

Case Report

Ankylosing spondylitis with chin-on-chest deformity combined with Anderson lesion as the apex of cervicothoracic kyphosis: case report

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Abstract: Ankylosing spondylitis patient with chin-on-chest deformity and Anderson lesion as the apex of kyphosis in the cervicothoracic region is rare, and there was no consensus on the best surgical strategy for it. Here we report such a rare case of a 28-year-old man, who presented with a history of unsatisfactory appearance, cervicothoracic kyphosis, neck pain (VAS = 7), difficulty in ambulation, and inability to maintain frontal gaze. On physical examination, the patient presented severe kyphotic deformity of the cervico-thoracic spine with inability to keep horizontal gaze, the chin-brow angle was 58°. We performed extension osteotomy combined with intralesional curettage of the inflammatory lesion. At one-year follow up, the patient got significant neck pain relief (VAS = 1), expressed satisfaction for the restoration of forward gaze, with grade one of Patient Satisfaction Index (PSI). In conclusion, for AS patients with chin-on-chest deformity and AL as the apex of kyphosis in the cervicothoracic region, extension osteotomy combined with intralesional curettage of the inflammatory lesion is an effective surgical strategy. Our data is of great value in decision making and surgical planning for both spinal surgeon and the patients.

Keywords: Anderson lesion, cervicothoracic kyphosis, ankylosing spondylitis

Introduction

Anderson lesion (AL), a localised vertebral or discovertebral lesion of the spine described by Anderson in 1937, is a known complication of ankylosing spondylitis (AS), with a reported prevalence of 1.5% to over 28% [1]. It usually involves only a single spinal level and the most common affected site is thoracolumbar junction. Up to now, there was no report of AS patient with chin-on-chest deformity and AL as the apex of kyphosis in the cervicothoracic region in the previous literature, with little knowledge about surgical strategy for it. Here we report the first rare case, and introduce the surgical strategy of extension osteotomy combined with intralesional curettage of the inflammatory lesion.

Case report

A 28-year-old man presented in April 2014 with a history of unsatisfactory appearance, cervicothoracic kyphosis, neck pain (Visual Analogue

Score, VAS = 7), difficulty in ambulation, and inability to maintain frontal gaze. On physical examination, the patient presented severe kyphotic deformity of the cervico-thoracic spine with inability to keep horizontal gaze, the chin-brow angle was 58° (**Figure 1**). Laboratory tests of Erythrocyte sedimentation rate and C reactive protein were 31 mm per hour and 8 mg/l, respectively. Blood counts and other serum chemistries were normal. Radiographic examination, including antero-posterior and lateral X-ray of cervico-thoracic spine in standing position and CT scan in supine position, demonstrated a severe sagittal deformity of cervico-thoracic junction, with kyphotic angle of C5-T1 was 54° and Anderson lesion (AL) within C6-7 disc space (**Figure 2**). The patient failed to undergo magnetic resonance imaging (MRI) scans of the CTJ region due to the constrained body posture.

The surgical procedure: Following the induction of general anesthesia, the patient was placed



Figure 1. The patient presented severe kyphotic deformity of the cervico-thoracic spine with inability to keep horizontal gaze and the chin-brow angle of 58° .

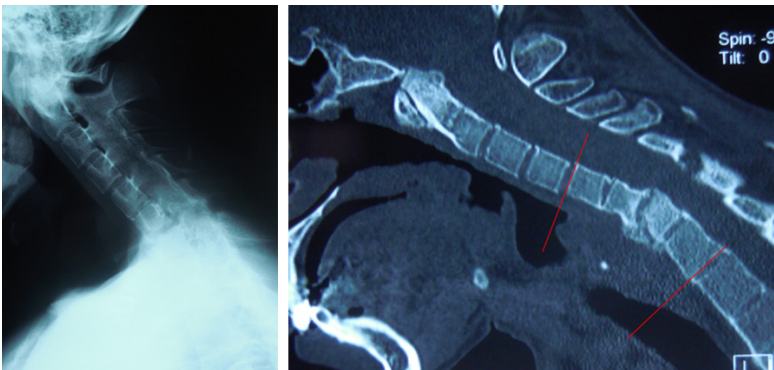


Figure 2. Lateral X-ray and CT scan demonstrated cervicothoracic kyphotic deformity with C5-T1 Cobb angle of 54° and Anderson lesion (AL) within C6-7 disc space.

in Mayfield pin fixation. Exposure was extended 3 levels above and below the intended osteotomy level (C6-7). We placed pedicle screws at

T1-3, and lateral mass screws at C3-5. Partial resection of the spinous processes and laminotomies of adjacent levels were performed, facetectomy was created bilaterally to provide adequate exposure of the 2 exiting nerve roots, laminectomy of C6 as well as part of C5 and C7 laminae were done to expose the dura sac completely. A temporary holding rod was placed on one side to prevent any inadvertent translation of the spine and perform a controlled correction (**Figure 3**). Intralesional curettage was done by forceps or curette, then the anterior osteoclasis was done by gentle elevation of the patient's head held in the Jackson table cranial fixation frame, which was accomplished manually by the assistant. The temporary rods were removed, and the permanent rods were then attached and locked into place. Further reduction was obtained using gentle compression across the osteotomy prior to screw tightening, the laminae of C5 and C7 were closed finally without kinking of dura sac (**Figure 4**). Autograft bone was placed across the closed osteotomy defect and over the length of the posterior instrumented levels, the wound was closed in multiple anatomical layers.

The patient was maintained with cervical collar until bony fusion was achieved and confirmed on CT scan at six months postoperatively

(**Figure 5**). At one-year follow up, the patient got significant neck pain relief (VAS = 1), expressed satisfaction for the restoration of forward gaze,

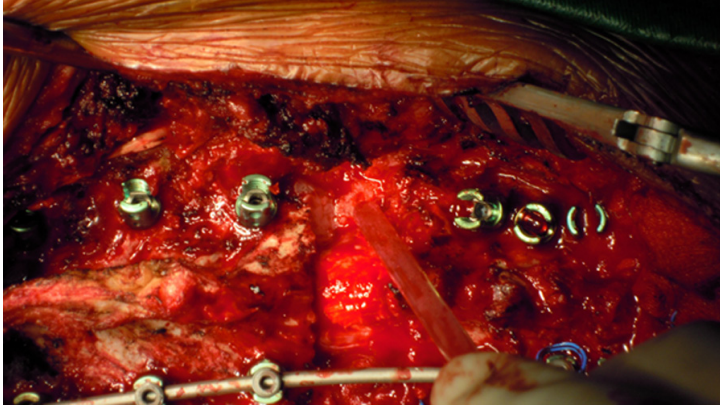


Figure 3. Laminectomy of C6 as well as part of C5 and C7 laminae, and facetectomy of C6-7 were done to expose the dura sac and 2 exiting nerve roots completely.

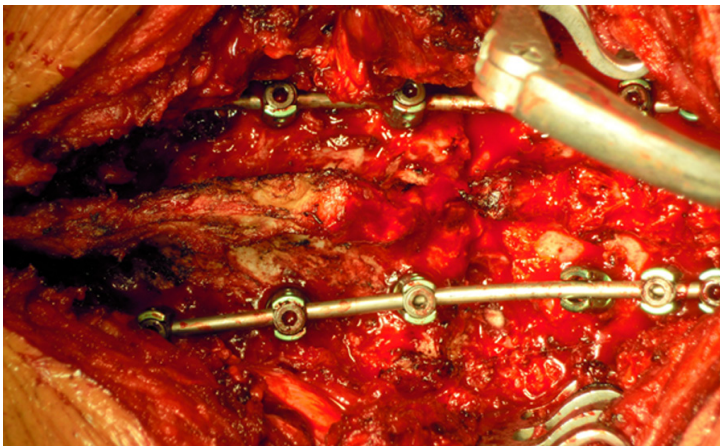


Figure 4. Further reduction was obtained using gentle compression across the osteotomy prior to screw tightening, the laminae of C5 and C7 were closed finally without kinking of dura sac.

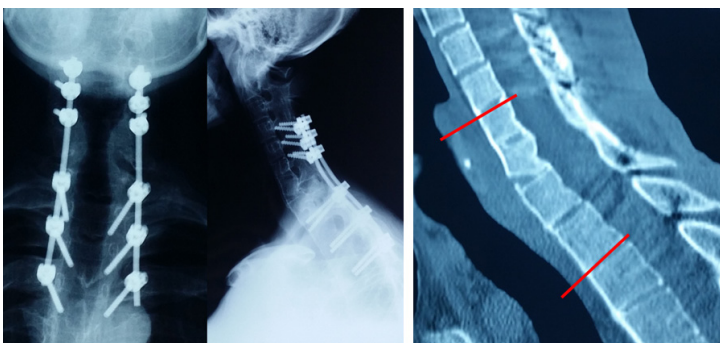


Figure 5. The X-ray and CT scan on six-month follow up demonstrate that solid bone fusion between posterior part of lower C6 endplate and upper C7 endplate has been achieved, bone fusion of intra-laminae between C5 and C7 has also been demonstrated.

with grade one of Patient Satisfaction Index (PSI), which means that “the surgery met my

expectations” [2] and the chin-brow angle of -8° (Figure 6).

Discussion

The AL is likely to be seen in place where the stress concentration is high, the most common affected site of AL in AS patients is thoracolumbar junction, while cervicothoracic junction (CTJ) is rare to be mentioned in previous literature [3-6]. The CTJ is a site of transition from a flexible, lordotic cervical spine to a rigid, kyphotic thoracic spine, and is also a stress concentration region. The CTJ kyphosis derived from AS is a variable local deformity with the most severe cases presenting as chin-on-chest deformity, and always results in problems of forward gaze, hygiene, chewing and swallowing, chronic neck pain, myelopathy, possible radicular symptoms, dysphagia and respiratory distress, with a dramatic reduction in quality of life [7, 8]. The specialty of our case is CTJ kyphosis and the AL appeared simultaneously in the same patient with AS, and the apex of the kyphosis is located at the level of the AL. The AL could be confined to the vertebral body (transvertebral), through the disc space (transdiscal), or both (discovertebral). In this patient with transdiscal lesion, both the X-ray and CT scan indicate that the disc space is conspicuously more radiolucent than its neighbor disc spaces which are fused.

Differing from the AS patients with CTJ deformity in previous literature, the patient in this case report demonstrate both CTJ kyphosis and the AL in C6-7 disc space, and the AL is revealed by osteolytic destruction with a surrounding zone of reactive sclerosis and vertebral osteophytes. Extension osteotomy is the predominant procedure in treating CTJ deformity in ankylosing spondylitis (AS). In 1958, Urist firstly described a patient with AS



Figure 6. The patient was very satisfied for the restoration of forward gaze and the chin-brow angle of -8° .

and a severe cervical kyphosis which was treated by anterior osteoclasia, also named as an extension osteotomy procedure. Modifications of the original technique developed continuously. The goal of all the surgical techniques is to correct the regional CTJ kyphosis, restore forward gaze, and reduce the pain associated with the deformity, with kyphotic deformity correction range from 13.7 degrees to 35.6 degrees [7-9] (Table 1).

The surgical strategy of this patient differs from the extension osteotomy technique reported in the literature, and the main differences are summarized as follows: First, C7-T1 is always selected as the ideal osteotomy site in the previous reports, for safety consideration, the vertebral artery (VA) remains anterior to the trans-

verse processes of C-7, while compromising the bilateral cerebral blood flow and kinking of the VA during osteotomy procedure can be avoided at T-1 or C-7. For the patient in this study, the C6-7, instead of C7-T1, is the apex of the kyphosis and presents with AL, preoperative vertebral artery CT scan demonstrated that the VA does not enter the transverse foramen until C-6, extension osteotomy within the C6-7 is safe, but without alternative. Second, extension osteotomy procedure alone is not enough for this patient, because the inflammatory lesion within the C6-7 disc space is supposed to be the main source of neck pain, which may not be relieved without intervention. The intralesional curettage was performed by forceps or curette to promote fusion, the CT scan on six-month follow up demonstrate that solid bone fusion between posterior part of lower C6 endplate and upper C7 endplate has been achieved, bone fusion of intra-laminae between C5 and C7 has also been demonstrated.

The clinical outcome of this patient is inspiring, as he has got significant kyphosis correction, maintained frontal gaze, expressed satisfaction about the appearance. However, the risk of spinal cord kinking, intraoperative sagittal translation, and postoperative instrumentation failure shouldn't be underestimated. First, laminectomy of C6 as well as part of C5 and C7 laminae, and facetectomy of C6-7 were done to expose the dura sac and 2 exiting nerve roots completely, which could provide enough room and decrease the risk of neurological complication. Second, in order to prevent intraoperative sagittal translation, a temporary holding rod was placed on one side to prevent any inadvertent translation of the spine and perform a controlled correction, and the anterior osteoclasia was done by gentle elevation of the patient's head held in the Jackson table cranial fixation frame, which

Table 1. Surgical strategy for CTJ deformity in the previous literature

N	Type	Case	Surgical option	CBVA	Correction	CTJ angle	Correction	Surgery time	Blood loss
1	Clinical article	8	Pedicle subtraction osteotomy	–	–	38.67°	35.63°	–	800 ml
2	Case report	1	Pedicle subtraction osteotomy	–	–	65.2°	21.9°	7 hours	2,700 ml
3	Database review	18	Posterior non-instrumented open-wedge osteotomy	43.1°	25°	2°	13.7°	–	–

was accomplished manually by the assistant. Third, the bone fusion of vertebrae and laminae in osteotomy space is the critical point to prevent postoperative instrumentation failure, and the patient should be immobilized with cervical collar until bony fusion was achieved, which was confirmed on CT scan at follow up.

In conclusion, AS patients with chin-on-chest deformity and AL as the apex of kyphosis in the cervicothoracic region is rare, extension osteotomy combined with intralesional curettage of the inflammatory lesion is an effective surgical strategy and the outcome is satisfactory.

Disclosure of conflict of interest

None.

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