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SHORT REPORT



Vertebro-basilar stroke due to Bow-Hunter syndrome: an unusual presentation of rotatory atlanto-axial subluxation in a fourteen year old

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ABSTRACT

Bow Hunter's syndrome is a rare disorder usually producing transient ischemic symptoms as a result of dynamic compression of the vertebral artery during head turning. We report a case of a 14 year old male presenting with stroke due to occlusion of vertebral artery due to rotatory atlanto-axial subluxation. The patient presented with sudden onset vertigo and ataxia. History revealed led mild torticollis since childhood which was never investigated. MRI and MRA showed infarcts in the bilateral cerebellar hemispheres and the occipital lobes with a hypoplastic left vertebral artery and kinking of the right vertebral artery at the cranio-vertebral junction due to rotatory atlanto-axial subluxation. The patient was successfully treated by C1 lateral mass and C2 sub-facetal screw with rod fixation. Bow-Hunter's syndrome producing transient ischemia is well reported but stroke in the vertebro-basilar territory in a 14 year old due to rotatory atlanto-axial subluxation is uncommon, and to the best of our knowledge, this is the eighth such reported

ARTICLE HISTORY

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KEYWORDS

Bow Hunter syndrome: rotatory atlanto-axial subluxation; vertebral artery hypoplasia; posterior circulation stroke

Introduction

Bow-Hunter syndrome (BHS), also known as rotational vertebral artery compression syndrome, is a rare disorder, characterised by ischemic symptoms due to vertebro-basilar insufficiency as a consequence of dynamic vertebral artery compression during head turning.

We report a case of vertibro-basilar stroke, as result of rotatory atlanto-axial subluxationleading to Bow-Hunter's syndrome.

Case presentation

A 14-year-old male presented with sudden onset vertigo and imbalance while walking. Examination found slurring of speech, severe ataxia, and nystagmus with normal vision, power and sensations. He had a mild torticollis present since childhood, but never investigated. MRI of the brain revealed areas of diffusion restriction in the bilateral cerebellum and occipital lobes (Figure 1). MR angiography revealed a hypoplastic left vertebral artery and kinking of the right vertebral artery near the craniovertebral junction (Figure 2, 3). CT and MRI scan of the cervical spine revealed rotatory subluxation of the right atlanto-axial joint with aplasia of the right C1 arch and occipitalisation of the left C1 arch (Figures 4 and 5). He was treated by C1 lateral mass to C2 subfacetal screw and rod fixation.

He required tracheotomy on the second postoperative day, and Ryle's tube feeding was instituted. He was mobilised and was able to walk with support from the third day. He gradually improved and was able to walk with minimal support over the next 3 weeks. Tracheostomy and Ryle's tube were removed on the 20th postoperative day.

Discussion

Bow-Hunter syndrome was first described in 1978 by Sorenson¹ in a bow hunter, derives its name from the position a hunter takes while aiming. This syndrome usually produces transient ischemic symptoms as a result of impaired flow in the vertebrobasilar complex, Jost and Dailey² in their analysis of 126 patients with BHS reported C1-C2 as the site of occlusion in 36% while Rastogi et al.3 in a review of 153 patients reported the atlantoaxial (C1-C2) joint as the site of involvement in 69% (99 patient out of 142 patients).

The cranio-vertebral junction appears more prone to rotational occlusion of vertebral artery because the rotation of the atlas-occiput complex over the axis, which is about 30-40 degrees, places a torsional force on the vertebral artery. In a normal person this temporary reduction of blood flow through one vertebral artery is without any clinical consequence, because the contralateral flow through the opposite vertebral artery and the anterior circulation maintains blood flow to the brain. However, when one of the vertebral artery is aplastic, like in this case, any compromise in the blood flow through the normal vertebral artery may lead to ischemia in the vertebrobasilar territory.

The rotatory atlanto-axial dislocation in this case produced a "fixed" kinking of the right vertebral artery, which in the absence of the left vertebral artery, produced infarcts in the cerebellum and occipital lobes. Such a presentation of cranio-vertebral junction anomaly with a vascular event is uncommon, with only eight cases reported till date.4-10

Rotatory atlanto-axial dislocations may present with a "cockrobin" position of the head. In pediatric age group they typically occur after an upper respiratory tract infection, called Griesel's

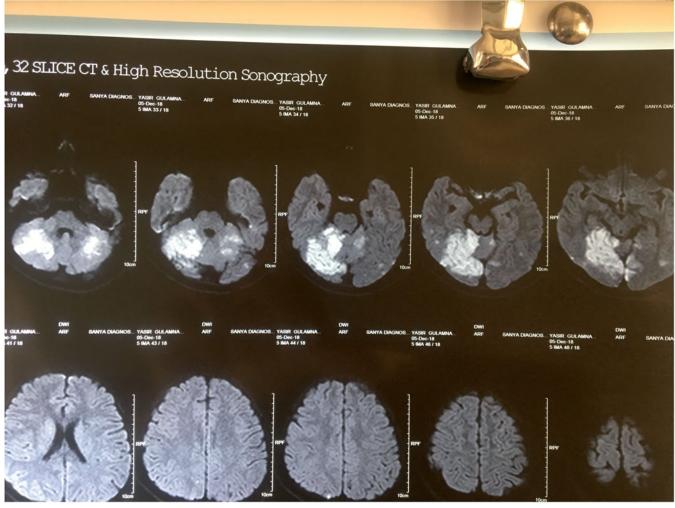


Figure 1. MRI brain (diffusion weighted) showing bilateral cerebellar and occipital infarcts.



Figure 2. MR Angiography showing kinking of right vertebral artery (red arrow) and hypoplastic left vertebral artery.

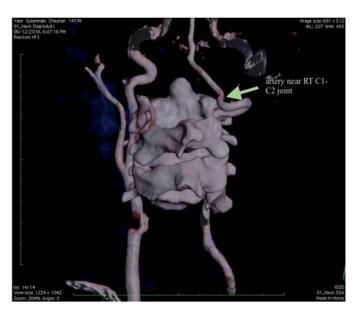


Figure 3. Showing the right vertebral artery near the right C1–C2 joint.

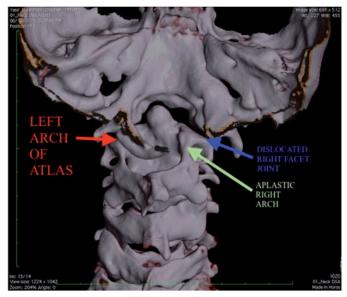


Figure 4. Showing the bony anatomy. Left occipitalised arch of atlas(red arrow), dislocated right atlanto axial joint (blue arrow), hypo plastic right C1 arch (green arrow).



Figure 5. MRI axial cut showing the rotated C2.

syndrome⁽¹¹⁾ from leading to effusion and laxity of the atlantoaxial joints.

Our case is an example of a rare presentation of a congenital cranio-vertebral junction anomaly with vertebro-basilar infraction.

Conclusion

Posterior circulation stroke or symptoms of vertebro-basilar insufficiency in young should be investigated for cranio-vertebral junction anomaly.

Disclosure statement

No potential conflict of interest was reported by the author.

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