Case Report

An unusual clinical manifestation of ossified styloid complex: Diagnosis and treatment

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Abstract

The stylohyoid complex (SC) connects the temporal bone to the hyoid bone and may suffer anatomical variations that are represented by a length increase of the styloid process or ossification of the stylohyoid ligament. Reported here is a case of a 58-year-old asymptomatic patient who presented an unusual clinical manifestation on the floor of the mouth, which precluded the installation and adaptation of dental prostheses. The first diagnostic hypothesis was sialolithiasis. Panoramic radiography and cone-beam computed tomography revealed a massive ossified SC. Preprosthetic surgery was performed. The present case report highlights that the styloid complex may present unusual anatomical variations which can be clinically and radiographically detectable.

Key words: Cone-beam computed tomography, elongated styloid process, styloidectomy

Introduction

Stylohyoid complex (SC) is a bone ligament structure that connects the temporal bone to the hyoid bone, involving the styloid process (SP), the stylohyoid ligament (SL), and the lesser cornu of the hyoid bone.

The most common anatomical variations of the SC are ossification of the SL and an increase in length of the SP. It is considered elongated when it reaches a length over 30 mm.

The etiology of elongation and ossification of the SC is poorly explained in literature. The pathogenesis seems to be linked to chronic irritations, surgical trauma, growth of osseous tissue, or trauma during the development of SC could result in calcified hyperplasia of the SL.

The prevalence of SP elongation or SL ossification has been reported to be between 19.4% and 52.1% in the general population and up to 76% in patients with temporomandibular disorder.

This anatomic variation usually is an asymptomatic condition. Only in 4% of the cases, symptoms are associated with syndromes, such as Eagle syndrome. This syndrome manifests as cervicofacial, ear, and throat pain. In addition, headaches and dysphagia may be present.

This report presents a rare case of a massive SC associated with an asymptomatic swelling on the floor of the mouth. The patient was submitted to styloidectomy for prosthesis rehabilitation.

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**Case Report**

A 58-year-old edentulous male sought dental treatment at the Stomatology Clinic of the Bauru School of Dentistry, University of São Paulo, Brazil. He was referred by his dentist because of a swelling on the floor of the mouth. The mucosa had normal color at inspection revealing a mobile fusiform-shaped swelling 3 cm in length, hard on palpation, and next to the left alveolar ridge. The swelling had never been noticed by the patient [Figure 1].

The first diagnostic hypothesis was sialolithiasis of the submandibular gland. An occlusal radiograph revealed a thin fusiform radiopaque structure not compatible with sialolith. The patient has never had any symptoms of gland obstruction to strengthen this hypothesis [Figure 2].

Panoramic radiograph presented bilateral ossified SC [Figure 3]. On the right side, the ossification was unclear due to the superposition of the structures. On the left side, there was a thick rounded ossified image reaching the angle of the mandible. The end tip of this image was protruding on the floor of the mouth as a clinically palpable swelling.

The patient reported no history of pharyngeal trauma, otalgia, cervicofacial or throat pain, or any difficulty in swallowing. Due to the proximity of the swelling to the alveolar ridge, it was not possible to install a prosthesis. The treatment established was surgical resection of the ossified SC, only on the left side.

A cone-beam computed tomography (CBCT) examination was performed to evaluate the full extent, length, and angulation of the ossified SC and to assist in the surgical planning. It showed that there was a partial ossification on the right side, 83.40 mm long, segmented into two fragments, and finishing at the lesser cornu of the hyoid bone. On the left side, the ossified SC was a single structure, measuring 72.24 mm [Figure 4], ending in a round tip.

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**Figure 1:** Fusiform swelling on the floor of the mouth next to the alveolar ridge

**Figure 2:** Occlusal distorted radiograph showing a fusiform radiopaque structure on the posterior floor of the mouth

**Figure 3:** Panoramic digital radiograph showing the ossified stylohyoid complex on both sides of the mandible. On the left side, the arrow points to the round-shaped end tip of the structure

**Figure 4:** (a) Sagittal reconstruction of cone-beam computed tomography showing the ossified SC on the right side. The lower part of the image reaches the lesser cornu of the hyoid bone. (b) Three-dimensional image of the cone-beam computed tomography scan showing the ossified stylohyoid complex on both sides. Note that on the right side, the ossified stylohyoid complex is segmented into two parts and on the left side, it is unique. (c) Sagittal reconstruction of cone-beam computed tomography showing the round end tip of the left ossified stylohyoid complex near the mandible angle. The end tip diameter was 4.88 mm
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responsible for the protrusion on the floor of the mouth. The mean diameter was 5.83 mm (range from 3.19 to 9.57 mm) of the entire extension.

Surgical resection using an intraoral approach was performed under local infiltrative anesthesia. The protuberance of the round end tip of SP was identified by digital palpation in order to determine the position of the incision, which was performed using a number 15 surgical scalp knife blade from the lingual surface of the mandible parallel to its ascending branch. The end tip of the SP was exposed and stripped of all attachments. Tissue was detached superiorly as posteriorly as possible. It was possible to apprehend the SP with a Luer rongeur instrument (Quinelato®), promoting its fracture and removal. Silk suture was performed promoting coaptation of the edges of the surgical wounds. Anti-inflammatory drugs were prescribed to the patient [Figure 5]. Figure 6 shows a 7-month follow-up indicating that the patient has recovered well, with no signs of abnormalities. The patient continues to have no symptoms.

DISCUSSION

The prevalence of SP elongation or SL ossification has been reported to be between 19.4% and 52.1% in the general population.[7,8] Its etiology is poorly understood in the literature.[3]

Diagnosis of ossified SC consists of the clinical history of pain, physical examination, and radiographic interpretation. Physical examination consists of palpation of the tonsillar fossa. [11] An atypical case of an ossified SC in a 58-year-old patient with an intraoral swelling on the floor of the mouth is presented. Any mention of this unusual position was not found in the literature. In 2012, Baharudin et al. reported a case of SP elongation with a bony hard mass located over the tonsillar fossa.[12]

Patients are usually asymptomatic: Only 4% of them present symptoms associated with syndromes, such as the Eagle syndrome.[5,10] Compressions on the neural and vascular structures are probably the reason for this. However, the SC may reach a long length without symptoms, as in the present case. Similar cases are described in the literature as incidental radiographic findings.[2,5,13]

Several conventional radiographic methods can be applied to diagnose an ossified SC: Posterior-anterior skull radiograph, lateral oblique radiographs of the mandible, panoramic radiograph, and computed tomography.[3] However, there are some limitations in conventional radiographs when the mandibular bone and the teeth superimpose on the SC, and they reduce the visibility of the SC on the images. In addition, the panoramic radiograph may distort the dimensions of the stylohyoid chain complex.[3,14]

CBCT might be considered as an additional examination alternative to panoramic radiographs due to its effectiveness in evaluating the SP length, angulation, and morphological characteristics, mainly prior to surgery.[3,14]

In the present case, the ossified SC measured 72.24 mm on the left side and 83.34 mm on the right side. According to the literature, the average length of the SP is <30 mm, with normal length ranging from 15.20 to 47.70 mm.[2] In a Brazilian study, Rizzatti-Barbosa et al. concluded that the ossification extension varied from 26.1 to 65 mm.[6] This is not in line with the present report.

In cases where treatment is necessary, conservative drug approaches and surgical techniques are available.[9,12,15] In conservative treatments, anti-inflammatory, analgesic,
The surgical technique involves amputating or removing the elongated SP. It is well accepted in the literature that surgery is the primary treatment for a symptomatic patient, which can be performed using an intraoral/transoral or an external/transcervical approach. To choose between these two options, a surgeon must have an intimate understanding of each procedure and balance their advantages and disadvantages. The intraoral approach is considered technically easier and less time-consuming, creates no external scar, and can be done simultaneously with other surgeries, such as tonsillectomy, under local or general anesthesia. On the other hand, its disadvantages must be taken into account. In the literature, they include cervical infection due to contamination from the pharynx into the neck and restricted field visualization, which can lead to injuries to the external carotid artery and facial nerve.

The external technique provides adequate exposure of the surgical field, in addition to less risk of infection and hemorrhage. On the other hand, it can result in a cutaneous scar, which can be a disadvantage for the patient. Table 1 shows the main aspects of the most recent case reports in the literature comparing gender, age, symptomatology, oral clinical manifestation, image examinations, and the established treatment. It is possible to notice that the treatment is chosen based on individual characteristics.

In the present case, the patient was asymptomatic but requested surgical treatment to be able to wear a prosthesis. Thus, surgical resection was performed only on the left side. On the right side, no clinical manifestation was detected and it was decided to follow up the case.

### Conflicts of interest

There are no conflicts of interest.

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### References


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CBCT: Cone-beam computed tomography; CT: Computed tomography

antidepressant, and anticonvulsant medications are cited in the literature as options. Glogoff et al. stated in 1981 that other methods include steroids or local anesthesia with long-term effects on the tonsil or tender areas. Nonsurgical therapy is considered an adjunct treatment after surgery aimed at reducing reossification of the SL or in cases where surgery is contraindicated. Antidepressant, and anticonvulsant medications are cited in the literature as options. Glogoff et al. stated in 1981 that other methods include steroids or local anesthesia with long-term effects on the tonsil or tender areas. Nonsurgical therapy is considered an adjunct treatment after surgery aimed at reducing reossification of the SL or in cases where surgery is contraindicated. Antidepressant, and anticonvulsant medications are cited in the literature as options. Glogoff et al. stated in 1981 that other methods include steroids or local anesthesia with long-term effects on the tonsil or tender areas. Nonsurgical therapy is considered an adjunct treatment after surgery aimed at reducing reossification of the SL or in cases where surgery is contraindicated. Antidepressant, and anticonvulsant medications are cited in the literature as options. Glogoff et al. stated in 1981 that other methods include steroids or local anesthesia with long-term effects on the tonsil or tender areas. Nonsurgical therapy is considered an adjunct treatment after surgery aimed at reducing reossification of the SL or in cases where surgery is contraindicated. Antidepressant, and anticonvulsant medications are cited in the literature as options. Glogoff et al. stated in 1981 that other methods include steroids or local anesthesia with long-term effects on the tonsil or tender areas. Nonsurgical therapy is considered an adjunct treatment after surgery aimed at reducing reossification of the SL or in cases where surgery is contraindicated. Antidepressant, and anticonvulsant medications are cited in the literature as options. Glogoff et al. stated in 1981 that other methods include steroids or local anesthesia with long-term effects on the tonsil or tender areas. Nonsurgical therapy is considered an adjunct treatment after surgery aimed at reducing reossification of the SL or in cases where surgery is contraindicated.
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