Atlantoaxial rotatory subluxation in Eagle's syndrome: Is the styloid process protective?

ABSTRACT

A styloid process >3 cm is known as Eagle's syndrome (ES). This syndrome can lead to neurovascular symptoms. Traumatic atlantoaxial rotatory subluxation (AARS) is very rare in adults. We diagnosed AARS in a patient with ES after high-energy trauma. Posterior C1–2 stabilization was performed under traction. We wanted to discuss the mechanism of AARS in ES based on this case.

Keywords: Atlantoaxial, Eagle's syndrome, rotatory subluxation, stabilization

INTRODUCTION

Atlantoaxial rotatory subluxation (AARS) may develop due to rheumatoid arthritis, inflammatory process, and congenital or neoplastic causes.[1] Traumatic AARS in adults is extremely rare. Unlike children, AARS in adults occurs after high-energy trauma. The pathophysiology of AARS is not well defined.[2] The atlantoaxial joint contributes to approximately 50% of axial neck rotation. The transverse ligament and facet joint capsule prevent anterior translation of C1 over C2. The alar ligaments connect the posterolateral apex of the odontoid with the lateral aspect of the foramen magnum and generally limit anterior displacement of the atlas over the odontoid.[2,3]

A styloid process > 3 cm is known as Eagle's syndrome (ES). This syndrome can lead to neurovascular symptoms.[4] ES can cause ligament calcification, vertebral osteophytes, and vertebral enthesopathies.[5,6] We wanted to discuss the mechanism of traumatic AARS in a patient with ES.

CASE REPORT

A 23-year-old man is brought to the emergency department due to a nonvehicle traffic accident. His medical history does not include known ES or neurovascular symptoms. General condition is poor, unconscious, intubated, with a Glasgow Coma Scale score of 7. The investigations revealed a right clavicle fracture, lung contusion, and diffuse axonal injury in the brain. He was managed in the intensive care unit for an extended period. Two months posttrauma, with improvement in his general condition, the patient was transferred to the physical therapy clinic. Balance and in-bed exercises were initiated. The patient complained of neck pain, exhibited a Cook Robin position, and experienced restricted movement in the right upper extremity due to the right clavicle fracture. A computed tomography scan performed in response to these complaints revealed a 6-cm-long styloid process and C1–2 Fielding Type 1 AARS [Figure 1]. The patient underwent surgery under general anesthesia in the prone position with Gardner-Wells traction. Following a posterior occipitocervical skin incision, access to the vertebrae was achieved through posterior dissection. Posterior stabilization of C1–2 was performed utilizing bilateral C1 lateral mass and C2 pedicle screws. Early postoperative evaluation showed improvement

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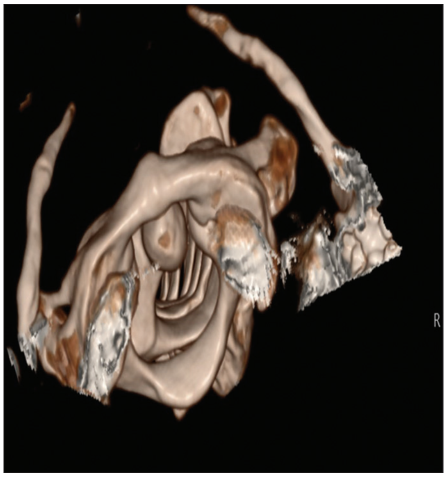

Figure 1: Three-dimensional computerized tomography; elongated styloid process and rightward subluxation of C1

in neck deformity. After adequate wound healing, the patient was transferred to the physical therapy clinic for further rehabilitation.

DISCUSSION

Mild neck extension produces a 30° angular movement at the tip of the styloid process.[4] The effect of the elongated styloid process on cervical vertebral biomechanics in ES remains unknown. Case studies involving ES, particularly with cervical vertebral pathologies, have been documented. These pathologies include cervical transverse ligament calcification, cervical osteophytes, diffuse idiopathic skeletal hyperostosis, and cervical disc herniation, often accompanied by neck pain.[4-8] In our case, AARS was detected alongside ES on cervical computed tomography, prompted by the presence of the Cook Robin position with neck pain. To our knowledge, this case represents the first reported instance in the literature.

In a study of 5 cases of ES, Ho *et al*. suggested that symptoms may be attributed to the proximity between the styloid process and the transverse process of C1.[9] The elongation of the stylohyoid process may represent an anatomical region in the cervical spine associated with enthesopathy.[6] Guo *et al*. proposed that the variable types of the stylohyoid process-stylohyoid ligament complex have a significant relationship with osteophytes and ligamentous osteophytes in the cervical spine.[7] However, in our case, no osteophytes or abnormal ossification were observed in the cervical region.

In our patient with ES following severe trauma, Fielding Type 1 AARS developed. It is plausible that fibrosis around the prolonged styloid process contributed to the low degree of subluxation. However, Miller pointed out that a prolonged calcified styloid process in ES might exacerbate the effects of hyperflexion/hyperextension (whiplash) injuries.[10] Green *et al*. reported an old fracture of the C6–7 spinous process and C6–7 disc degeneration in a patient with ES.[11]

Chronic instability at the craniovertebral junction is associated with musculoskeletal and structural changes in the spine. Chiari malformation, basilar invagination, syringomyelia, and Klippel–Feil, omovertebral bone anomalies are among the secondary changes that occur due to chronic atlantoaxial instability. Goel *et al.* have noted that all secondary changes at the craniovertebral junction, which are traditionally considered pathological and compressive/deforming, are actually protective in nature.[12,13] Therefore, an elongated styloid process in ES could be a protective secondary change for chronic atlantoaxial instability. Hence, further studies and biomechanical tests are necessary to determine whether a prolonged styloid process predisposes to or protects against traumatic AARS.

CONCLUSION

Traumatic AARS is extremely rare in adults. It usually occurs after high-energy trauma. ES is characterized by a styloid prominence >3 cm, potentially leading to neurovascular symptoms. In ES cases, cervical vertebral abnormalities such as transverse ligament calcification, vertebral osteophytes, and vertebral enthesopathies may be observed. Further studies and biomechanical tests are essential to determine whether the elongated styloid process predisposes individuals to or protects them from traumatic AARS.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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