

Case Report
Trauma

Trauma induced eagle syndrome

A. Koivumäki, M. Marinescu-Gava, J. Järnstedt, G.K. Sándor, J. Wolff: Trauma induced eagle syndrome. Int. J. Oral Maxillofac. Surg. 2012; 41: 350–353. © 2011 International Association of Oral and Maxillofacial Surgeons. Published by Elsevier Ltd. All rights reserved.

Abstract. Eagle syndrome is characterized by secondary calcification and elongation of the styloid process. Eagle syndrome is often associated with sharp, intermittent pain along the path of the glossopharyngeal nerve located in the hypopharynx and at the base of the tongue. In some cases, the stylohyoid apparatus can compress the internal and/or the external carotid arteries and their perivascular sympathetic fibres, resulting in a persistent pain radiating throughout the carotid territory. The pathogenesis of the syndrome is not understood. The authors report the case of a 52-year-old woman with post traumatic Eagle syndrome-like pain and pseudoarthrosis of the stylohyoid ligament.

**A. Koivumäki¹,
M. Marinescu-Gava², J. Järnstedt²,
G. K. Sándor³, J. Wolff¹**

¹Oral and Maxillofacial Unit, University Hospital of Tampere, Finland; ²Department of Radiology, Pirkanmaa Hospital District, Medical Imaging Centre University of Tampere, Finland; ³Department of Oral and Maxillofacial Surgery, University of Oulu, Finland

Accepted for publication 19 December 2011
Available online 12 January 2012

The styloid process normally measures 2.5–3 cm in length and is situated at the base of the temporal bone, immediately posterior to the mastoid apex^{1,10}. Developing from the Reichert cartilage of the second branchial arch, the styloid process, the lesser horn of the hyoid bone and the ligament between them form the stylohyoid apparatus. When the length of the styloid process exceeds 3 cm, it is considered to be elongated and can be associated with a wide variety of symptoms that were first described by Eagle in 1937¹⁰.

Eagle originally described two morbidity forms^{10,3}. The first classic styloid process syndrome form is caused by the calcification of the stylohyoid complex resulting in the scar tissue impinging on cranial nerves V, VII, IX, X or XII, all of which are situated near the styloid process.

The second carotid artery Eagle syndrome form is caused by compression of the sympathetic chain in the carotid sheath.

It often occurs in the absence of ossification of the stylohyoid complex. Only a slight deviation of the styloid process medially is required for the tip of the process to start impinging on an artery and cause a wide variety of symptoms¹⁰.

Patients often experience a wide variety of symptoms, including pain in the throat and ear, vertigo, voice alteration, cough, dizziness, sinusitis, conjunctival injection, headaches, swallowing problems, and pain when turning the head^{7,11}. The symptoms often cause continuous discomfort over long periods of time and are often difficult to diagnose.

The differential diagnosis of Eagle's syndrome may include any condition that can result in cervicofacial pain⁴. These include temporomandibular joint (TMJ) diseases; trigeminal, sphenopalatine, and glossopharyngeal neuralgias; myofascial pain; mastoiditis; otitis; temporal arteritis; dental pain; chronic

tonsillitis or pharyngitis; submandibular sialadenitis or sialolithiasis; esophageal diverticulosis; benign or malignant neoplastic disease; and pharyngeal foreign bodies^{5,6}. Problems that arise in the diagnosis of Eagle's syndrome can result in various unnecessary treatments^{10,5}.

The calcification of the stylohyoid is often detected as an incidental feature on panoramic radiographs when consulting a dentist or an oral and maxillofacial surgeon. In most cases, the patients are over 40 years of age. Eagle syndrome occurs three times more frequently in women than in men² and usually with bilateral calcification⁸. Bilateral involvement does not always involve bilateral symptoms.

The cause of stylohyoid calcification is unknown, but it has been suggested that it may be associated with a genetic polymorphism, trauma, or early onset menopause⁴. When the terms 'trauma' and 'Eagle syndrome' are used as search

headings on PubMed, only three references emerge^{9,12}. On closer examination, these three papers have little to do with trauma.

Surgical and nonsurgical treatment regimens have been described. Nonsurgical treatment involves patient reassurance, the use of analgesics, or the local injection of steroid. The surgical treatment of the calcified stylohyoid is performed using two different anatomical approaches.

The transpharyngeal approach, first described by W. Eagle, is performed through a small incision in the tonsillar fossa by excising the calcified stylohyoid ligament and leaving no visible external cutaneous scar. This operative technique is associated with an increased risk of nerve and vessel injury, deep space infections, and significant postoperative oedema⁴. Talking and swallowing are often difficult for a few days following the procedure. This approach can be difficult to perform on patients with limited mouth opening.

Some authors think that the extraoral approach is superior to the transpharyngeal approach as it provides a better view of the operative field and better wound

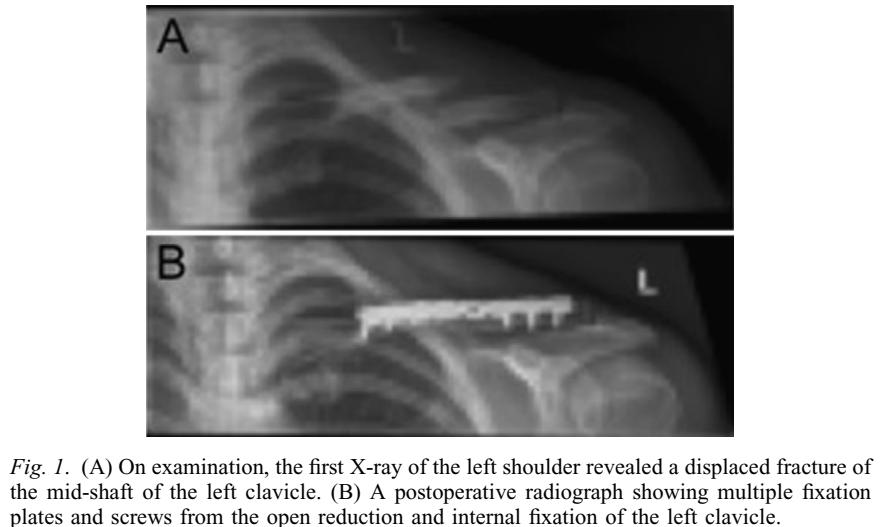


Fig. 1. (A) On examination, the first X-ray of the left shoulder revealed a displaced fracture of the mid-shaft of the left clavicle. (B) A postoperative radiograph showing multiple fixation plates and screws from the open reduction and internal fixation of the left clavicle.

hygiene^{11,4,5}. The extraoral approach takes longer, carries the risk of facial nerve injury, and leaves an external scar.

Case report

At the beginning of 2009, a 52-year-old woman with suspected temporomandibular dysfunction was referred by her dentist.

The patient had been physically assaulted by her husband in 2006. She sustained a blow to her left shoulder resulting in a left clavicle fracture. The initial treatment comprised a clavicle brace and restricted activities for 3 weeks.

After 7 months (2007), the clavicle fracture showed no signs of healing and the patient complained of ongoing pain



Fig. 2. (A) Orthopantomogram showing calcified stylohyoid apparatus. Coronal (B) and sagittal (C) CT scans showing calcified stylohyoid with two pseudoarthrosis; central segment is marked with an asterisk.

and difficulty in lifting her shoulder. Physical examination revealed crepitus and abnormal motion in the middle third of the left clavicle. Radiographs (Fig. 1A) revealed a displaced clavicle fracture. Attempts to achieve apposition of the bone ends had been unsuccessful. In 2007, an open reduction was performed with internal fixation and cancellous bone grafting from the anterior iliac crest (Fig. 1B). The patient was immobilized postoperatively with a sling for 4 weeks. Follow-up examination revealed the fracture to be radiographically united.

Since her assault, the patient had been experiencing continued discomfort in her left jaw and throat when swallowing. The pain was assumed to be due to TMJ dysfunction with progressive worsening. Oral examination revealed no limitation of mouth opening but a slight tenderness and discomfort was observed when palpating the auricular, facial and infraorbital area. Intraoral palpation of the styloid process through the tonsillar fossa aggravated the discomfort and pain.

An orthopantomogram revealed dense calcification of the left stylohyoid ligament (Fig. 2A). Computed tomography (CT) scans revealed dense calcification of the left stylohyoid ligament from the base of the skull to the lesser horn of the

hyoid. Two pseudoarthrosis were present (Fig. 2B and C).

The central part of the calcified left stylohyoid ligament was excised under general anaesthesia using the transpharyngeal approach through a 2–3 cm incision in the palatoglossal arch. The palatoglossus muscle and superior pharyngeal muscle were then gently dissected exposing the calcified stylohyoid. The segment located between the two pseudoarthrosis was separated and removed (Figs 3 and 4).

The wound was sutured in two layers. There were no intra or postoperative complications. The patient recovered uneventfully and was reported to be symptom free at the 2-, 6-, and 12-month follow-up visits.

Discussion

Symptoms induced by the secondary calcification of the stylohyoid ligament are often mistaken for a variety of facial neuralgias, migraine, as well as oral, dental, and TMJ diseases⁸. Calcification of the stylohyoid ligament is often detected as an incidental finding on panoramic radiographs. The elongated and calcified styloid process can be easily palpated on the surface of the tonsil as a hard and pointed structure.

The actual cause of the elongation is poorly understood. Several theories have been proposed: congenital elongation of



Fig. 3. Excised central segment of the left calcified stylohyoid ligament marked with an asterisk.

the styloid process due to the persistence of a cartilaginous anlage an embryologic precursor of the styloid process; calcification of the stylohyoid ligament precipitated by trauma; and growth of osseous tissue at the insertion of the stylohyoid ligament⁶.

CAMADRA et al. reported that when symptoms exist there is little correlation between the extent of the ossification and

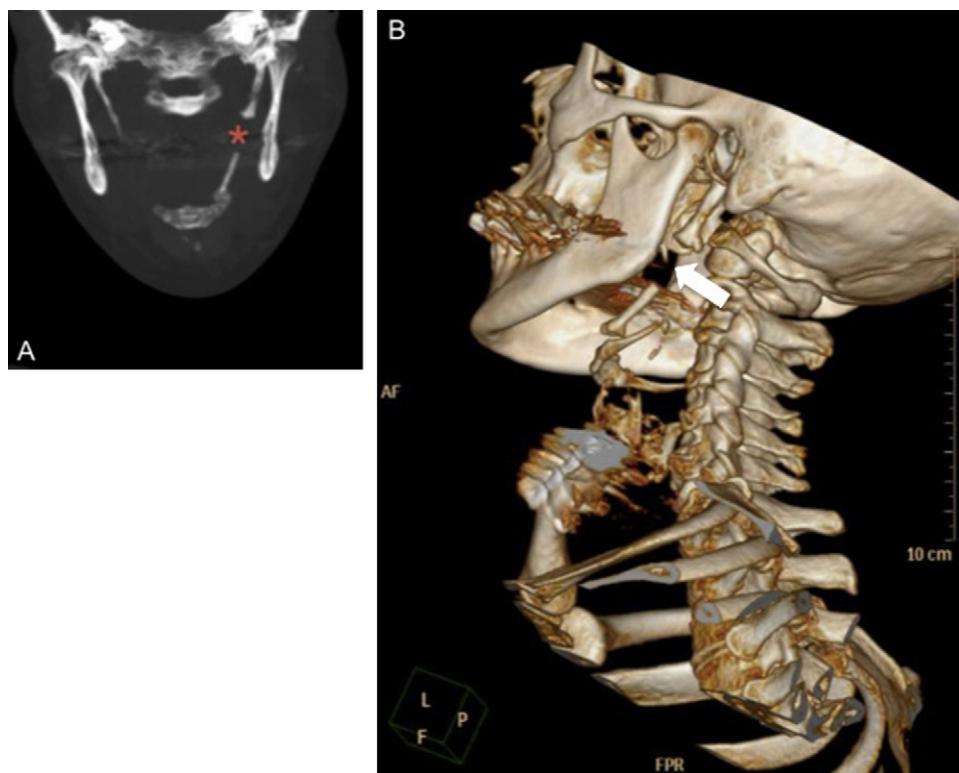


Fig. 4. (A) Sagittal and coronal view showing proximal and distal fragments of calcified stylohyoid ligament and excised central segment marked with an asterisk. (B) Postoperative 3D reconstruction showing absent central segment of the left stylohyoid ligament.

the severity of the symptoms². In this case the central part of the calcified left stylohyoid ligament was excised using the transpharyngeal approach and followed by relief of the patient's symptoms. Other authors have also illustrated subtotal removal of the stylohyoid ligament⁴.

The patient's radiographic and clinical findings revealed a particularly unusual pseudo-articulated and calcified stylohyoid ligament. One explanation for the unusual calcification could have been a consequence of her previous physical assault that resulted in the fracture of her left clavicle and possibly the fractures of the calcified stylohyoid ligament. The patient developed pain immediately after the assault both in the left shoulder and in her left throat and jaw. Her jaw and throat pain worsened over time and the radiographs demonstrated two pseudoarthrosis of the calcified left stylohyoid ligament. Her pain resolved after the removal of the central segment of the calcified ligament and the pain has not recurred.

Funding

None.

Competing interests

No competing interests.

Ethical approval

Ethical approval given by Tampere Yliopiston Sairalla Ethics Committee May, 2011.

References

- Ahmat S, Lokman U, Godden DR, Adam S. Three dimensional CT of Eagle's syndrome. *Diag Interv Radiol* 2005;11:206–9.
- Camadra AJ, Deschamps C, Forest I. Stylohyoid chain ossification: a discussion of etiology. *Oral Surg Oral Med Oral Path Oral Radiol Endod* 1989;67:508–10.
- Carter L. Soft tissue calcification and ossification. In: White SC, Pharoah MJ, editors. *Oral Radiology, Principles and Interpretation*. Missouri: Mosby; 2009. p. 526–40.
- Ceylan A, Köybaşıoglu A, Celenk F, Yilmaz O, Uslu S. Surgical treatment of elongated styloid process: experience of 61 cases. *Skull Base* 2008;18:289–95.
- Diamond LH, Cottrell DA, Hunter MJ, Papageorge M. Eagle's syndrome: a report of 4 patients treated with a modified extraoral approach. *J Oral Maxillofac Surg* 2001;59: 1420–6.
- Godden DRP, Adam S, Woodwards RTM. Case report: Eagle's syndrome: an unusual cause of a clicking jaw. *Br Dent J* 1999; 186:489–90.
- Gokce C, Sisman Y, Sipahioglu M. Styloid process elongation or Eagle's syndrome: is there any role for ectopic calcification? *Eur J Dent* 2008;2:224–8.
- Gokce C, Sisman Y, Tarim Ertas E, Akgunlu F, Ozturk A. Prevalence of styloid process elongation on panoramic radiography in the Turkey population from Cappadocia region. *Eur J Dent* 2008;2:18–22.
- Martin TJ, Friedland DR, Merati AL. Transcervical resection of the styloid process in Eagle syndrome. *Ear Nose Throat J* 2008;87: 399–401.
- Neville BW, Damm DD, Allen CM, Bouquot JE. Developmental defects of the oral and maxillofacial region. In: Neville BW, Damm DD, Allen CM, Bouquot JE, editors. *Oral and Maxillofacial Pathology*. Philadelphia: W.B. Saunders Company; 2009 . p. 1–53.
- Raina D, Gothi R, Rajan S. Eagle syndrome. *Indian J Radiol Imaging* 2009;19:107–8.
- Renzi G, Mastellone P, Leonardi A, Becelli R, Bonamini M, Fini G. Basicranium malformation with anterior dislocation of right styloid process causing stylalgia. *J Craniofac Surg* 2005;16:418–20.

Address:

*Jan Wolff
Oral and Maxillofacial Unit
University Hospital of Tampere
P.O. Box 2000
FI-33521 Tampere
Finland
Tel: +358 40 557 11 99
E-mail: Jan.wolff@uta.fi*