

Delayed diagnosis of isolated alar ligament rupture: A case report

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Abstract

Ligament disruptions at the craniovertebral junction are typically associated with atlantoaxial rotatory dislocation during upper cervical spine injuries and require external orthoses or surgical stabilization. Only in few patients isolated ruptures of the alar ligament have been reported. Here we present a further case, in which the diagnosis was initially obscured by a misleading clinical symptomatology but finally established six months following the trauma, demonstrating the value of contrast-enhanced high resolution 3 Tesla magnetic resonance imaging in identifying this particular lesion.

Key words: Alar ligament rupture; Cervical spine injury; Contrast-enhanced magnetic resonance imaging

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Core tip: Upper cervical spine injuries are common and bear a relevant medical and socioeconomic impact. While most of such lesions are related to atlantoaxial rotatory dislocation, thus far only few patients with isolated alar ligament ruptures have been reported. This particular trauma is a challenge to both clinicians and radiologists and diagnosis might thus be delayed. Here we present a further case of a young adult and discuss the value of sequential contrast-enhanced magnetic resonance imaging in establishing this diagnosis at a late stage and in the follow-up of a subsequently prolonged recovery.

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INTRODUCTION

Upper cervical spine injuries are common and mainly caused by car and sport accidents or falls. They frequently are associated with long-term impairment or work disability of involved individuals and bear a relevant medical and socioeconomic impact^[1-4]. Particularly, cases with hyperextension and rotation of the neck may eventually result in ligament ruptures, though this incident is not necessarily correlated with the intensity of the trauma^[5,6]. While most of such lesions are related to atlantoaxial rotatory dislocation, thus far only few patients with isolated alar ligament ruptures have been reported^[7]. Probably these cases are underdiagnosed, since they might be missed on initial presentation and only be identified in the context with persistent cervical instability. Here we present a case of a young adult and discuss the value of sequential contrast-enhanced magnetic resonance imaging (MRI) in establishing this diagnosis at a late stage and in the follow-up of a subsequently prolonged recovery.

CASE REPORT

A 25-year-old man was diagnosed with rupture of the tympanic membrane of the left ear following a blunt fist hit trauma to the left side of his head associated with a short period of retro- and ante-grade amnesia, hearing impairment and tinnitus. While topical membrane patching led to complete healing with reestablishment of hearing, ipsilateral tinnitus remained and was accompanied by intermittent occipital pain. Moreover, during the following weeks and after repetitive sport exercises including headstands the patient developed further symptoms such as projecting aching in both shoulders, neck stiffness, dysphagia, fasciculation predominantly in the left arm, paraesthesia along the thoracic spine, neuralgiform pain attacks in the chest, few episodes of unexplainable shivering without fever and some other vague symptoms. On repetitive neurological examination there was no evidence of any objective deficits apart from an impaired neck rotation to the left side. A plain MRI excluded cervical disc herniation and except a straightening of the cervical spine reported no otherwise pathology. Mobilization and physiotherapy was advised leading to further exacerbation of symptoms. After a chiropractic manoeuvre attempted by an orthopaedic surgeon the tinnitus was felt louder and of higher frequency. An additional osteopathic treatment with repetitive sessions of rotational overstretching of the cervical spine above the tolerable pain threshold further aggravated the symptomatology. Recalling the earlier blunt injury a traumatologist was finally consulted 6 mo after the initial event who disclosed a pathologic cervical hypermobility on rotating the neck to the

right site, which was considered highly suspicious of a ligament lesion within the atlantoaxial joint. At this time a computed tomography (CT) scan ruled out a fracture or atlanto-occipital dislocation but revealed a slight shift of the dens towards the right lateral mass of C-1 (Figure 1). An MRI (Magnetom Prisma 3T, Siemens Healthcare) confirmed loss of lordosis on sagittal plane while the contrast-enhanced axial T1-weighted sequences disclosed increased signal intensity within the apex of the dens as well as within the widened left lateral dens-atlas space indicative of edema. Moreover, the dark signal of the left alar ligament proved to be interrupted (Figure 2), whereas the tectorial membrane and transverse ligament as well as the spinal cord appeared intact. Taken together, the findings were suggestive of an isolated rupture of the left alar ligament.

Subsequently, the cervical spine was immobilized by means of a Philadelphia collar, leading to a partial relief of the symptoms. Follow-up MRI of the cervical spine 3 mo later still showed signal hyperintensity within the alar ligament and the apex of the dens while its deviation apparently almost had resolved. After allowing for less cervical immobilization using a soft collar the patient's complaint worsened again and a subsequent MRI two months later still confirmed hyperintense signalling of the involved ligament and dens. The Philadelphia collar was reintroduced and following another 3 mo of immobilization a 3rd MRI sequence showed marked improvement (Figure 3). The patient gained a progressively increasing range of neck motion in each plane and was nearly free of any discomfort except a feeling of cervical tension increasing during the day, sporadic periods of head and chest pain and persistent tinnitus of varying intensity.

DISCUSSION

Isolated unilateral alar ligament rupture is a diagnosis made by excluding associated dislocation, fracture, or disruption of other ligamentous structures in the craniovertebral junction. Only recently Wong *et al*^[7] emphasized the special anatomical and pathophysiological aspects of this particular trauma and the value of CT and MRI to confirm the diagnosis discussing a 9 years old girl and reviewing 6 additional cases from the literature aged between 5 and 21 years, all of which fully recovered after conservative immobilization therapy within 1 year^[7].

While cervical X-ray, CT and MRI (T2-weighted and STIR sequences) of spinal ligamentous and soft tissue trauma are normally initiated in an acute setting^[8,9], in some cases appropriate imaging of cervical injuries might be missed initially or performed with delay for a variety of reasons^[10-12]. Also in our patient an appropriate diagnostic work-up had finally been delayed for several months, since the initial trauma and symptoms were apparently considered inadequate for further imaging analysis. MRI studies of patients with suspected occult cervical injury are well established to detect ligamentous



Figure 1 Multidetector-computed tomography coronary construction. Verification of a relationship of dens axis, atlas and occipital condyles. Asymmetry of the denso-axial joint space between left and right (white arrows) with widening on the left side. No micro- or macro-fracture.

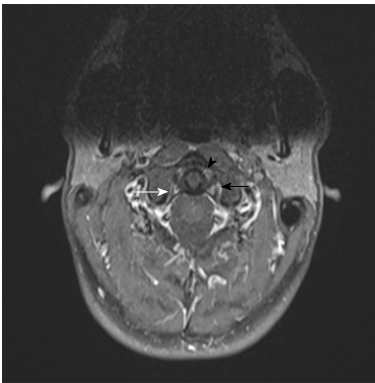


Figure 2 High resolution contrast-enhanced 3T magnetic resonance imaging, fat-saturated gradient echo sequence performed 6 mo following the trauma. In the contrast-enhanced T1w fat-suppressed magnetic resonance imaging sequence note the contrast enhancement in the periligamentous venous plexus (arrow head). On the left side lower than on the right side. No TS symmetry of the joint space left vs right side (black and white arrow).

injuries including the alar ligament^[13-15]. However, in the case presented here an earlier non contrast-enhanced MRI was performed in a private practice to check exclusively for cervical disc herniation as a potential cause of the unexplained symptoms and a ligament lesion was not suspected at that time. Only after subjective symptoms worsened and were possibly linked to the earlier trauma the potential lesion became evident after simply demonstrating contralateral hypermobility on physical testing.

For an optimal detection of ligamentous lesions, the strength of the MRI has been suggested to be at least 1.5 Tesla, which corresponds to half of the magnetic field strength used in our case for an optimal resolution. A slice thickness of 2 mm is reported to give excellent spatial resolution of the injured alar ligaments^[16]. Since T1-weighted images provide poor contrast resolution and thus less ability to differentiate small variations in signalling we in addition used a Gadolinium contrast enhanced imaging technique. We evaluated the unenhanced and enhanced images in comparison and could better stage the amount of ligamentous injury and an

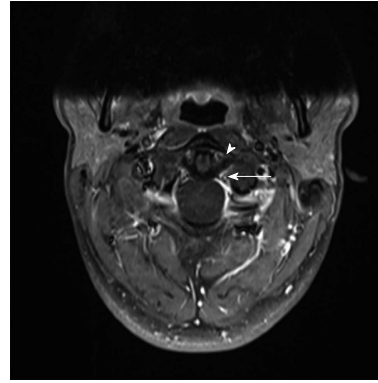


Figure 3 Follow-up magnetic resonance imaging. High resolution contrast-enhanced 3T magnetic resonance imaging, fat-saturated gradient echo sequence. After physical therapy stabilization. Note the clear contrast enhancement in the periligamentous venous plexus (arrow head) and the symmetric space evaluation (white arrow).

oedema of surrounding tissues.

On our final MRI after initiating the 12 mo immobilization therapy no relevant ligament or dens pathology could be documented. However, our patient still complaint of tinnitus and recurrent episodes of neck pressure, headaches and chest pain. Persisting symptoms following cervical injuries are well documented in the literature and seem likely if a healing after two years has not been achieved and more frequent in older individuals^[17,18]. Whether in our patient an instant diagnosis followed by immediate external orthoses or surgical therapy according to recent recommendations would have led to an entire and earlier relief of symptoms remains however hypothetical. In conclusion, we emphasize the value of contrast high resolution 3 Tesla MRI for the detection of ligamentous injuries at the craniovertebral junction^[19].

COMMENTS

Case characteristics

Patient presented with neck stiffness, dysphagia, fasciculation, paraesthesia and neuralgiform pain attacks.

Clinical diagnosis

Findings were suspicious of cervical ligament lesion.

Differential diagnosis

Spectrum of atlantoaxial rotatory dislocation.

Imaging diagnosis

3T Magnetic resonance imaging (MRI), fat-saturated gradient echo sequence (contrast-enhanced T1w fat-suppressed MRI sequence) showed a contrast enhancement in the periligamentous venous plexus with an asymmetry of the joint spaces.

Pathological diagnosis

Alar ligament rupture.

Treatment

Twelve months immobilization therapy.

Related reports

Wong ST, Ernest K, Fan G, Zovickian J, Pang D. Isolated unilateral rupture of the alar ligament. *J Neurosurg Pediatr* 2014; 13: 541-547.

Experiences and lessons

This particular trauma is a challenge and diagnosis might be delayed. We emphasize the value of contrast high resolution 3 Tesla MRI for the detection of ligamentous injuries at the craniocervical junction.

Peer-review

The authors present a case report on a delayed diagnosis of isolated alar ligament rupture and the added value of 3 Tesla MRI for the proper assessment. The paper is well written. Appropriate iconography. The purpose is well defined and it transmits properly the message becoming of potential interest for the readers.

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