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Case report

Stylohyoid syndrome – Radiological progression and bilateral hypoglossal palsy complication

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ABSTRACT

Objective: Stylohyoid syndrome diagnosis is still challenging nowadays and its symptoms can be easily confused with other orofacial pathologies. The aim of this study is to report a case with stylohyoid syndrome, highlighting the symptoms, diagnosis, and surgical treatment, and describing an unusual major complication. Methods: A 48-year-old woman presented to Portuguese orofacial pain department with a history of progressive and unspecified neck pain lasting for 6 years. After clinical investigation, a stylohyoid syndrome was diagnosed based on previous computed tomography (CT) imaging demonstrating progressive calcification of the stylohyoid complex. Results: Surgery by external approach solved the patient's symptoms, but a transient bilateral hypoglossal palsy was found. It was managed conservatively and recovered within 6 weeks. Conclusion: From our knowledge, this is the first study documenting a stylohyoid complex calcification, improving our knowledge about the calcification progression. Depending on surgical and anatomical conditions, the surgeon should consider approaching the contralateral side in a second surgery to avoid major bilateral complications of the parapharyngeal space, like bilateral hypoglossal palsy.

1. Introduction

In 1948, Dr Watt Weems Eagle described two new categories of orofacial pains, named stylohyoid syndrome, caused by the calcification of the stylohyoid ligament - elongated styloid process (SP) [1]. The first typical stylohyoid syndrome, occurred after tonsillectomies where the local trauma apparently stimulates the SP growth, compressing the pharynx wall at the tonsillar fossa. This leads to a predominant odynophagia and referred ear pain triggered by tongue or pharynx movements, sialorrhea, and a local foreign body sensation. The second, atypical syndrome, the SP compresses the carotid artery walls conducting to sympathetic nerves pressure and restriction of its flow and, consequently neck pain, headache in the external carotid territory, and vascular tinnitus. The latter was named carotid artery syndrome. An important aspect already recognized by Eagle was that not every SP elongation was symptomatic. Later, Camarda, et al. [2], observed that stylohyoid syndrome might not be exclusively related to SP elongation, but it could be associated with other calcifications in any part of the stylohyoid complex defined as the SP, the stylohyoid ligament and the lesser hyoid horn. Even no calcification at all could cause the syndrome, assumed to be stylohyoid ligament tendinitis, also called pseudostyloid syndrome [2,3]. Since Eagle's landmark article, it was clear that symptoms were easily confused with a wide range of orofacial, cervicofacial, and dental chronic pains including temporomandibular joint (TMJ) disorders, headaches like temporal arteritis or migraine, cranial nerves neuralgias, hyoid bone syndrome and head and neck neoplasms leading to frequent misdiagnosis in clinical practice [4–6]. Even when the pain origin is correctly identified, clinicians usually name it Eagle's syndrome [4,7], despite many works distinguishing the different types of stylohyoid syndrome [2,3,6]. Regardless of the radiologic finding, pathophysiology seems related to increased rigidity of the stylohyoid complex. This would cause restrictions of the normal hyoid elevation and depression during local movements (swallowing, head rotation, chewing) and irritation of surrounding neurovascular structures, like the carotid artery and the glossopharyngeal, trigeminal, vagus, and sympathetic chain nerves, leading to local and referred pain (secondary neuralgia) or neurologic vascular symptoms respectively [2,3,6–8].

Once the diagnosis is determined, surgical treatment is accepted as

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the gold standard for stylohyoid syndrome. Some authors prefer intraoral and others external approaches, both with low morbidity [1–3,6,9]. Conservative management can be considered, particularly for those who decline surgery or are in pseudostyloid syndrome [2,7].

This study aims to report a progressive calcification of the stylohyoid complex, the surgical treatment and an uncommon major complication in a case of bilateral stylohyoid syndrome.

2. Case report

A 48-year-old woman was attended with a 6-year complaints of an intense odynophagia irradiated to the submandibular region triggered by opening the mouth or talking. The same pain also limited head rotation and upper limbs raising (Fig. 1a-c). No previous trauma or infection history was reported. Analgesics, non-steroidal, anti-inflammatory agents, and muscle relaxants were prescribed in previous medical appointments in multiple specialties. Initially, the prescribed drugs restricted the pain, but over the last 6 years the pain progressively worsened, and medication failed to control it.

Bilateral tongue base, anterior pillars, and submandibular regions palpation were normal, but painful, the same pain referred to the patient history. Bilateral painless clicking in TMJ and myofascial pain in the masticatory muscles were also identified, with a first suspicion of bilateral dislocated disc with reduction and mastication muscles myositis. The TMJ magnetic resonance imaging (MRI) presented a slight anterior displacement of the posterior band of the temporomandibular disc. No other findings were reported, and temporomandibular disorder was excluded as a possible cause for the main symptoms.

Previous computed tomography (CT) was retrieved from 2012 and compared with new CT images acquired between 2014 and 2018 (Fig. 2a-d). A progressive bilateral lesser hyoid horns elongation was observed (Fig. 2a-d). A gradual SP elongation was verified on the left side from 32 mm to 41 mm (Fig. 3).

The failed previous clinical treatment and clinical investigation, associated with the radiological evolution presenting a progressive and active calcification of the stylohyoid complex over time led to a stylohyoid syndrome diagnosis, and a surgical procedure was planned.

The proposal was an external bilateral stylohyoid complex excision through a submandibular approach. Considering that the patient reported pain in the submandibular region and the hyoid radiological findings, we planned a surgical procedure that could address both the hyoid bone and the SP in the parapharyngeal space.

Under general anesthesia, a bilateral transverse cervicotomy was made respecting the security distance from the marginal facial branch. Subplatysmal flaps were performed, and the superficial cervical fascia was opened over the hyoid bone and both posterior digastric muscle bellies under the submandibular glands. At one side, the lesser hyoid horn was identified and fractured, then it was traced upwards, resecting the entire stylohyoid ligament into the parapharyngeal space (Fig. 4). When the tip of the SP was reached, it was dissected cranially under its periosteum and fractured the most cranial as possible (Fig. 5). All relevant anatomical structures were identified and preserved including the hypoglossal nerve, facial artery, and the carotid artery. The facial vein was divided, and some degree of neighboring structures had to be deviated to allow safe exposure of the parapharyngeal space as the patient presented a short and fatty neck. The same procedure was repeated on the contralateral side. Postoperatively, systemic corticoid was given for 3 days to avoid tongue/pharynx edema.

After surgery, the patient immediately improved the pain and movement restrictions (Fig. 1d-f), but presented an unexpected bilateral hypoglossal palsy compromising speech and swallowing. We managed this major complication with a nasogastric feeding tube for 3 weeks and with a protocol of speech therapy (3 times in the first two weeks, then once a week until full recovery). As we preserved both nerves anatomically, we expected complete spontaneous recovery which occurred progressively between 4 and 6 weeks. A postoperative CT scan confirmed the removal of both SP (Fig. 6).

3. Discussion

Stylohyoid syndrome diagnosis can be very challenging due to unspecific symptoms and diverse differential diagnoses like TMJ disorders, glossopharyngeal neuralgia, trigeminal neuralgia, and myofascial pain dysfunction. The multiple diagnostic possibilities and the wide range of health professionals (neurologists, otolaryngologists, neurosurgeons, maxillofacial surgeons, and dentists, among others) working in this area contributed to the delayed stylohyoid syndrome diagnosis in this patient. Stylohyoid syndrome misdiagnosis is common in the literature [1,4,9]. In clinical assessment, a movement that triggers or aggravates somatic (such as lifting the upper limbs, talking, chewing, or rotating the

Fig. 1. Preoperative limited movements (A, B, C) and normal postoperative movements (D, E, F). (A) Limited upper arms abduction; (B) Limited head rotation to the left; (C) Limited head rotation to the right; (D) Normal upper arms abduction; (E) Normal head rotation to the left; (F) Normal head rotation to the right.

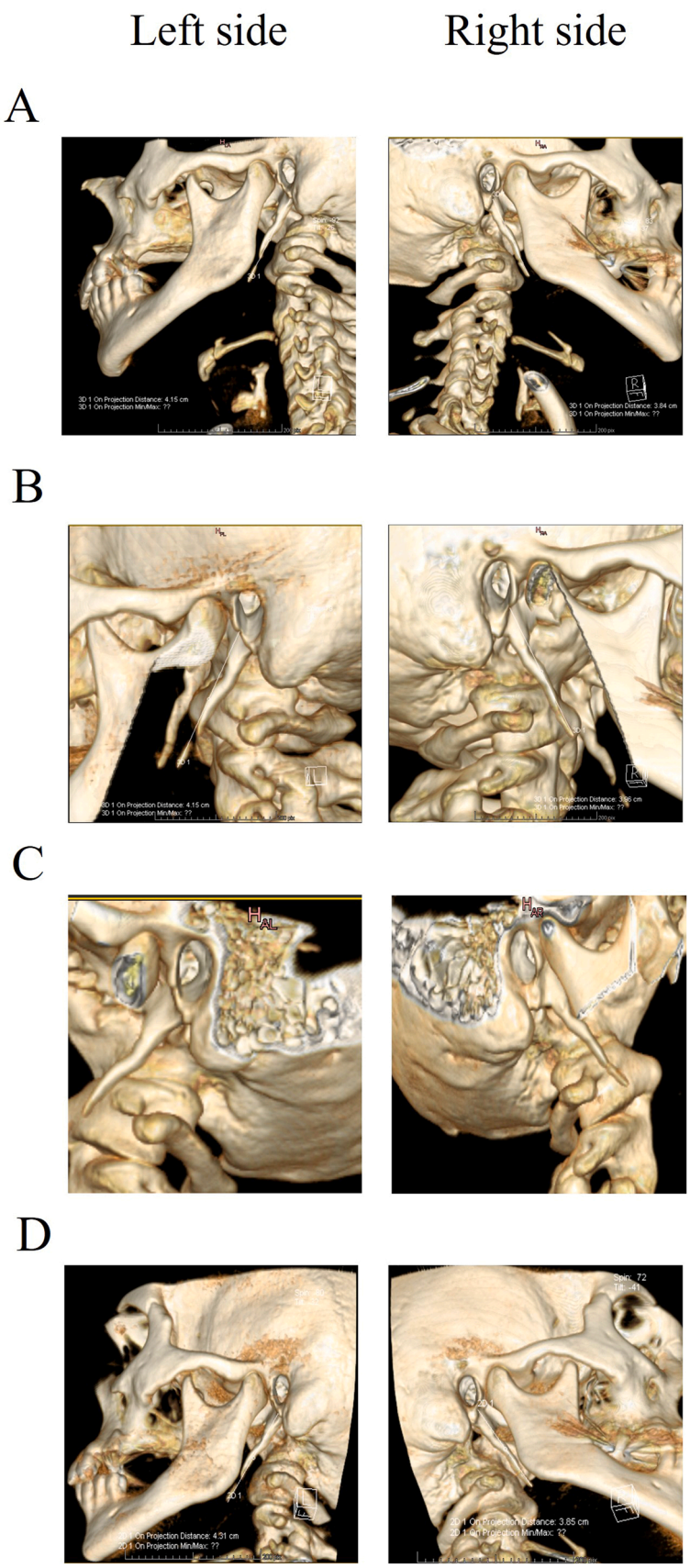

Fig. 2. Computed tomography (CT) images of the styloid process length over the years. (A) 17th April, 2014; (B) 16th June, 2015; (C) 10th, November 2016; (D) 14th March, 2018.

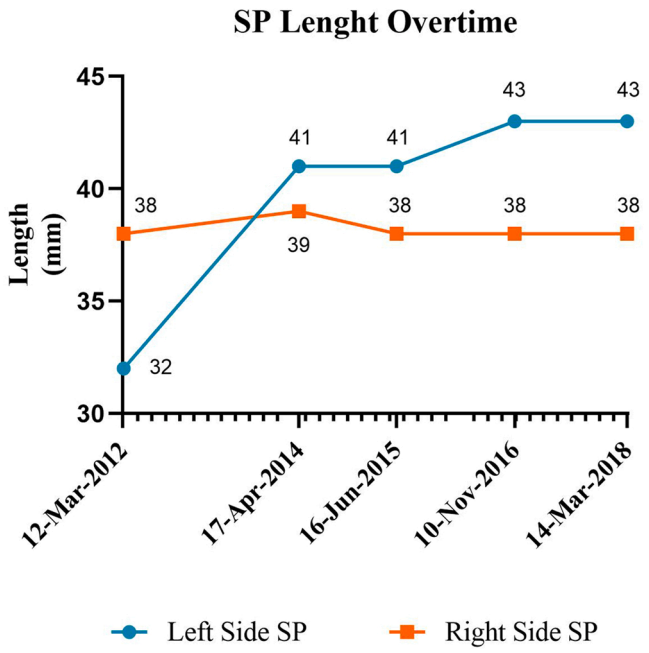

Fig. 3. Graphical representation of styloid process length over time.

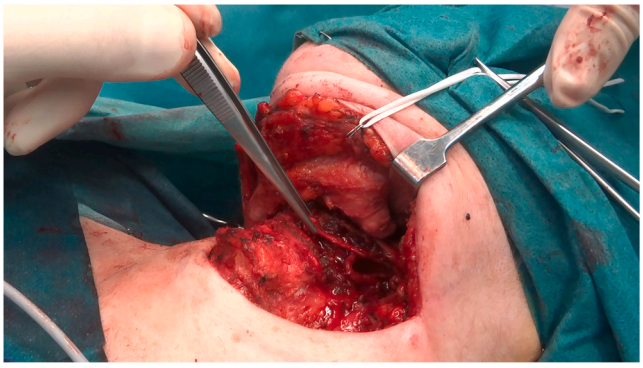

Fig. 4. Bilateral transverse cervicotomy surgical approach. Surgical forceps are holding the stylohyoid ligament to be removed.

head) or/and vegetative (such as swallowing or yawning) pain should raise clinical suspicion of osteomuscular or articulatory origin.

Many clinicians base the stylohyoid syndrome diagnosis on the radiologic finding of elongated SP. But defining an elongated SP is far from consensus in the literature. Many studies are measuring it in cadavers and radiological exams like dental panoramic radiographs and CT scans, but only two calculated the natural variation expected for normal SPs [9,10]. Different methodologies were used, which difficult comparisons between them. Moffat, et al. [9] measured in cadavers from the SP posterior face finding a normal Gaussian distribution between 15.2 mm and 47.7 mm encompassing 95.4% of his sample. Jung, et al. [10] measured in panoramic radiographs from the SP anterior face reporting a right-skewed distribution below 46 mm for 90% of his sample. In our case, the SP was not abnormally long for both reports. But, over time, it was growing, and not only the left SP itself, but also the lesser hyoid horns. There is no previous report of follow-up of this condition. Still, clinically, it is plausible that a progressive stylohyoid complex calcification can be the etiology of an unspecific pain in a symptomatic patient. This finding cannot be extrapolated to the asymptomatic population because there is evidence that SP length increases with aging [10].

Unilateral hypoglossal palsy is reported as a possible complication of parapharyngeal surgery. Stylohyoid hypoglossal symptoms preoperatively are rare, but may occur and may not fully recover after surgical treatment [8]. Asymptomatic stylohyoid ligament complex calcification can also be present with unilateral hypoglossal paralysis after airway management for general anesthesia [11] or even for surgical positioning/maneuvers for non-neck procedures [12].

To the best of our knowledge, only one report presented a bilateral hypoglossal nerve paralysis after external stylohyoid syndrome surgery, in a very similar case to our report [13]. This work does not describe the surgery in detail but the burden was shared between anesthesia and surgical procedures. Other bilateral hypoglossal paralysis was reported in surgical settings and was usually related to orotracheal intubation or the use of laryngeal masks for ventilation [13]. In our patient, we selected a submandibular approach because of the need to address the lesser hyoid horn which would be impossible with a classic more lateral SP-only approach. But due to a short and fatty neck with a narrow and deep parapharyngeal space, adequate surgical exposure of SP was challenging from our submandibular approach. The timespan taken for complete and spontaneous recovery (between 4 and 6 weeks) shows that the predominant hypoglossal injury was neuropraxia, probably by surgical traction of both nerves.

It is not the first time our group performed a submandibular approach for parapharyngeal space (unpublished data), but it was the

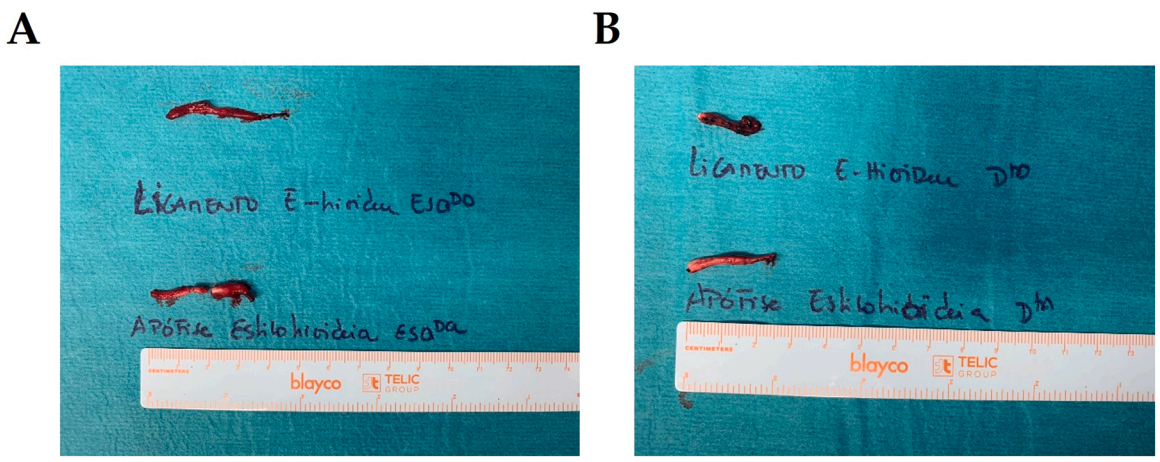

Fig. 5. Surgical resections. (A) In the superior part of the figure we observe the left stylohyoid ligament and in the lower part of the figure we observe the left styloid process; (B) In the superior part of the figure we observe the right stylohyoid ligament and in the lower part of the figure we observe the right styloid process.

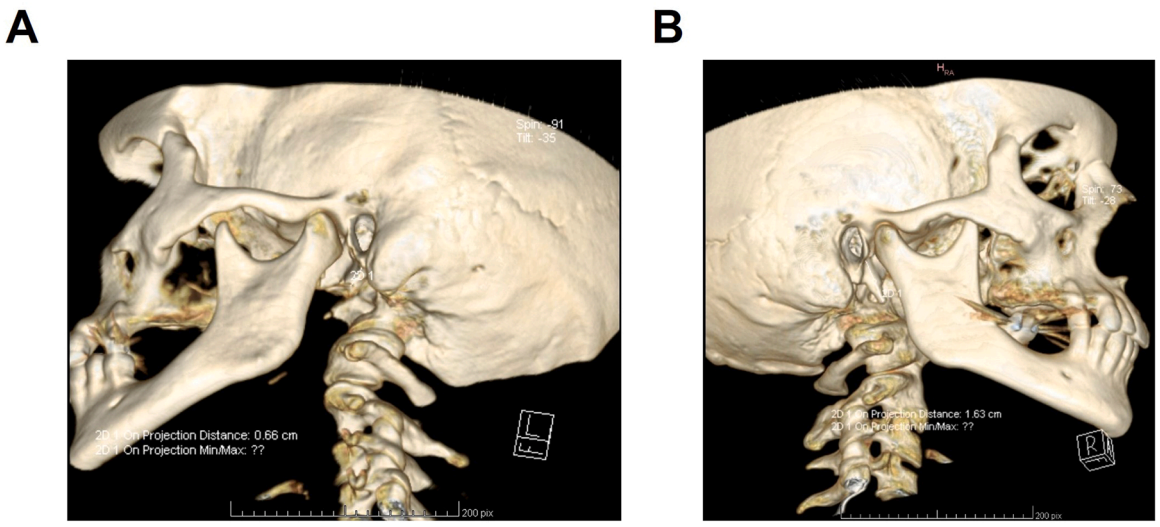

Fig. 6. Postoperative computed tomography (CT) confirms the correct surgical treatment in the left side (A) and right side (B).

first time that hypoglossal palsy occurred. We believe that it is due to the particular patient anatomy already described. Retrospectively reviewed, our other patients had skinnier and longer necks, making the parapharyngeal space exposure much easier to perform with the same access. We learned that if difficulty in exposing the parapharyngeal space exposure is anticipated or found during a submandibular approach, the surgeon should either consider delaying the contralateral side to a second surgery, or either extending laterally the incision to allow a wider exposure of the parapharyngeal space through the anterior upper third of sternocleidomastoid muscle, trying to avoid injuries to the external jugular vein or great auricular nerve.

4. Conclusion

From our knowledge, this is the first study documenting stylohyoid complex calcification over time in a symptomatic patient. It is postulated that this radiological evolution can support stylohyoid syndrome diagnosis in difficult clinical cases. When external bilateral approaches are planned, the surgeon must balance surgical morbidity with adequate and safe exposure of the parapharyngeal space to avoid bilateral concomitant parapharyngeal space complications. In complex cases, the surgeon should consider approaching the contralateral side in a second surgery to avoid major bilateral complications.

Provenance and peer review

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Competing interests

The authors have no conflict of interest to declare.

Patient consent

The participant gave the informed, clarified, and free term of consent in writing and per current legislation.

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