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Occipitoatlantal instability associated with articular tropism

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Abstract The authors report the case of a patient with rotatory posterior subluxation of the occiput on the atlas associated with tropism of the O-C1 articulations. Lateral flexion-extension plain films demonstrated 5 mm of posterior translation of the occiput on the atlas. Tomographs revealed tropism of the O-C1 articulations, and CT scans on extension demonstrated posterior rotatory subluxation of the occiput on the atlas. The patient had no osseous abnormality caudally below the atlantoax-

ial joint, and underwent posterior occipitoatlantal fusion by wiring. At the 4-year follow-up, the grafted bone had been incorporated, and the patient was completely free from preoperative symptoms. In the present patient, occipitoatlantal instability is presumed to have derived from articular tropism.

Key words Occipitoatlantal joint · Instability · Spinal tropism · Posterior fusion

Introduction

Non-traumatic occipitocervical instability has rarely been reported. Wiesel and Rothman described two patients with occipitoatlantal hypermobility associated with congenital fusion of the upper part of the cervical spine [15]. Georgopoulos et al. described occipitoatlantal instability in five patients [7]. The etiology of the occipitoatlantal instability in their patients was injury in three patients, congenital anomaly of the occipital condyle in one, and unknown in one. Harris et al. demonstrated significant instability at the occipitoatlantal articulation in one patient with Klippel-Feil syndrome [9]. Uno et al. reported that occipitoatlantal hypermobility was observed in Down's syndrome, but only when atlantoaxial instability was present [12]. Facet tropism is defined as asymmetry between the left and right facet joint angle [2]. The present paper discusses the case of a 28-year-old man who suffered symptoms caused by occipitoatlantal instability presumed to have been derived from tropism of the occipitoatlantal articulations.

Case report

A 28-year-old man was referred to our hospital with history of progressive neck pain, vertigo, nausea, and tightness of the chest. The patient had no history of trauma. Physical examination of the neck revealed that forward bending of the cervical spine was restricted by neck pain. Neurologic examinations revealed no abnormalities except hyperreflexia of deep tendon reflexes at the upper and lower extremities. Plain films by lateral projection demonstrated excessive motion from maximum flexion to maximum extension of 5-mm posterior horizontal translation of the occiput on the atlas, based on the Wiesel's measuring procedure (Fig. 1) [15]. In contrast, rotatory angular motion of the occipitoatlantal segment in the sagittal plane was within normal range [14]. Antero-posterior tomographs revealed asymmetry of the left and right occipitoatlantal articulations. The angle of the O-C1 joints relative to the horizontal plane was about 25° at the left side and 45° at the right at the center of the joints (Fig. 2). Lateral tomographs in extension and neutral position showed flattening of the posterior portion of the left joint surface of the atlas (Fig. 3A), and demonstrated posterior subluxation of the occiput on the atlas in extension and its reduction in neutral position (Fig. 3A, B). In addition, CT scans, taken with the cervical spine in extension, demonstrated posterior translation of the occiput on the atlas associated with axial rotation (Fig. 4). In contrast, radiological examinations revealed no osseous abnormalities or abnormal segmental motion in the atlantoaxial joint

Fig. 1 Plain films in **A** maximum extension and **B** in maximum flexion by lateral projection demonstrating 5 mm of posterior horizontal translation based on the Wiesel's measuring procedure

Fig. 2 Angle of the O-C1 joints relative to the horizontal plane was more horizontally oriented at the left side than at the right side

Fig. 3 Lateral tomographs in flexion and neutral position showed flattening of the posterior portion of the left joint surface of the atlas (**A**), and demonstrated posterior subluxation of the occiput on the atlas in extension (**A**) and its reduction in neutral position (**B**)

Fig. 4 CT in flexion (**A**) shows normal relationship between the occiput and the atlas. In contrast, CT in extension (**B**) shows posterior translation with coupled axial rotation of the occiput on the atlas

Fig. 5 Four years postoperatively, plain film by lateral projection demonstrating the solid posterior occipitotlantal fusion

and the subaxial cervical spine. Angiograms of the vertebral artery demonstrated no abnormal findings.

The symptoms of the patient were improved by application of a Philadelphia collar indicating that surgical stabilization could improve occipitotlantal instability and relieve the symptoms. The patient underwent posterior occipitotlantal fusion using wiring and iliac bone grafting. Double wires were passed through the occiput and the posterior arch of the atlas bilaterally. After the placement of monocortical iliac bone between the occiput and the atlas,

the wires were tightened. His consistent symptoms disappeared completely postoperatively. The patient wore a Philadelphia collar for 8 weeks postoperatively, and solid fusion was obtained 3 months after surgery. At the final follow-up, 4 years postoperatively, the patient was completely free from preoperative symptoms, and lateral plain films demonstrated solid bony union of posterior occipitotlantal fusion (Fig. 5).

Discussion

There are several conceivable etiologies of occipitotlantal instability including injuries, congenital anomalies, spinal tumors of the occipital condyle or atlas, Down's syndrome, and rheumatoid arthritis. According to previously reported studies, occipitotlantal instability related to Down's syndrome, rheumatoid arthritis, or tumor of the atlas is always associated with atlantoaxial instability [8, 12, 13]. However, reports of an isolated instability of the occipitotlantal junction without atlantoaxial instability

have been rare, except for several reports of traumatic occipitoatlantal dislocation [5, 6, 9, 11, 15]. Wiesel and Rothman and Harris et al. reported one case each of occipitoatlantal instability secondary to congenital fusion of the upper part of the cervical spine [9, 15]. Wiesel and Rothman postulated that abnormal stress on the ligaments of long duration may result in excessive motion of the occipitoatlantal joint. Georgopoulos et al. reported on one patient with congenital enlargement of the occipital condyles who presented with excessive motion of the occipitoatlantal joint [7].

In the present report, the patient showed tropism of the occipitoatlantal articulations and flattening of the posterior portion of the left joint surface of the atlas. Those osseous abnormalities were presumed to be one of the causes of abnormal rotatory translation of the occiput on the atlas in extension. It is difficult to distinguish whether the osseous abnormality is congenital or acquired. However, relatively young age and no history of trauma or other lesions in this patient indicate that the congenital osseous abnormalities were probably a cause of instability at the occipitoatlantal motion segment. It was impossible in the present report to demonstrate abnormality of the ligaments important for stability of the occipitoatlantal joint, such as the tectorial membrane, alar ligament, apical ligament, etc. However, the authors presume that osseous abnormalities at the cranio-cervical junction in the present patient had produced abnormal stress on those stabilizing ligaments, which resulted in ligamentous insufficiency and led to abnormal mobility of the occipitoatlantal motion segment.

Biomechanical studies in vivo and in vitro showed that there is relatively large rotatory motion in flexion-extension, but there is almost no anterior or posterior translation in flexion-extension motion [14]. Wiesel and Rothman stated that, at the occipitoatlantal joint in a normal individual, there is 0–1 mm of horizontal translation from maximum flexion to maximum extension [15]. They advocated that movement greater than 1 mm was clinically significant. Flexion-extension films in lateral projection were useful to evaluate the occipitoatlantal stability in the present report. Nevertheless, it is impossible to demonstrate the coupled rotatory motion by this procedure. CT scan in extension showed the coupled rotatory motion to posterior translation in the present report.

Regarding the lumbar spine, there have been several studies that referred to the relation between facet tropism and intervertebral disc diseases [1, 3, 4, 10]. In contrast, there have been no reports related to tropism of the occipitoatlantal articulation and intersegmental instability. In normal subjects, the joint surfaces of the atlas are cup shaped, with the arcuate occipital articulation fitting into the cup of the atlas. This characteristic osseous anatomy of the occipitoatlantal articulation seems to limit anterior-posterior translations and axial rotation of the occiput on the atlas. The patient in this report had abnormalities in the posterior portion of the left joint between the atlas and the occipital condyle including flattening of the left joint surface of the atlas with loss of its posterior edge, which permitted the excessive posterior translation with coupled axial rotation of the occiput on the atlas. In other words, facet tropism of the occipitoatlantal joint was presumed to be one of the causes of excessive posterior translation associated with axial rotation.

The clinical manifestations of occipitoatlantal instability vary widely. The symptoms of the present patient resembled those caused by insufficiency of the vertebral artery. However, angiograms did not reveal any abnormal findings. Therefore, those symptoms may have been caused by dynamic compression of the medulla oblongata by excessive occipitoatlantal motion.

In the present patient, intersegmental instability and anatomical abnormalities were limited only to the cranio-cervical junction, and there were no mechanical or anatomical abnormalities in the atlantoaxial complex. Simple posterior fixation by wiring limited to the occipitoatlantal segment was conducted, and resulted in successful fusion with complete resolution of symptoms.

Conclusion

Isolated occipitoatlantal instability, excepting that caused by trauma, is a rare condition. In the patient of this report, tropism of the occipitoatlantal articulation was presumed to be one of the causes of occipitoatlantal instability indicated by rotatory posterior translation of the occiput on the atlas. The patient was successfully managed by posterior occipitoatlantal fusion with wiring.

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