Surgical treatment of a cervical spine fracture in an ankylosing spondylitis patient with severe global spine kyphosis and chin-on-chest deformity

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To the Editor: Kyphotic deformity is one of the main manifestations of ankylosing spondylitis (AS). Usually, the hyperkyphosis is distributed along the lumbar, thoracolumbar, and thoracic spine, since the ossification of the spinal column mainly starts at the sacroiliac joint and then progresses towards the head.[1] Occasionally, the kyphotic deformity occurs in the cervicothoracic spine, resulting in the chin resting on the chest, called chin-on-chest deformity. Patients with chin-on-chest deformity may have problems with horizontal gaze, chewing and swallowing, maintaining and performing proper personal hygiene, and they may suffer from neck pain and myelopathy.[2,3] However, severe global spine kyphosis in AS, resulting in a curvature of the upper body resembling the letter C, is an extremely rare condition with a simultaneous lumbar, thoracic, and cervicothoracic kyphosis. When cervical spine fractures happen in these patients, it is very challenging to deal with them, because of the severity of the deformity and the instability of the fracture. In this study, we report our experience in the treatment of a patient reporting these problems.

A 71-year-old woman arrived in our department 11 days after she fell while walking. She complained of neck pain (visual analog score [VAS] = 7), limited cervical mobility, and disability in chewing and swallowing. Her neck disability index (NDI) was 78%. She had a history of 40 years of AS and 20 years of severe global spine kyphosis. The physical examination showed a severe spine kyphosis with chin-on-chest deformity, neck tenderness, but without neurologic deficits. Computed tomography (CT) revealed a three-column fracture through the C7 vertebral body and C6 appendix. The fracture aggravated her pre-existing chin-on-chest deformity, and worsened the ability to chew and swallow. The whole spine X-ray images showed a lumbar kyphosis of 18° (L1-S1), thoracic kyphosis of 72° (T5-T12), and cervicothoracic kyphosis of 56.5° (on CT, C2-T4) [Figure 1A–E]. Her chin-brow vertical angle (CBVA) was approximately 130°, leaving her sitting even in a supine position. We placed a skull traction of 4 kg to relieve her neck pain. Since the patient and her family did not accept a conservative treatment and its neurologic risks, in which the skull traction and the custom-made halo-vest would last for several months, we decided to perform a posterior internal fixation and fusion surgery. After an awake nasotracheal intubation and general anesthesia, the patient was carefully placed in a sitting position with continuous skull traction [Figure 1F]. Intra-operative neurophysiologic monitoring was performed. A posterior approach was used and C2-C5 lateral mass screws and T1-T3 pedicle screws were placed before C6-C7 laminectomy decompression. Next, multiple layers of sheet were placed between the chin and chest to correct the kyphotic deformity and reduce the dislocation. When the intra-operative fluoroscopy confirmed that the dislocation was reduced and enough space between the chin and sternum was achieved (approximately 10 cm), the rod was placed and the screws were locked. The estimated blood loss was 400 mL. The patient recovered without complication. She was discharged at the 6th post-operative day wearing a soft collar. Post-operative CT scan showed that the cervical spine returned to the pre-injury alignment with a cervicothoracic kyphosis of 34.5° (C2-T4). At 3-month follow-up, the cervical spine fracture showed healing signs and no loss of deformity correction [Figure 1G]. The patient was satisfied with the treatment and returned to her pre-injury daily life (VAS = 0, NDI = 13%).

Cervical spine fractures are not uncommon in patients with AS. Of the 131 patients with cervicothoracic kyphosis reported by Simmons et al.,[23] 31% presented cervical spine fractures. However, such a severe deformity case as we reported is extremely rare. In addition to the severe chin-on-chest deformity caused by cervicothoracic kyphosis, the patient had a simultaneous global spine kyphosis with a
CBVA of 130°. Kim et al.[3] reported a case of AS treated with staged corrective osteotomy with a CBVA of 140°. The patient described by Kim et al.[3] underwent six surgeries within approximately 5 months. Cervical spine fracture combined with such a severe deformity represents a challenge for the doctors.

Anesthesia and surgical position: For patients with AS, most researchers choose endotracheal intubation under general anesthesia and prone position. However, for patients with chin-on-chest deformity and severe global kyphosis of the whole spine, endotracheal intubation is extremely difficult due to the inability to be in a complete supine position and limited mobility of the jaw and neck. Anesthesiologists usually choose awake nasotracheal intubation for these patients.[2,3] In case of intubation failure, local anesthesia can be used. Simmons et al.[2] applied local anesthesia with intra-venous sedation and a sitting position to treat cervicothoracic kyphosis in AS patients, achieving good results. Because the patient had a thoracolumbar kyphosis, a prone position on a straight table was almost impossible. Therefore, in our case, nasotracheal intubation was used and the sitting position was adopted [Figure 1F]. When using a sitting position, surgeons should be aware of the risk of air embolism. The case reported by Kim et al.[3] also used this method to perform cervicothoracic osteotomy.

Surgical strategy: Did this patient need osteotomy to correct the pre-existing deformity, or did she just need to resolve the fracture to restore the pre-injury status? Which approach should be chosen, anterior, posterior, or circumferential? C7-T1 extension osteotomy, C7 pedicle subtraction osteotomy, and closing-open wedge osteotomy were reported to treat chin-on-chest deformity, all achieving good results.[2,3,5] For patients with chin-on-chest deformity before injury, some authors believe that the fracture can be used to correct the pre-existing deformity if the patient’s physical condition allows this procedure and has the will of improving the physical appearance.[4] In this case, the fracture was well aligned and the patient had a good physical condition, so we chose to use the fracture to correct the deformity.

Figure 1: Patient’s appearance (A and B); Anterior-posterior position (C) and lateral position (D) of whole-spine X ray; (E) CT image showing the fracture through the C7 vertebra and C6 appendix (the red arrow showing the fracture and the yellow arrow showing there was no space between the chin and the chest); (F) Nasotracheal intubation and sitting position with skull traction; (G) CT image at 5-month follow-up (the red arrow showing the fracture had healed and the yellow arrow showing there was space between the chin and the chest). CT: Computed tomography.
In conclusion, in old patients with severe global spine kyphosis and chin-on-chest deformity combined with cervical spine fracture, the treatment strategy should be aimed at stabilizing the fracture and relieving symptoms, rather than correcting the deformity. In terms of treatment methods, nasotracheal intubation, a sitting position with skull traction, and posterior long segmental fixation should be adopted. The follow-up time of this patient is short, but long-term follow-up is needed.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be published. The patient understands that her name and initials will not be published and efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Conflicts of interest**

None.

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