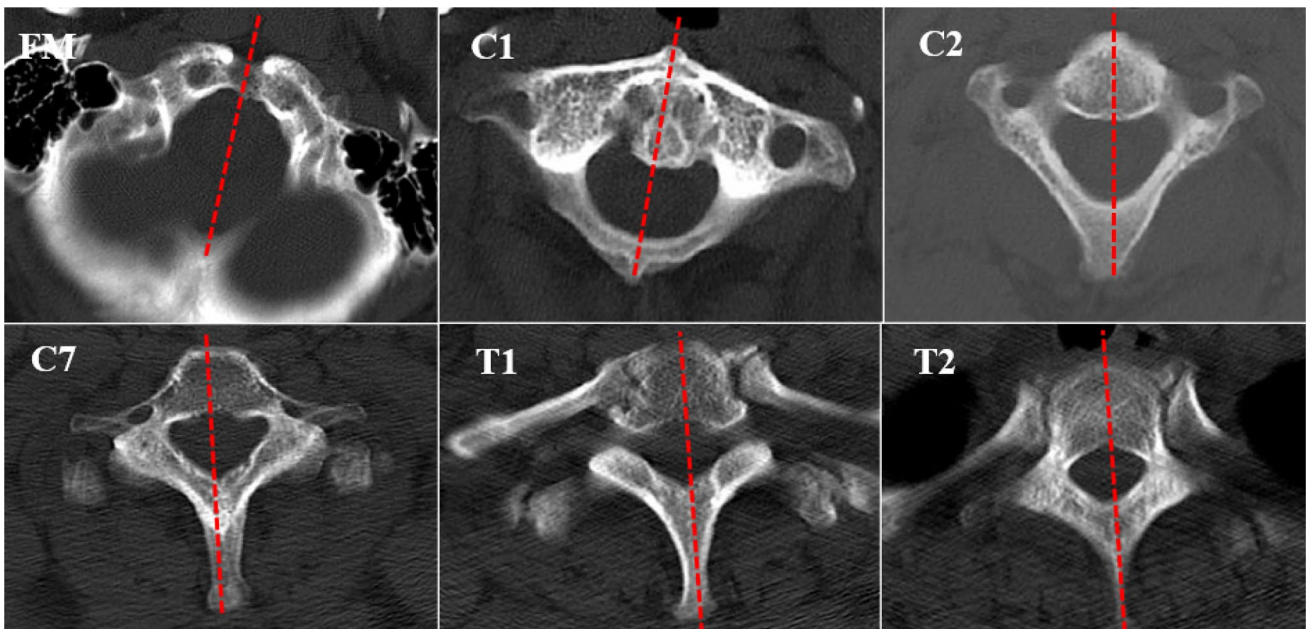
Fig. 4 Axial rotation of the cervical spine. The angle between the axial midline of FM and C1 was 2°. The angle between the axial midline of C1 and C2 was 9°. The central axes of C2 and C7 were approximately 7°. The angle between the axial midline of C7 and T2 was 0°. The total angle between the axial midline of FM and T1 was 18°. FM: foramen magnum

midline of C1 and C2 was noted as 9°. The angle between the axial midline of C2 and C7 was recorded as 7°. In total, the angle between the axial midline of the FM and C7 is recorded as 18°.

Surgical strategy

The goal of this surgery was to correct the axial misalignment and restore the patient's ability to look forwards without having to turn his body to the right. Given that his cervical facet joints were fused, the patient first requires a posterior osteotomy, followed by an anterior osteotomy to achieve a 360-degree release, and finally posterior instrumentation [1]. Thus, one-stage posterior-anterior-posterior approach surgery was carried out with intraoperative neuromonitoring.

The patient was placed in a prone position on the operating table with his head secured by a Mayfield horseshoe headrest. Bilateral lateral mass screws were placed in C6, and pedicle screws were placed in C7–T2. The posterior osteotomy included laminectomies of C7 and T1, followed by a C7/T1 facetectomy with exposure of the C8 nerve roots in the neural foramina. Bilateral intervertebral foramens were enlarged by partial resection of C7 pedicles to make sure the C8 nerve root was fully decompressed (Fig. 5a). The wound was temporarily sutured after gauze packing.

The patient was then turned to a supine position. At this time, his head could not be turned upwards (Fig. 6a, b). Therefore, a regular Smith–Robinson approach was used
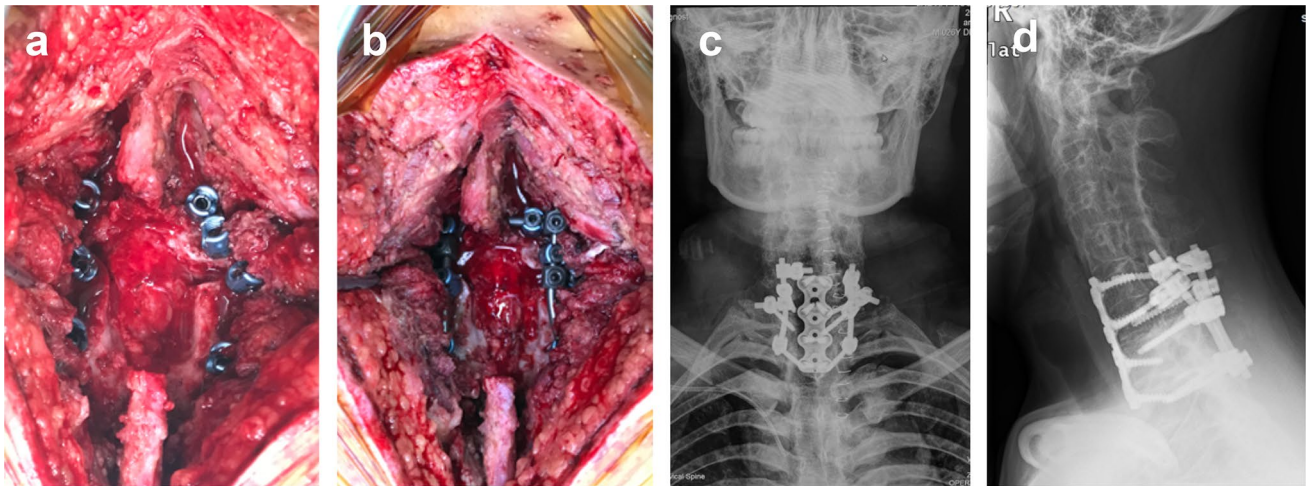
Fig. 5 Screws were inserted from C6–T2 in the first-stage surgery. The posterior osteotomy included laminectomies from C6–T2, followed by a C7–T1 facetectomy with exposure of the C8 nerve roots in the neural foramina (**a**). Two rods spanning from C6 to T2 were fixed using connectors on both sides in the third-stage surgery (**b**). Postoperative X-ray showed that the neck rotational deformity was largely corrected (**c**, **d**)
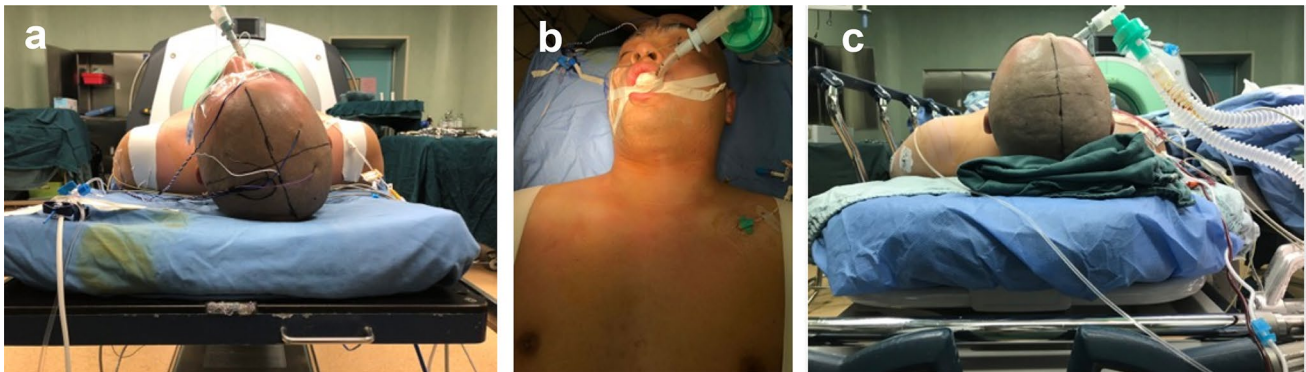
Fig. 6 The patient's neck was fixed in a position of about 20° left rotation after general anesthesia (**a**, **b**). After cervical rotational osteotomy at C7/T1, his face and torso were able to simultaneously face frontward (**c**)

to expose the C7/T1 intervertebral disc. Discectomy, bony resection of the lateral body and uncovertebral joints, and entry into the transverse foramen were among the surgical procedures involved. After complete release at the C7/T1 level, the head was then rotated manually, to upturn his face until the tip of his nose faced frontward. A titanium plate was then fixed.

After the prior procedures, the patient was then placed in a prone position once again. The spinal cord and nerve roots at the osteotomy site were probed to confirm that there was no obvious impingement of the neural structure. Rods spanning the screws from C6 to T2 were fixed using the connector on both sides (Fig. 5b). The patient's face and torso were able to simultaneously face forwards after the rotational osteotomy surgery (Fig. 6c). Postoperative X-ray showed that the neck rotational deformity was largely corrected (Fig. 5c, d). Postoperative CT axial slices showed that while the angle between the axial midline of the FM and C7 was unchanged, the angle between the axial midline of the FM was almost parallel with that of T1(Fig. 7).
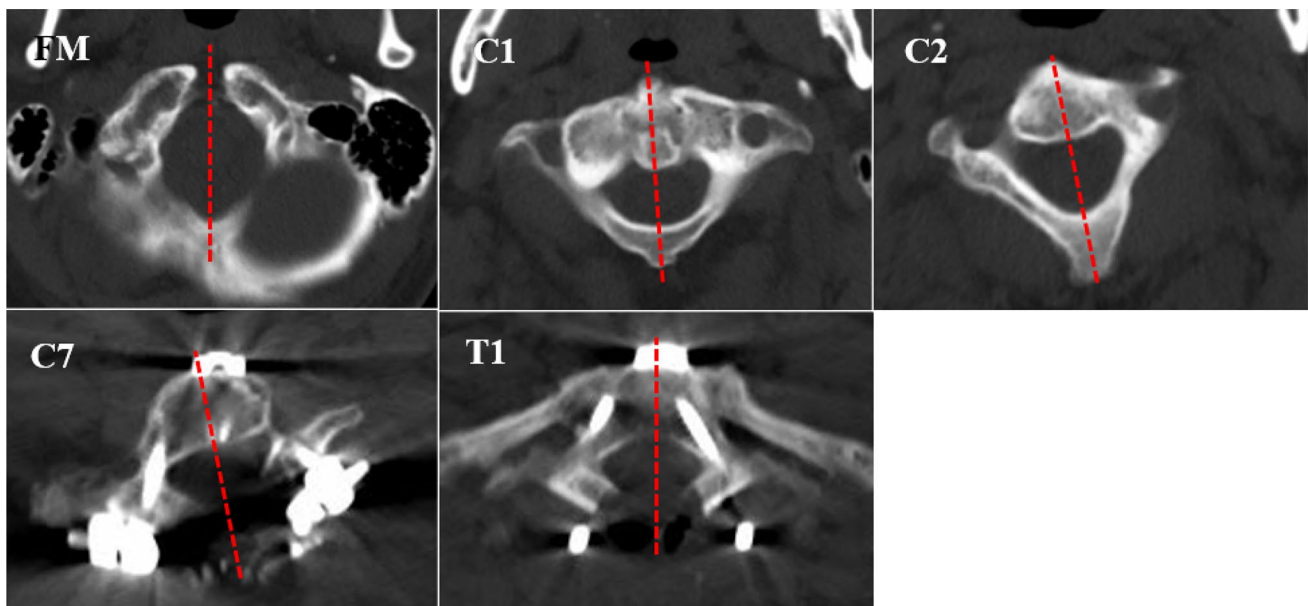
Postoperative course

It was reported that the patient had experienced left C8 nerve root paralysis after surgery. He displayed left upper-extremity pain and dysesthesias, as well as left hand grip strength weakness in the immediate postoperative period. These symptoms, however, improved after methylprednisolone and methycobal treatment, and he had fully recovered after

a period of 3 months. The patient was mobilized 2 days after surgery and wore a Philadelphia collar for 3 months. The patient was able to walk with his face and trunk frontward simultaneously and like a 'normal' person (see supplementary video 3). This clinical outcome was well maintained during the one-year and three-year follow-up (Figs. 8 and 9). Our reports showed that cervical axial rotation had not worsened at the three-year follow-up (Fig. 9).

Discussion

Whilst the correction of sagittal deformity in AS patients has been extensively studied [1, 2, 9, 10, 14], correction of axial malalignment is poorly reported. In a normal person, the spine is composed of multiple kinematic segments, comparable to that of a flexible hose. In the axial plane, the axial rotation of the spine reaches 90°–95° (averaging 60° [3], 25° [4], and 9° [5] in the cervical, thoracic, and lumbar spine, respectively). In slow walking, the pelvis and thorax rotate around the supporting leg [6]. However, in this AS patient, his spine resembled a rigid stick, and had lost most of this rotational function. To walk with his face forwards, he usually turned his body to the right, which results in asymmetric bilateral rotational axes. As a result, the patient tires easily and suffers hip pain after a long walk. After the index surgery, he was able to walk with his face and trunk facing forward simultaneously. Although he had lost most of the axial rotation of the spine, his pelvis and thorax were able to rotate around two symmetric instantaneous rotational axes

Fig. 7 The axial midline of the foramen magnum and T1 was almost paralleled postoperatively, so that axial rotation was largely corrected. FM: foramen magnum
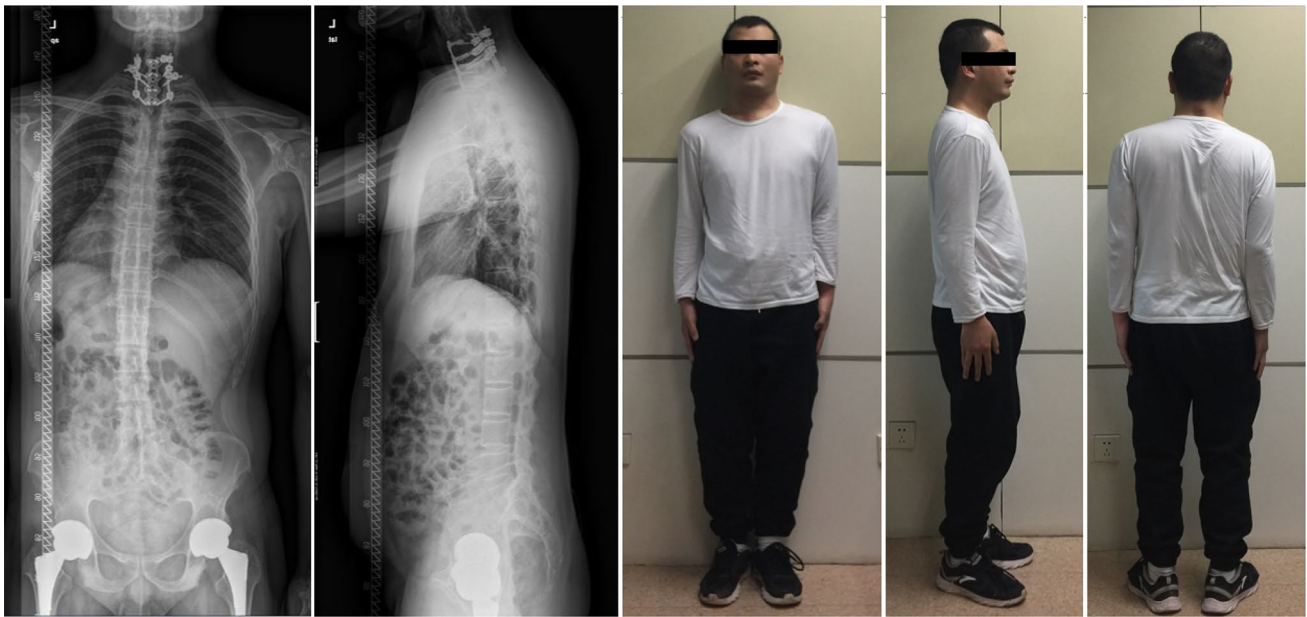
Fig. 8 Full-length spine plain radiographs and photograph at 1-year follow-up

Fig. 9 Full-length spine plain radiographs and photograph at 3-year follow-up

(the supporting leg). Thus, he was able to walk in a more normal manner, with less energy cost.

Cervical osteotomies are among the most difficult procedures to perform. The selection principles of the osteotomy site are feasibility, effectiveness, and safety. The use of three-column osteotomy in the cervical and upper thoracic spine for cervical deformity is extremely powerful, but it is associated with significant morbidity in the form of medical and

surgical complications, with mortality rates ranging from 1.3 to 6.7% and neurological complication rates ranging from 13.5 to 43.6% [7–10] including nerve root weakness, surgical site infection, dysphagia, and comorbid medical-related complications. In this case of cervical axial malalignment, osteotomy at the thoracic or lumbar segments would not correct his axial mismatch of his head and torso. Furthermore, because his axial deformity extends primarily from the occiput to C7, osteotomy at the completely ankylosed atlantoaxial segments cannot fully correct his deformity and poses a high risk of vertebral artery (VA) injury. Thus, we performed osteotomy at the cervicothoracic junction for the following reasons, the relatively large size of the spinal canal at C7/T1, mobility of the spinal cord and eighth cervical nerves in this region, maintenance of good hand function in the event of C8 nerve injury, and a fairly safe position of the VA in front of the transverse process of C7 prior to the artery entering the transverse foramen at C6 [11, 12]. Osteotomy at C7 is often preferred given the lower risk of devastating neurological injury and VA injury compared to more proximal cervical levels [13, 14]. Although we did not perform VA angiogram in this case, we can see his VAs are located within their respective transverse foramens at the level of C4-6 on cervical MRI, and are missing

in the transverse foramens at the level of C7(Fig. 10). Published data showed that abnormal VAs entrance is more often been seen at C3-C5 and minimal at C7. Bruneau et al. [15] reported that abnormal VAs entrance was observed in 7.0% cases, in comparison with a level of entrance into the C7 transverse foramen reported at 0.8%. Hong et al. [16] reported that atypical VA entrance was observed in 5.1% of cases, relative to entrance into the C7 transverse foramen being reported as 0.3% of cases. Shin et al. [17] also revealed that entry into the left of transverse foramen of C7 was only 0.2%. Despite the lack of previous cervical rotational correction reports, we used intraoperative monitoring in this case and manually rotated the patient's head to about 20° intraoperatively. We were unsure whether twisting his spine would result in nerve root or spinal cord injury after performing this maneuver. Probably due to the relatively large space available for the spinal cord in C7/T1 [18], no severe neurological deterioration occurred after surgery except for a mild left C8 paralysis.

Another limitation to this case is the short-segment fixation. Many would recommend that long-segment fixation may be necessary to counteract the long lever arms incurred by a rigid ankylosed spine [13, 19]. Long-segment screw/rod constructs offer stronger stabilization, thereby reducing the

Fig. 10 Axial slices of MRI showing vertebral artery course. Red arrows indicate vertebral arteries. His VAs are located within their respective transverse foramens at the level of C4-6 on cervical MRI, and is missing in the transverse foramens at the level of C7
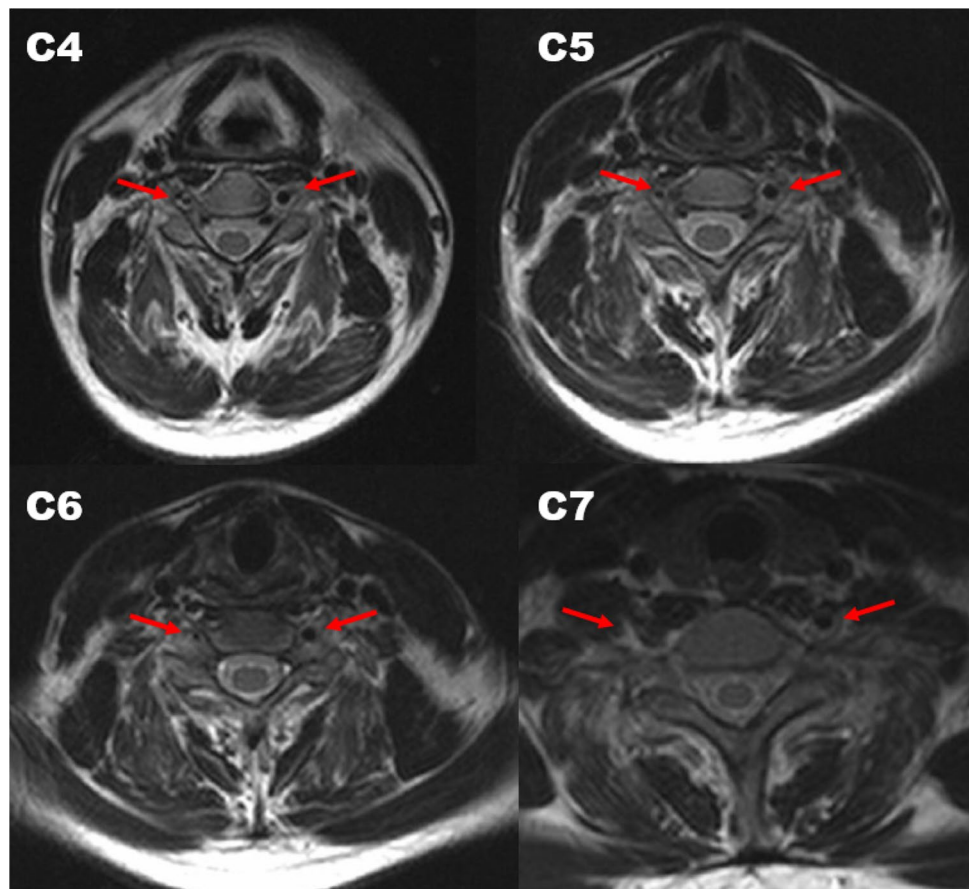
risk of postoperative subluxation or implant failures. This patient was only a young 24-year-old male at the time of surgery. We found that his bone quality was good intraoperatively. And we did not perform extensive osteotomy such as wedge osteotomy or complete vertebral column resection. Thus, we used fixations to extend 2 levels above and below the osteotomy site both anteriorly and posteriorly. Postoperatively, he was asked to wear a Philadelphia collar for 3 months. At the 3-year follow-up, his cervical alignment was well-maintained. However, a longer follow-up will be needed.

This is the first case report of cervical rotational osteotomy for correction of axial deformity. More case reports will be needed to finesse the technique and evaluate the clinical outcome of cervical rotational osteotomy. Through this case, we hope to draw the attention to spinal axial deformity and provide a reference in the surgical treatments of spinal axial deformity.

Compliance with ethical standards

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