

Styloid-Stylohyoid Syndrome: Literature Review and Case Report

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The first reports on enlargement of the styloid process date from the 17th century, when anatomists explored this region. These postmortem findings were only useful as a mere anatomic curiosity and had no clinical correlation. In 1937, W.W. Eagle reported various cases of a cervicopharyngeal symptomatology and associated them with radiographic findings. It was believed that trauma in the cervicopharyngeal region, especially after tonsillectomy, might stimulate a subsequent growth of the styloid process.¹⁻³

Various names were proposed for the syndrome: styloid process neuralgia,⁴ styloid syndrome,⁵ stylohyoid syndrome,³ elongated styloid process syndrome,⁴ Eagle's syndrome,⁶ and styloalgia.^{7,8} The most appropriate, however, is styloid-stylohyoid syndrome because such anomalies may be of the styloid process, of the stylohyoid ligament, or a combination of both,³

although it has been divided into 4 syndromes in recent literature.¹

Differential diagnoses are innumerable because many of the symptoms detected in the enlargement of the styloid process (orofacial pains and dysfunctions) are also found in patients without presence of the elongated process. Furthermore, its attachments may be susceptible to stretch and whiplash type injuries, in which an acute force exceeds the physiologic limits of the temporal bone attachment.⁹

The objective of this article is to present a comprehensive systematic review of literature and to report a case of stylohyoid ligament ossification with 2 pseudoarticulations.

Literature Review

The styloid process is a slender projection connected to the inferior aspect of the petrous part of the temporal bone just below the tympanic membrane and behind the tympanic plaque which shields its attachments. It lies behind the pharyngeal wall of the palatine fossa, between the internal and external carotid arteries.⁹ Innervation comprises the glossopharyngeal nerve in the posterior lateral wall of the tonsillar fossa (medial to the process), and the facial nerve emerging from the stylomastoid foramen which is slightly posterolateral to the base of the styloid process. The accessory nerve, the hypoglossal nerve, and the vagus nerve are placed medially to the process, together with the internal jugular vein and the internal carotid artery with its sympathetic chain.^{9,10}

The normal size of the styloid process varies significantly in the literature (Table 1).

According to Camarda et al,¹ the ceratohyal element degenerates with time. Nonetheless, its fibrous sheath persists as the stylohyoid ligament, containing a cartilaginous and bone potential because the styloid process normally ossifies 5 to 8 years after birth.

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Table 1. NORMAL LENGTH OF THE STYLOID PROCESS ACCORDING TO THE LITERATURE

	Normal Length				
	Both Sides	Each Side		Gender	
		Left	Right	Male	Female
Frommer ¹⁸	3-5 cm	-	-	3.26 cm	2.97 cm
Eagle ²⁶	<2.5 cm	-	-	-	-
Correl et al ¹⁷	2.5 cm	-	-	-	-
Langlais et al ²¹	2.5 cm	-	-	-	-
Montalbetti ³⁴	2.5 cm	-	-	-	-
Gossman and Tarsitano ³	2.5 cm	-	-	-	-
Stafne and Hollinshead ³⁵	2-3 cm	-	-	-	-
Lindeman ³⁶	2-3 cm	-	-	-	-
Kaufman et al ²⁰	-	2.95 cm	2.99 cm	-	-
Silva et al ²²	-	3.135 cm	3.372 cm	-	-
Moffat et al ⁷	1.52-4.77 cm	-	-	-	-

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Variations in the ossification and fusion of the 4 elements of the second brachial arch (under appropriate stimulation) in youngsters may lead to a marked variation in the radiographic appearance of the whole stylohyoid chain.

Steinmann⁵ proposed 3 theories to explain such ossification. The theory of reactive hyperplasia implies that if the styloid process is adequately stimulated, as in pharyngeal trauma, ossification would take place in the terminal portion of the process at the expense of the stylohyoid ligament. The theory of reactive metaplasia also involves traumatic stimulus, which would induce some sections of the stylohyoid apparatus to undergo metaplastic changes and thereby become intermittently ossified. The third theory, of anatomic variation, involves the stylohyoid ligament and/or the styloid process as ossified structures that develop in the early formative years after birth. The theory may fit in radiographic findings of ossification in children and young adolescents and in the absence of antecedent cervicopharyngeal trauma (as an inductive stimulus).¹ This could not be adjusted to the classical Eagle syndrome because there is no prior trauma.¹¹

Camarda et al¹ added the theory of aging developmental anomaly, in which there is an increased inelasticity of the soft tissue. This may lead to the development of tendinosis in the junction of the stylohyoid ligament with the lesser cornu of the hyoid bone, secondary to the increased ligament resistance to joint movement (between ligament and bone) in some older patients.

As such, there are 4 syndromes: classical Eagle syndrome and carotid artery syndrome, with the reactive hyperplasia and metaplasia theories of ossification which explains the first and maybe the second (once trauma is not necessarily the cause of the carotid artery syndrome); stylohyoid syndrome, which

would be justified by the theory of anatomic variation; and finally pseudostylohyoid syndrome, which is explained by the theory of aging developmental anomaly.¹

The symptoms of classical Eagle syndrome and carotid artery syndrome are well described in literature.^{1,3,5,6,9,10,12-24} The symptoms of the other 2 are the same as classical Eagle syndrome, but without prior trauma history and involving a specific age group, usually above 40 years for pseudostylohyoid syndrome but not necessarily for stylohyoid syndrome. In classical Eagle syndrome, the chief complaint is continuous throat pain during the convalescent period in patients submitted to tonsillectomy and a sensation of foreign body lodged in the throat. Pain related to swallowing and speech is frequently referred to the ear on the side of the elongated styloid process.³ It is assumed that healing tonsillectomy scar tissue tightens the mucosa across the tip of the elongated styloid process and that movements of this mucosa during function across it is thought to cause the symptoms.⁹ Pharyngeal pains are theoretically generated by stretching or fibrous compression of the V, VIII, IX, and X cranial nerve endings in the tonsillar fossa during the healing phase. When the stylohyoid ligament is ossified, contraction of the stylopharyngeal muscle lifts the pharynx upward and laterally and, with the ossified ligament remaining fixed in this maneuver, the glossopharyngeal nerve is pulled across it during the swallowing act and may be stimulated mechanically to produce pain.³ Symptoms of dysphagia, pain referred to the ear, dysphonia, and the sensation of a foreign body in the pharynx during functional movements (eg, eating, yawning, turning the head, and swallowing) are part of classical Eagle syndrome.⁹

An important feature of carotid artery syndrome is that it is not dependent on a tonsillectomy. Because the styloid process lies between the internal and ex-

ternal carotid, any deviation in the process or ossification of the ligament may produce pressure on either of these structures and produce regional carotidynia (pericarotid sympathetic plexus irritation). When pressure is exerted on the external carotid artery, the pain is regionalized to the anatomic structures supplied by this artery. Clinically, the patient may complain of constant pain in the neck, pain on turning the head, regional carotidynia, or tenderness of a cervical lymph node. Pressure on the internal carotid artery may produce symptoms as a result of the unique arterial supply to the cranial cavity and its contents.^{3,25,26} Symptoms of carotid artery syndrome present as chronic neck pain, pain on turning the head, and pain irradiating to the eye.⁹

Normally, panoramic radiography is indicated for best visualizing the styloid process, but other x-ray options (such as anteroposterior and lateral cephalometric radiography) help to visualize the process in 2 planes.^{3,27}

The use of 2-dimensional and currently 3-dimensional (3D) computed tomography (CT) is better for defining length, angulation, and anatomic relationships of the stylohyoid process.^{19,28}

The radiographic images may be divided into 3 types.²¹ Type I (elongated) is characterized by an uninterrupted integrity of the styloid image. Processes up to 25 mm in length are accepted as normal. However, because of the magnification of panoramic projections, those of 28 mm were also considered normal. In type II (pseudo articulated), the styloid process is apparently joined to the mineralized stylohyoid or stylomandibular ligament mineralized by a single pseudoarticulation, usually located superior the level tangential to the inferior border of the mandible, giving an appearance of an articulated elongated styloid process. This type is much less frequent than the first. Type III (segmented) consists of either long or short noncontinuous portions of the styloid process or interrupted segments of the mineralized ligament. In either instance, 2 or more segments are seen with interruptions either above or below the level of the inferior border of the mandible, or both.

Regarding prevalence of gender in styloid-stylohyoid syndrome, the subject is quite controversial. Various authors did not find predominance by gender.^{14,17,27,28} Other authors report a higher frequency in females.^{5,8,13,15,19,29} Diverging from Correl et al,¹⁷ Eagle,² Strauss et al,⁶ and Zohar et al²⁴ reported a higher prevalence in the male gender.

According to Correl et al,¹⁷ the average age was 56 years in patients presenting roentgenographic anomalies, while for Härmä,⁸ Gossman and Tarsitano,³ and Strauss et al,⁶ the syndrome is more frequent in patients over 30 years of age and may be hereditary. More recently, Camarda et al¹ reported that it affects

patients over 40 years of age, while it was those over 50 for Fini et al.¹⁹

There are many differential diagnoses concerning the oral and maxillofacial area and other specialities. They are presented in Table 2.

To palpate the elongated styloid process, the gloved finger is inserted along the occlusal line posteriorly to the region of the tonsillar fossa. If the process is palpable, it will be firm and pointed.^{3,14} Pressure to the region exacerbates symptoms and local sensitivity.^{3,13,14,23,24,30} Care must be taken by the examiner not to manipulate the lateral and dorsal borders of the tongue as this will evoke the gag reflex.³

After a few minutes of infiltration of 1 mL 2% lidocaine clorhydrate at the site at which the styloid process was palpable on the tonsillar fossa, the patient's symptoms and local tenderness subsides temporarily and the result of the test is regarded as positive.^{8,13,14,23}

According to Prasad et al,¹³ the intraoral technique is made with the patient under general anesthesia, the tonsillar bed is palpated and the tip of the styloid process is identified. With Negus curved arterial forceps, the muscles of the tonsillar bed are dissected with a blunt instrument,^{3,13} separated and retracted downward between the 2 arms of the forceps. An incision is made in the periosteum at the tip of the styloid process. Then, the periosteum is stripped from the tip to the base using a freer's mucoperiosteal elevator, while constant reflection of the muscles is made with the forceps. The styloid process is then excised with a bone nibbling rongeur. The tonsillar bed is sutured with catgut and then a nasogastric tube is inserted. Postoperatively, patients continue fasting for 48 hours. Parenteral antibiotics are administered during these 48 hours and then oral diet and oral antibiotics are instituted. Patients are discharged on the fifth postoperative day.

To Strauss et al⁶ and Zohar et al,²⁴ the procedure is conducted under local anesthesia; the muscles beneath the mucosa are separated and dissected with a curved septal elevator. The distal end of the process is grasped with a clamp and then carefully broken and excised using a bone nibbling rongeur. Suture may be performed with polyglycolic acid or catgut in the pharyngeal mucosa. In the presence of palatine tonsils, tonsillectomy should be performed first^{6,19,24} and special care is necessary to avoid injuring the closely associated structures (external and internal carotid arteries and the glossopharyngeal nerve) during detachment.³ In the past, the manual fracture of the elongated styloid process was advocated, but postoperative results were unsatisfactory.^{12,16,31}

The extraoral procedure was first described by Loeser and Cardwell.³² Surgery is conducted under

Table 2. DIFFERENTIAL DIAGNOSES OF STYLOID-STYLOHYOID SYNDROME ACCORDING TO THE LITERATURE

Differential Diagnosis	Amancio et al ³⁷	Aral et al ¹⁵	Balasubramanian ³³	Breault ¹⁶	Camarda et al ¹	Diamond et al ¹²	Fini et al ¹⁹	Gossman and Tarsitano ³	Grossmann and Paiano ³⁰	Nakamaru et al ²⁸	Sivers and Johnson ¹⁴	Strauss et al ⁶	Tiago et al ²³	Zohar et al ²⁴
Degenerative alterations of the cervical column														
Esophageal diverticula														
Dysesthesia of the laryngopharynx														
Temporal arteritis														
Temporal arthritis														
Tumors														
Pain of dental origin														
Migraine headaches														
Chronic pharyngotonsillitis														
Atypical facial pain														
Regional carotidynia														
Cervical arthritis														
Otitis														
Glossopharyngeal and vagoglossopharyngeal neuralgia														
Upper laryngeal neuralgia														
Geniculate neuralgia														
Atypical trigeminal neuralgia														
Sphenopalatine neuralgia														
Vidian neuralgia														
Ciliary neuralgia														
Syndrome of myofascial pain dysfunction														
Fault in dental prostheses														
Pharyngeal and lingual tumors														
Salivary glands diseases														
Disorders of the temporomandibular articulation														
Nonerupted teeth and impacted third molars														

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general anesthesia, with the patient placed in a supine position with the ipsilateral shoulder slightly elevated off the surgical table.¹² The head and neck are extended and rotated to the opposite side. Skin incision is made well below the inferior border of the mandibular ramus parallel to the sternocleidomastoid muscle. After identification and division of the platysma muscle, the deep cervical fascia is incised and the anterior border of the sternocleidomastoid muscle is identified and retracted. A combination of sharp and blunt dissection is used to identify the posterior belly of the digastric muscle. At this point, in contrast to the classical extraoral, which involves identifying and retracting of the carotid artery system, the elongated styloid process is simply palpated beneath the digastric muscle. After retracting this muscle inferiorly and posteriorly, the fascia overlying the surface of the stylohyoid process is divided, the peri-

osteum and the muscle attachments are incised and reflected. The elongated styloid process is removed to a point limited by both surgical access and surgeon's confidence in not damaging any vital adjacent anatomic structures. Wounds are all closed in the traditional manner. The estimated blood loss averages 60 mL (range, 10 to 150 mL). Inpatient hospital time averages 2.8 days, including the day of surgery. The incision may extend from the mastoid process, along the sternocleidomastoid muscle, until the level of the hyoid.¹⁶

More recent works report the use of anesthetic associated with corticosteroids in periodical inoculations in the site, based on the presence of an inflammation. This, combined with an intraoral appliance that restricts eccentric mandibular movement during the healing phase, may be responsible for significant reduction in pain and dysfunction.^{9,33}

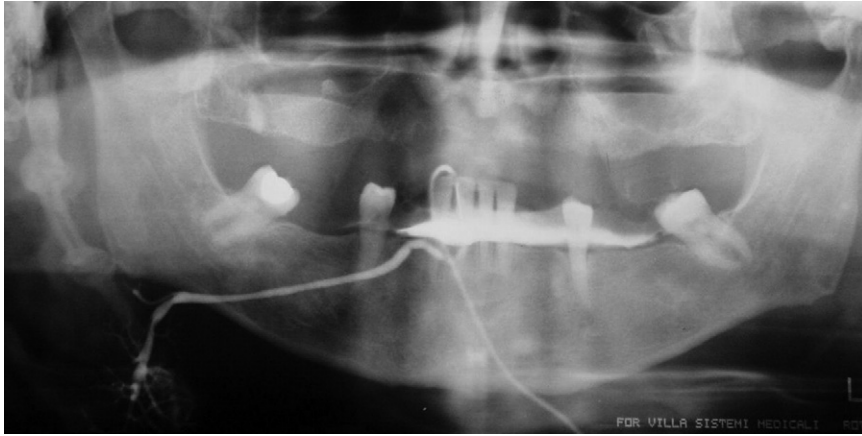


FIGURE 1. Sialography of the right submandibular gland showing the styloid-stylohyoid complex.

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Report of a Case

A 59-year-old Caucasian male was referred to the day care unit of oral lesions of State University of Maringá (Paraná, Brazil) with a suspicion of sialolithiasis or sialadenitis, and presenting with a sialography of the right submandibular gland.

The patient was partially edentulous, with painful symptomatology when turning the head to the right side. At palpation of the region, the right mandibular angle presented pain irradiating to the ipsilateral ear. Cracking at the right temporomandibular articulation was perceived. Submandibular and parotid glands of the right side were pressed to express saliva, and the drainage was normal.

The tonsillar fossa was palpated and worsening of the patient's symptoms was noted. The pharyngeal tonsils were

in place and he presented no prior history of cervical trauma.

At radiographic examination, an ossification of the styloid-stylohyoid complex was noticed along the entire course to the hyoid bone, with 2 pseudoarticulations along it (type II by Langlais et al's²¹ classification; Fig 1).

A 3D CT was requested to confirm the diagnosis of ossification of the stylohyoid ligament (Fig 2).

The surgery was conducted under general anesthesia. The region of the right angle was incised in an anteroinferior direction, following the anterior border of the sternocleidomastoid muscle. The planes were dissected, the sternocleidomastoid muscle was individualized and reflected, followed by retraction of the posterior belly of the digastric muscle. Finally, the maxillary and lingual arteries were individualized and the styloid process exposed with its muscles and ligaments. The periosteum of the entire stylohyoid process was incised and detached. An osteotomy of the complex was performed with a drill as near as possible to the temporal basis, while protecting the adjacent structures. The same was carried out in the inferior part of the process and then excised and measured 4.9 cm (Fig 3).



FIGURE 2. 3D CT of the cervical region.

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FIGURE 3. Excised styloid-stylohyoid complex.

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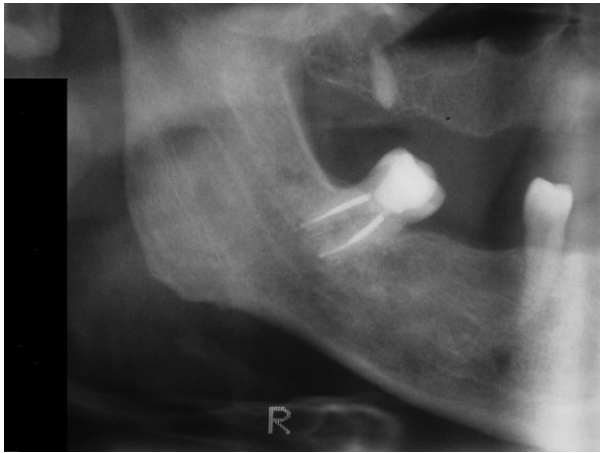


FIGURE 4. Panoramic radiography 2 weeks after surgery.

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Suture of the planes was performed with Vicryl 4-0 and intradermal with nylon 5-0. After 2 weeks, a panoramic radiograph was made (Fig 4).

The specimen was sent for histologic examination, which revealed mineralized material compatible with lamellar cortical bone tissue exhibiting concentric layers, and also with trabecular bone, intermingled by fibrous-adipose medulla, rich in hemorrhagic foci, and marked by lamellas parallel among themselves. In focal areas, remnants of red osseous medulla were observed. The anatomic region coincident with the pseudoarthrosis showed a mineralized tissue of mature appearance, pale stained in irregularly distributed incremental lines, and exhibiting exuberant cellularity. In the periphery of the specimen, a connective tissue rich in collagen fibers was present (Figs 5, 6).

The patient healed uneventfully during the 27-month postoperative period. Minimal scar tissue, no peripheral neurological deficit, and remission of symptoms were found.

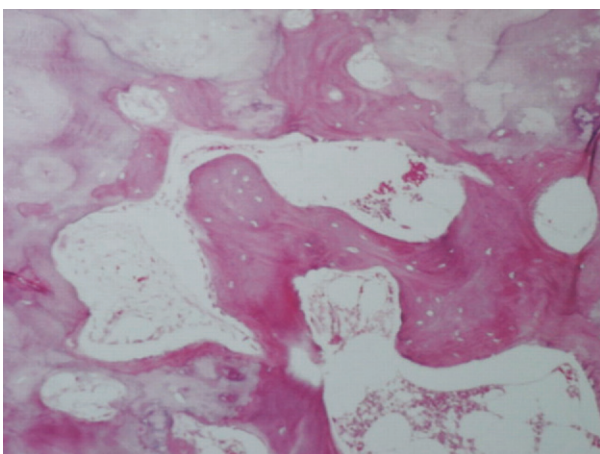


FIGURE 5. Medullar area of pseudoarthrosis, with incremental lines distributed in an irregular way. (Magnification $\times 250$.)

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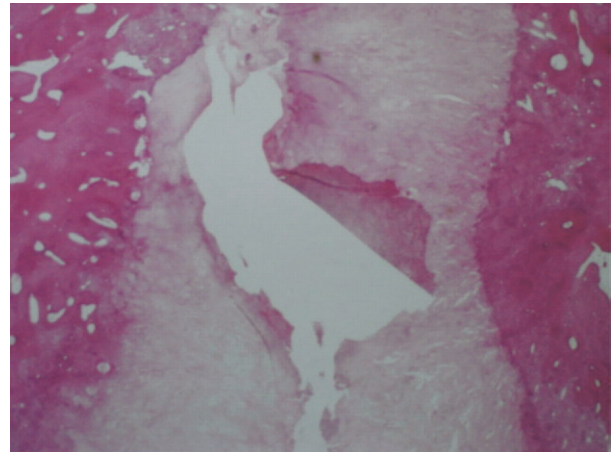


FIGURE 6. In the area of pseudoarthrosis, immature ossified tissue appears, weakly stained. (Magnification $\times 63$.)

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Discussion

Therapeutic approaches and possible etiologic factors of the syndrome described by Eagle in 1937, which includes the classical as well as the carotid artery syndromes, remained unchanged for nearly 30 years. At the end of the 1960s and the beginning of the 1970s new studies were published about the etiology, ossification theories, classification, and treatment options and are still in use today. Conservative approaches involving the use of corticosteroids associated with local anesthetics were considered wrong or inadmissible, but nowadays are performed as routine.⁹ The ossification theories proposed by Steinmann⁵ and Camarda et al¹ divided the previous 2 syndromes described by Eagle into 4 syndromes: the classical (because of the theory of reactive hyperplasia and of reactive metaplasia); the carotid artery (maybe because of the reactional theories, once trauma is not necessarily the cause); the stylohyoid (because of the theory of anatomic variation); and the pseudostylohyoid (from the theory of aging developmental anomaly). Knowledge of all these syndromes is important to define treatment. Conservative treatment is recommended in pseudostylohyoid syndrome, as it is in the beginning of treatment for Stylohyoid syndrome, but can be followed by surgical excision if needed.¹

Etiologies of these syndromes has been altered. The original etiology described by Eagle remains unchanged, suggesting that tonsillectomies and regional traumas are the cause of elongation of the styloid-stylohyoid complex. Nonetheless, cases of patients with no history of prior trauma and with radiographic evidences of elongation in the young (less than 40 years), and those older than 40 years with no prior trauma history but with symptoms, were added.¹

CT offers the chance to plan intervention more precisely. However, use of posteroanterior and panoramic radiographs are still of great help when CT is not possible. In the present case, although the patient already had panoramic radiographies, a 3D CT scan allowed a better perception of the styloid-stylohyoid complex.

The patient was at the age in which the syndrome is normally diagnosed and had signs and symptoms similar to those of Stylohyoid syndrome. According to the classification by Langlais et al,²¹ the case in question fits into type II because it presented with 2 pseudoarticulations along its course.

Extraoral access was chosen because of the extent of the ossification, the lower risk of infection, better visualization, and exposure of the process and its associated structures,¹⁹ especially of the bifurcation of the external carotid with the maxillary artery and superficial temporal artery.²³ If hemorrhage of a larger vessel takes place, it can be handled in a controlled manner and with good visualization. The use of a sterile surgical approach reduces the risk of bacterial contamination.^{12,19} This approach further permits a more ample resection of the styloid process.²³

The disadvantages are the larger dissection and surgical intervention (drains, sutures), presence of a skin scar, the need of general anesthesia, longer recovery, and a possibility of impairing the facial nerve.^{10,13} Tiago et al²³ reported that the scar is aesthetically acceptable.

The authors who advocate the intraoral approach believe it is safe and relatively easy, while avoiding cutaneous scars and extensive dissections.^{6,19,24,25} Surgery can be performed under local anesthesia in a short time and recovery is more rapid. The morbidity and mortality associated with general anesthesia are reduced. Its disadvantages are the possibility of infection of the deep cervical spaces, poor visualization of the surgical field, increased risk of vascular (and its subsequent control), and nerve damage (VII and VIII).^{6,10,19,24} Intraoral access can be performed only if it is possible to palpate the process in the tonsillar fossa.^{12,16}

The patient is still in follow-up, but total remission of the symptoms was noted in the first days of the postoperative period.

Styloid-stylohyoid syndrome is difficult to diagnose because its symptoms are similar to those of other pathologies from areas in addition to the oral and maxillofacial area. Clinical examination and conventional radiographic findings associated with CT are often necessary to exclude other entities with similar manifestations.

Treatment options must be carefully evaluated for each case. Imageological examination of good quality is fundamental for planning and, when indicated, the surgical approach is one of the best treatments.

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Simultaneous Occurrence of 2 Different Low-Grade Malignancies Mimicking Temporomandibular Joint Dysfunction

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The incidence of 2 histologically distinct concurrent malignant tumors in adjacent anatomical regions of the head and neck is uncommon. When the presenting symptoms overlap, it can make their diagnosis very difficult. This article presents a case of a 72-year-old female patient with a left parotid low-grade mucoepidermoid carcinoma and a myxoid fibrosarcoma of the left temporomandibular joint (TMJ) condylar head in which the presenting symptoms were similar to those found on temporomandibular joint disorders (TMD).

Report of a Case

A 72-year-old female patient was referred to our institution in May 2003 from a private otolaryngologist. Her

initial complaint to him consisted of a history of hearing loss for 1.5 years. A mixed hearing loss was demonstrated by audiogram, and a magnetic resonance imaging (MRI) scan was obtained as part of her evaluation. The MRI showed a lesion in the left parotid parenchyma as well as a mass in the left infratemporal fossa (Fig 1). A fine-needle aspiration biopsy of the parotid mass was reported to be consistent with Sjögren's syndrome. The otolaryngologist then referred the patient to Loyola University Medical Center to evaluate the mass on the left infratemporal fossa.

The patient reported feeling a lump behind her left ear for the past 1.5 years. She also complained of hearing clicking noises in the left ear and later in the right ear. She also reported a slight decrease on her mandibular opening, but she was able to maintain a regular diet. The patient denied other constitutional symptoms with the exception of hearing loss. Her medical history was significant for rheumatoid arthritis, Sjögren's syndrome, colon cancer, diverticulitis, and hypothyroidism. Her surgical history included a segmental colon resection, total abdominal hysterectomy, appendectomy, and cataract surgery. She reported allergies to penicillin and sulfa antibiotics. Her medications included Premarin (Wyeth, Philadelphia, PA) 0.3 mg/day, Synthroid (Abbott Laboratories, Abbott Park, IL) 0.125 mg/day, Plaquenil (Sanofi-Aventis, Bridgewater, NJ) 400 mg/day, Bextra (Searle, Skokie, IL) 10 mg/day, Viactiv (McNeil Nutritionals, Ft Washington, PA) 1,500 mg/day, and B-complex 100 mg/day. She reported a 10-year history of smoking a half pack of cigarettes per day, but quit 4 years prior.

On physical examination, she was a well-developed and nourished 72-year-old female in no acute distress. Her vital signs were as follows: blood pressure 135/72 mm Hg, pulse rate 78 beats per minute, and respiratory rate 17 breaths per minute. Her facial examination showed a mild prominence of the left preauricular region. There were no changes in the skin color or texture. There was tenderness on digital

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