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CASE REPORT

A case of fourth atlanto-axial facet joint: anatomical description

Xing-Wen Wang[1,2] · Feng-Zeng Jian[1] · Feng Ling[1]

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Abstract
Purpose To report a rare fourth atlanto-axial joint.
Methods A cadaveric specimen of a young male adult from occiput to C4 was dissected for anatomical study of craniocervical region. A true fourth atlanto-axial joint was confirmed. Its morphological characteristics were described.
Results The fourth atlantoaxial joint is rarely seen. The possible embryogenesis is discussed. This case raises an anatomical possibility of a new variant in this region.
Conclusions A unique case with an anatomically proven fourth atlantoaxial joint is reported. This anomaly can lead to misdiagnosis. CT scan coupled with MRI can facilitate accurate diagnosis.

Keywords Anomaly · Facet joint · Atlantoaxial

Introduction

The articulations between the atlas and axis vertebrae are composed by one midline atlanto-odontoid joint and two lateral atlantoaxial (AA) joints. Congenital osseous anomalies in this region, such as occipitalization of the atlas, os odontoideum and basilar invagination, can lead to an increased risk of segmental instability and neurological implications. To the best of our knowledge, there have been only two case reports of a fourth AA joint confirmed during an operation [1, 2]. In this report, we present a true accessory facet joint between the posterior C1 arch and C2 lamina during an anatomical study of the lateral AA joint. The possible embryogenesis of this anomaly and its clinical significance are discussed in light of the literature.

Anatomical report

A fresh cadaveric specimen consisting of skull base and C1 to C4 was offered by the Department of Anatomy in Southern Medical University. The specimen was of a Chinese young male adult. An anatomical and biomechanical study was conducted for the lateral AA joint. The skin was removed first, and the posterior cervical muscles were separated in layers until the occiput, posterior arch of C1 and lamina of C2 had been exposed. At this stage, the surgical microscope was introduced (magnification 5–10×). A subperiosteal dissection was performed along the posterior arch of C1 to the lateral mass to expose the vertebral artery. Then dissection was continued along the posterior lamina of C2 to expose the lateral AAJ. The C2 nerve root on the right was normal as it coursed laterally between the posterior C1 arch and the C2 lamina. But on the left side the C2 nerve root could not be confirmed. After the joint capsule was opened, an extremely small facet joint on the left side was demonstrated compared to that on the right. At this point, the possibility of a congenital anomaly of the facet joint was proposed. Then all the muscles were removed completely. The oropharynx and the longus capitis muscles were transected. The anterior longitudinal ligament between the C1 and C2 was cut. The transverse ligament of the midline atlanto-odontoid

✉ Feng Ling
fengl15210027143@yeah.net

[1] Department of Neurosurgery, Xuanwu Hospital, Capital Medical University, 45 Changchun St, Beijing 100053, People's Republic of China

[2] Department of Neurosurgery, Beijing Hospital, Beijing 100730, People's Republic of China

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joint was cut via the foramen magnum from the superior direction, and the tectorial membrane was cut from the posterior direction. Then the joint capsule of the lateral AA joint was cut with caution not to damage the joint space. At this point, the atlas was separated completely from the axis (Fig. 1).

The fourth AA joint was between the posterior C1 arch and the C2 lamina on the left side. It was situated at the pars interarticularis. This joint was parallel to the lateral AAJ with synovium in place of a typical intralaminar space. This fourth AAJ was oval in shape and smaller than the lateral AAJ. Normal endplate cartilage and joint capsule were noted. The left C2 nerve root exited between the lateral AA joint and the fourth AA joint. No abnormality was observed of the lateral AA joints and odontoid process. No arthritis or joint degeneration was confirmed.

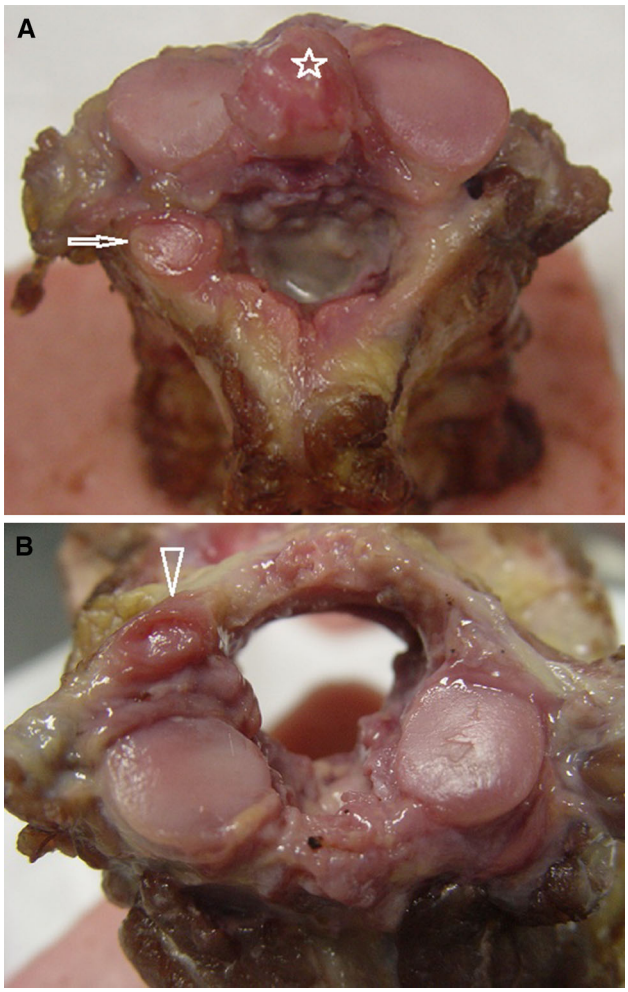

Fig. 1 **a** Superior view of C2 showed that the inferior articulation of the fourth AAJ situated posterior to the lateral AAJ on the left side (*white arrow*). Odontoid process was normal (*star*). Joint surface of the fourth AAJ was flanked with smooth endplate cartilage as the lateral AAJ. The C2 nerve root exited between the lateral AAJ and the fourth AAJ. **b** Inferior view of C1 showed the superior articulation of the fourth AAJ situated at the inferior margin of C1 posterior arch (*arrow head*). *AAJ* atlanto-axial joint

Discussion

The existence of a fourth AA joint is a rare congenital anomaly. Its embryological pathogenesis is unclear. The formation and development of the spine takes place by 4-week gestation. When errors in cellular proliferation, migration, differentiation and segmentation occur, congenital osseous anomalies develop. During the process of C1–C2 segmentation, the dense zone of lateral C1 sclerotome forms the posterior arch and lateral masses of C1; the hypochordal bow forms the anterior C1 arch; and the dense zone of lateral C2 sclerotome forms the C2 lamina and superior articular processes [3]. A possible explanation for this fourth AA joint anomaly is a normal segmentation of the C1–C2 sclerotomes followed by a predifferentiated group of cells forming an interzone just posterior to the normal lateral AA joint, which in turn lead to the formation of a true accessory synovial joint.

The accessory articulations are commonly reported in the sacroiliac region. In the craniocervical region these have been reported in the midline atlanto-occipital joint [4]. Osteoarthritis was speculated to play a role in the formation of osseous contact zone and hyaline cartilage between the basiocciput, atlas and axis. Salunke et al. reported pseudofacet joint formation between C1 and C2 in some cases of congenital AA dislocation [5]. Genetic aberration of HOX genes and abnormal movement at C1–C2 were hypothesized for the accessory joint formation. In our case, no joint degeneration or osteophyte was noted. The surface of the joint was flanked with smooth endplate cartilage and the accessory joint was surrounded by joint capsule. The aforementioned facts suggested that it was a congenital true facet joint. It is unlikely to be a result of osteoarthritis or acquired contact area between C1 and C2. Only two other cases of a fourth AAJ with similar characteristics had been reported in the literature [1, 2]. These two young patients suffered from dorsal compression of the spinal cord. Their symptoms relieved after the resection of the abnormal joint. The fourth AAJ was situated posterior to the C2 pars interarticularis. The hyaline cartilages covering the joint surface were identified, just like that seen in our case.

Congenital facet disorders are typically asymptomatic. These anomalies may be incidentally noted during a routine radiographic examination. However, AAJs account for nearly 60 % of the rotations in the cervical spine [6]. It is conceivable that this fourth AAJ was associated with altered load bearing at the AA region on account of the increased contact surface area. With the

stability between C1 and C2 increased, the shearing force could be transferred to the subaxial cervical spine. Subsequent cervical neuralgia and degeneration could be possible. If spinal cord compression caused by the accessory joint hypertrophy was present, surgical removal may be needed.

Because of its rarity, this anomaly has a high potential for radiographic misinterpretation and misguided clinical intervention. In the past 10 years, direct posterior reduction and fusion technique has been used widely for the treatment of AA dislocation. This fourth AA joint anomaly could block the surgeon's vision of the true lateral AA joint and cause confusion during C2 pedicle screw insertion.

The bony and neural anatomical anomalies in this case were noted during our cadaver dissection. Although the present report is limited by the absence of radiological and histopathological evaluation, it raises an anatomical possibility of a new variant in this region that needs to be taken into consideration during surgical interventions.

In conclusion, we report a unique case with an anatomically proven fourth AA joint for the first time. Young spine surgeons unfamiliar with this anomaly may misdiagnose it as an osteoma, fracture or other anomaly. CT scan coupled with MRI can facilitate accurate diagnosis. This anomaly should be kept in mind whilst operating in the craniocervical region.

Compliance with ethical standards

Conflict of interest The authors have nothing to declare. No funding or grants were provided for this report. The authors did not receive any outside payments or other benefits. The authors have full control of all primary data which could be reviewed by the journal if requested.

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