



CASE REPORT

Unilateral isolated alar ligament rupture in an adult female patient

Erişkin bir kadın hastada tek taraflı izole alar ligaman rüptürü

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Summary

Only seven cases of isolated unilateral rupture of the alar ligament had been previously reported. The authors report the first adult female case of this rare injury. The patient in their case, a 36-year-old female presented after a trauma due to falling, and at that moment, she had fainted due to a sudden pain between the neck and head. The radiological examinations [magnetic resonance imaging (MRI) and X-rays] had been interpreted as normal. She had a positive Alar ligament test at the right side, and a thin section craniocervical junction computed tomography was obtained which revealed an asymmetrically left-sided odontoid process and a new MRI revealed a right-sided alar ligament rupture. Thus she underwent a bilateral greater occipital nerve block together with pulse radiofrequency and trigger point injection at splenius capitis, levator scapula, and trapezius followed by the application of a halo orthosis to be worn for 3 months. The patient was found to be pain-free in the follow-up examinations. With pure unilateral alar ligament rupture, the atlantooccipital joint is not disrupted and the craniocervical junction is not destabilized. To date, only eight cases of isolated unilateral alar ligament rupture have been reported one of which was a 25 years old male; all of whom presented with marked neck pain and treated by external immobilization for 4 weeks to 4 months and our case is the first adult female patient.

Keywords: Adult; alar ligament; rupture; unilateral.

Özet

Daha önce tek taraflı izole alar ligament rüptürü yedi vaka bildirilmiştir. Yazarlar, bu nadir yaralanmanın ilk yetişkin kadın olgusunu bildirmektedir. 36 yaşında kadın hasta düşme sonrası merkezimize başvurdu. Düşme esnasında aldığı travma sonucu başlayan boyun ve baş arasında yoğun ağrı tarifledi. Radyolojik incelemeler (manyetik rezonans görüntüleme ve grafiler) normal olarak yorumlandı. Muayenesinde sağ tarafta alar ligaman testi pozitif bulundu. Çekilen ince kesitli baş boyun bileşke tomografisinde odontoid sürecin sol tarafta asimmetrik olduğu farkedildi. Sonrasında çekilen menyetik rezonans görüntülemesinde izole sağ alar ligaman rüptürü saptandı. Bu nedenle hastaya iki taraflı oksipital sinir blokajı, pulsed radyofrekans ve splenius capitis, levator scapula, trapezius kaslarına tetik nokta enjeksiyonu uygulanarak hasta üç ay boyunca halo ortez ile takip edildi. İlerleyen takiplerinde hastanın ağrısız olduğu gözlemlendi. Tek taraflı izole alar ligaman rüptürü ile atlantooksipital eklemde ayrışma gözlenmezken, kraniovertebral bileşkede instabilite gelişmediği gözlemlendi. Günümüze kadar izole alar ligaman rüptürü 8 kez rapor edilmiş olup bunlardan sadece biri 25 yaşında erkekti. Diğer hastalar çocukluk çağındaydı. Tüm olguların 4 hafta ile 4 aylık eksternal fiksatorlerle tedavi edildiği gözlemlendi. Bizim olgumuz ise tek taraflı izole alar ligaman rüptürü gelişen ilk erişkin kadın hastadır.

Anahtar sözcükler: Erişkin; alar ligaman; rüptür; tek taraflı.

Introduction

The alar ligaments are important stabilizers at the craniocervical junction, originating bilaterally from lateral or posterolateral aspects of the upper one-third odontoid tip and inserting to both medial tubercles of the occipital condyles and (in two-thirds of the cadavers) C1 lateral masses in close proximity to the occiput-C1 joint; preventing excessive rotation and lateral flexion.^[1–3]

Alar ligament rupture is seen in association with atlanto-occipital dislocation, condylar fracture, and atlantoaxial rotatory dislocation.^[4–7] However, isolated unilateral alar ligament rupture is rare; only eight cases have been reported. These ruptured ligaments can show high signal intensity on proton attenuation-weighted high-resolution magnetic resonance imaging (MRI), This high signal intensity has an unknown etiology; a controversial relation with trauma, and uncertain clinical relevance.^[8]

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Figure 1. Open mouth odontoid process x ray and thin section craniovertebral junction computed tomography revealing asymmetrically left sided odontoid process (arrow).

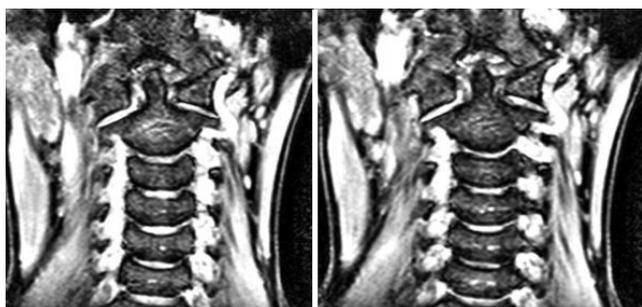


Figure 2. Magnetic resonance imaging showing the right sided alar ligament rupture.

Case Report

A 36-year-old female was admitted due to neck pain (Visual Analog Scale: 10) aggravated by head movements, together with paresthesia in her arms and neck although she had no motor or reflex loss. It was learned that months ago she had a trauma due to falling and at that moment, she had fainted due to a sudden pain between the neck and head. She could not get any relief from physical therapy or pain killers (even Gabapentin). All her radiological examinations (MRI and X-rays) had been interpreted as normal.

Since on our examination she had a positive Alar ligament test on the right side, an open mouth odontoid process X-ray and a thin section craniovertebral junction computed tomography (CT) were obtained which revealed asymmetrically left-sided odontoid process (Fig. 1) and an MRI revealed a right-sided alar ligament rupture (Fig. 2). Thus she underwent a bilateral greater occipital nerve block together with pulse radiofrequency and trigger point injection at splenius capitis, levator scapula, and trapezius followed by the application of a halo orthosis (KB 2001 by DBA, Turkey) to be worn for 3 months since it can provide more rigid immobilization than a cervical collar. The patient was found to be pain-free in the follow-up examinations and the MRI was normal (Fig. 3). The patient was informed that data concerning the case would be submitted for publication and agreed to this.

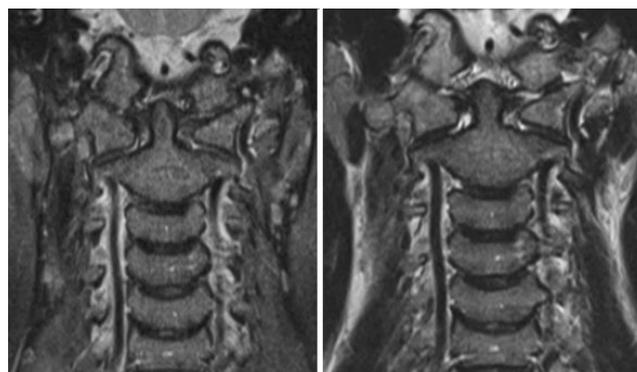


Figure 3. Follow-up magnetic resonance imaging showing the intact alar ligament.

Discussion

The main part of the alar ligament measures 11–15 mm in length, 3–8 mm in height, and 2–4 mm in thickness.^[2,9] The definitive evidence of alar ligament injury is provided by MRI. The most reliable, though indirect, sign of rupture is signal hyperintensity on axial T2-weighted images within the lateral dens-atlas space, now widened due to the ruptured ligament.^[10,11] In an MRI study of healthy individuals, coronal images showed the alar ligament running caudocranially from the dens to the occipital condyle in 67% of those studied, and horizontally in the rest. In the axial plane, the two alar ligaments formed an inverted V in 54%, a straight transverse line in 40%, and only rarely a true V.^[12] An association between the exposure to whiplash injury and high-signal type alterations of the alar ligaments has been observed.^[13,14] Furthermore, conditions that enhance ligamentous laxities such as Down syndrome, Morquio syndrome, and Marfan syndrome correlate with a higher incidence of instability.^[15]

Due to its anatomical and biomechanical characteristics, unilateral rupture of alar ligament most probably occurs when the head is subjected to sudden contralateral rotation and hyperflexion when the forces involved are violent and abrupt, yet not enough to disrupt the sturdier tectorial membrane and transverse atlantal ligament. With pure unilateral alar ligament rupture, the atlantooccipital joint is not disrupted and the craniovertebral junction is not destabilized.^[10] To date, only eight cases of isolated unilateral alar ligament rupture have been reported one of which was a 25 years old male; all of whom presented with marked neck pain and treated by external immobilization for 4 weeks to 4 months and our case is the first adult female patient.^[11]

The decision to take a surgical approach is based on the stability of the joint, its re-dislocation, and on the compromise of the transverse alar ligaments. Compared to conservative management, the arthrodesis of the atlantoaxial joint results in a loss of rotation to each side and therefore it is not recommended as the initial treatment.^[16] Up to this time, no isolated alar ligament rupture-related operation was found in the literature.^[17]

Conclusion

It should always be kept in mind that even if the vertebrae and spinal cord are intact in painful patients with cervical trauma, the craniovertebral junction ligaments should be controlled since their injuries may lead to symptoms related to instability if not treated.

Informed Consent: *Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.*

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References

1. Wong ST, Ernest K, Fan G, Zovickian J, Pang D. Isolated unilateral rupture of the alar ligament. *J Neurosurg Pediatr* 2014;13(5):541–7. [\[CrossRef\]](#)
2. Dvorak J, Panjabi MM. Functional anatomy of the alar ligaments. *Spine (Phila Pa 1976)* 1987;12(2):183–9. [\[CrossRef\]](#)
3. Heller JG, Amrani J, Hutton WC. Transverse ligament failure: A biomechanical study. *J Spinal Disord* 1993;6(2):162–5.
4. Karray M, M'nif N, Mestiri M, Kooli M, Ezzaouia K, Zlitni M. Concomitant alar and apical ligament avulsion in atlantoaxial rotatory fixation. Case report and review of the literature. *Acta Orthop Belg* 2004;70(2):189–92.
5. Pang D, Nemzek WR, Zovickian J. Atlanto-occipital dislocation part 2: The clinical use of (occipital) condyle-C1 interval, comparison with other diagnostic methods, and the manifestation, management, and outcome of atlanto-occipital dislocation in children. *Neurosurgery* 2007;61(5):995–1015; discussion 1015. [\[CrossRef\]](#)
6. Saternus KS, Thrun C. Traumatology of the alar ligaments. *Aktuelle Traumatol* 1987;17(5):214–8.
7. Bloom AI, Neeman Z, Floman Y, Gomori J, Bar-Ziv J. Occipital condyle fracture and ligament injury: imaging by CT. *Pediatr Radiol* 1996;26(11):786–90. [\[CrossRef\]](#)
8. Lummel N, Zeif C, Kloetzer A, Linn J, Brückmann H, Bitterling H. Variability of morphology and signal intensity of alar ligaments in healthy volunteers using MR imaging. *AJNR Am J Neuroradiol* 2011;32(1):125–30. [\[CrossRef\]](#)
9. Osmotherly PG, Rivett DA, Mercer SR. Revisiting the clinical anatomy of the alar ligaments. *Eur Spine J* 2013;22(1):60–4.
10. Briem D, Linhart W, Dickmann C, Rueger JM. Injuries of the alar ligaments in children and adolescents. *Unfallchirurg (Ger)* 2002;105(6):555–9. [\[CrossRef\]](#)
11. Caird MS, Hensinger RN, Vander Have KL, Gelbke MK, Farley FA. Isolated alar ligament disruption in children and adolescents as a cause of persistent torticollis and neck pain after injury. A report of three cases. *J Bone Joint Surg Am* 2009;91(11):2713–8. [\[CrossRef\]](#)
12. Baumert B, Wörtler K, Steffinger D, Schmidt GP, Reiser MF, Baur-Melnyk A. Assessment of the internal craniocervical ligaments with a new magnetic resonance imaging sequence: Three-dimensional turbo spin echo with variable flip-angle distribution (SPACE). *Magn Reson Imaging* 2009;27(7):954–60. [\[CrossRef\]](#)
13. Krakenes J, Kaale BR, Moen G, Nordli H, Gilhus NE, Rorvik J. MRI assessment of the alar ligaments in the late stage of whiplash injury: A study of structural abnormalities and observer agreement. *Neuroradiology* 2002;44(7):617–24.
14. Myran R, Kvistad KA, Nygaard OP, Andresen H, Folvik M, Zwart JA. Magnetic resonance imaging assessment of the alar ligaments in whiplash injuries: A case-control study. *Spine* 2008;33(18):2012–6. [\[CrossRef\]](#)
15. Mathern GW, Batzdorf U. Grisel's syndrome. Cervical spine clinical, pathologic, and neurologic manifestations. *Clin Orthop* 1989;244:131–46. [\[CrossRef\]](#)
16. Dvorak J, Penning L, Hayek J, Panjabi MM, Grob D, Zehnder R. Functional diagnostics of the cervical spine using computer tomography. *Neuroradiology* 1988;30(2):132–7. [\[CrossRef\]](#)
17. Radcliff KE, Hussain MM, Moldavsky M, Klocke NF, Vaccaro A, Albert TJ, et al. Stabilization of the craniocervical junction after an internal dislocation injury: An in vitro study. *Spine J* 2015;15(5):1070–6. [\[CrossRef\]](#)