Stylohyoid Complex Syndrome

A New Diagnostic Classification

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Objective: To describe stylohyoid complex syndrome (SHCS) as a new diagnostic classification of all lateral neck and/or facial pain conditions resulting from an elongated styloid process, ossified stylohyoid ligament, or elongated hyoid bone. All of these pathologic conditions result in tension and reduced distensibility of the stylohyoid complex (SHC), with resultant irritation of the surrounding cervical structures with movement of the complex.

Design: A retrospective medical chart review was performed to identify a cohort of patients who underwent surgical intervention for lateral neck and/or facial pain due to pathologic SHCS. Follow-up time of greater than 1 year is reported in 5 of 7 patients.

Setting: Tertiary, academic referral center.

Patients: Patients included were those given a diagnosis of SHCS who underwent surgical intervention from June 2006 through September 2009. There were 7 patients, 5 of whom were female. The age range was 38 to 53 years at time of presentation (mean age, 45.3 years). Common presenting complaints were lateral neck and oropharyngeal pain exacerbated by tongue and head movements.

Intervention: The pathologic areas were surgically addressed through transoral or cervical approaches.

Main Outcome Measure: Symptoms following surgical intervention.

Results: Seven patients (8 sides) were identified as having SHCS. Computed tomographic findings included elongated styloid processes (3 sides), ossified stylohyoid ligaments (2 sides), and elongated hyoid bones (3 sides). Computed tomographic scan, frequently with volume-rendered 3-dimensional reconstructions, identified the pathologic condition. All patients experienced clinically significant relief of presenting symptoms following surgical intervention.

Conclusions: Stylohyoid complex syndrome includes all lateral neck and/or facial pain conditions resulting from an elongated styloid process, ossified stylohyoid ligament, or elongated hyoid bone. Surgical intervention directed at any pathologic point to disrupt this complex relieves tension and offers patients relief of symptoms.


The stylohyoid complex (SHC) consists of the styloid process, the stylohyoid ligament, and the lesser cornu of the hyoid bone. The SHC arises from 4 cartilage segments: the tympanohyal, stylohyal, ceratothyal, and hypothyial. The styloid process originates from the temporal bone medial and anterior to the stylomastoid foramen and runs anteromedially, bordered on either side by the internal and external branches of the carotid artery. Three muscles (stylopharyngeus, stylohyoid, and stylomandibular) and 2 ligaments (stylohyoid and stylomandibular) originate at the styloid process, with the stylohyoid ligament and muscle inserting on the lesser cornu of the hyoid bone.

This complex traverses the area from the lateral skull base to the anterolateral neck, forming a band that crosses the upper lateral neck. Pathologic conditions associated with these structures, which can include an elongated styloid process, ossified stylohyoid ligament, or elongated hyoid bone, can cause cervical and pharyngeal symptoms including lateral neck pain in the area of the angle of the mandible, submandibular space, and upper neck that is exacerbated with speaking, swallowing, chewing, yawning, head turning, and other oral and cervical movements. The symptoms are likely related to irritation of the structures around the SHC, including the carotid arteries and the cranial nerves VII, IX, and X. We believe that the pathologic conditions of the individual structures result in reduced distensibility of the SHC, resulting in compression of the surrounding neurovascular structures with movement of the complex. In addition, tightness of the complex can result in a more posterior position of the greater horn of the hyoid bone, bringing it into closer proximity of the neurovascular structures of the lateral neck.

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The first description of a syndrome of vague orocervicofacial pain was in 1937 by Watt Eagle, MD, an otolaryngologist. His first 2 cases were patients with elongated styloid processes presenting with orocervical facial pain. Eagle considered any styloid process longer than the normal adult length of 25 mm to be abnormal and found that 4% of the population had elongated styloid processes, but only 4% of these individuals complained of any symptoms. Subsequent studies have estimated the incidence of elongated styloid process to be as low as 1.4% by Gossman and Tarsitano, who analyzed 4200 panoramic radiographs of men 18 to 22 years old, and as high as 30% by Keur et al in a study on 1135 men and women 18 to 91 years old. Keur et al found the incidence of elongated styloid processes to be equal in men and women, although women displayed symptoms consistent with Eagle syndrome more frequently. Gossman and Tarsitano studied only panoramic radiologic films of young males and acknowledged that most symptomatic patients are women older than 30 years; thus, they stated the incidence of elongated styloid processes is likely closer to 4%.

Eleven years after Eagle first made his diagnosis, he described 2 classifications of the syndrome: the first with the typical symptoms described herein, and the second as a carotid artery syndrome. Patients classified as having the carotid artery syndrome exhibited migraines, cluster headaches, or transient neurological symptoms caused by irritation of the periarterial sympathetic nerve plexus overlying the artery in the area of pain distribution. Eagle syndrome has also been reported in the neurosurgical literature as a cause for transient ischemic attack in a patient presenting with severe reversible narrowing of the internal carotid artery (ICA) on head movement to the affected side. The patient experienced right-sided weakness and syncope when he turned his head to the left, and on angiography there was a filling defect of the cervical ICA at the level C2-3, found to be caused by an osseous structure on imaging. This patient was treated successfully by transcervical styloid process excision with resolution of symptoms.

Camarda et al previously classified this constellation of symptoms into 3 distinct entities: Eagle syndrome as classically described with prior trauma, stylohyoid syndrome, and pseudostyloid syndrome. Stylohyoid syndrome was the most common of the 3, and applied when a patient’s symptoms appeared earlier in life owing to a developmental anomaly of ossified stylohyoid ligaments or elongated styloid processes, with no associated trauma. Pseudostyloid syndrome is caused by tendinosis at the junction of the stylohyoid ligament and the lesser cornu of the hyoid in older individuals with no history of trauma and no evidence of styloid process elongation or stylohyoid ligament ossification on radiologic examination.

We have seen that 3 different pathologic conditions within the SHC—elongated styloid process, ossified stylohyoid ligament, and an elongated hyoid bone—can cause the same symptom complex. Symptoms can include lateral neck and oropharyngeal pain in the area of the submandibular space and deep to the angle of the mandible exacerbated by chewing, swallowing, speaking, and head movement. We believe that the symptoms are related to the tension of the SHC that irritates surrounding structures with head and neck movement owing to the lack of distensibility of the complex. Surgery to interrupt this complex at any point is likely to improve the symptoms by relieving the tension of the SHC. We suggest a new diagnostic classification of stylohyoid complex syndrome (SHCS) to include the previously described Eagle syndrome, stylohyoid syndrome, and pseudostyloid syndrome because all entities originate from pathologic conditions within the SHC. Herein, we review our series of patients with SHCS.

METHODS

A retrospective medical chart review was performed at an academic tertiary care center of patients who presented with lateral neck and/or facial pain and underwent surgical intervention for SHCS from June 2006 through September 2009. Demographic data, presenting symptoms, radiologic findings, surgical procedures, and outcomes were gathered. Patients were asked to rate the percentage improvement of their pain symptoms at the first postoperative visit approximately 2 weeks after surgery and at all subsequent visits.

RESULTS

Seven patients (8 sides) were identified. The age range was 38 to 53 years at the time of presentation (mean age, 45.3 years). Five of the 7 patients were female. Common presenting complaints were lateral neck and oropharyngeal pain exacerbated by tongue and head movements, with tenderness over the affected structures when palpated during these activities. All patients underwent computed tomographic (CT) scan of the neck with contrast with multiplanar reconstructions, and when necessary 3-dimensional volume-rendered reconstructions were performed of the bony anatomy to show spatial relationships. The pathologic conditions identified on CT imaging included 3 elongated styloid processes (1 bilateral), 2 ossified stylohyoid ligaments, and 3 elongated hyoid bones. One of these patients had bilateral ossified stylohyoid ligaments, and 1 had bilateral elongated styloid processes, but each underwent unilateral resection due to unilateral symptoms. Two patients had a history of trauma or tonsilllectomy.

The surgical approach was determined by the location of the pathologic conditions along the SHC identified on examination and imaging. All operations were performed in a same-day surgical suite under general anesthesia, and patients were given perioperative antibiotics. The surgical approaches were as follows. The transoral approach was used to resect the elongated styloid processes. The styloid process was found by palpation of the tonsillar fossa. The overlying mucosa and superior constrictor muscle were incised vertically and the styloid process dissected out and skeletonized using a right-angled clamp. If necessary for exposure, a standard tonsillectomy was performed first. The stylohyoid ligament was isolated and resected off of the tip of the styloid process. The styloid process was then transected as high as possible. The superior constrictor muscle and mucosa were closed with interrupted absorbable sutures.
Ossified stylohyoid ligaments and elongated hyoid bones were resected through cervical incisions. In cases in which the stylohyoid ligament was ossified, an approach similar to that used for a submandibular gland resection was used. A horizontal incision was placed in a skin crease 2 to 3 cm inferior to the mandible, and dissection was carried down to the submandibular gland, taking care to protect the marginal mandibular branch of the facial nerve. The submandibular gland was reflected superiorly and the parapharyngeal space identified. The ossified ligament was identified by palpation, isolated from the surrounding structures with blunt dissection, and isolated along its entire length from its origin at the styloid process to its insertion onto the hyoid bone. The entire ligament was resected en bloc with the lesser horn of the hyoid bone and the tip of the styloid process. This resulted in immediate release of the tension in the SHC and an inferior release of the hyoid bone.

For those patients with an isolated elongated hyoid bone, the surgical approach involved a submandibular approach with a slightly more anterior incision. The greater horn of the hyoid bone was identified, and the lateral portion of the hyoid bone, including the greater and lesser horns, was skeletonized and resected.

All patients were discharged home the same day with oral pain medication and oral antibiotics for 7 to 10 days. No steroids were used. Those patients who underwent a transoral approach were placed on a soft diet. No intraoperative or postoperative surgical complications were encountered. One patient required an overnight readmission for inability to tolerate oral diet. Five patients experienced complete resolution, and 2 patients had partial resolution of symptoms within 2 weeks following surgical intervention. Of the 2 patients with partial resolution, 1 developed complete alleviation of symptoms within 3 months, the other had 75% improvement in presenting symptoms. The follow-up period was over 1 year in 5 of 7 patients.

The differential diagnosis of patients with lateral orocervicofacial pain includes, but is not limited to, glossopharyngeal neuralgia, trigeminal neuralgia, temporomandibular joint dysfunction, temporal arteritis, salivary gland disease, chronic tonsillitis, tumors of the pharynx and tongue base, laryngopharyngeal reflux, dental disease, carotidynia, atypical migraine, otitis media, otitis externa, and mastoiditis. SHCs should be considered in patients presenting with persistent lateral neck, throat, or sub-

Figure 1. Identifying pathologic conditions in the stylohyoid complex. Coronal computed tomographic (CT) multiplanar reconstruction (A) and 3-dimensional volume-rendered CT reconstruction of patient with symptomatic elongated styloid processes (B).

Figure 2. A 3-dimensional volume-rendered computed tomographic reconstruction of a patient with symptomatic elongated hyoid bone approximating the carotid bulb.
mandibular pain that cannot be explained by other etiologies. SHCS should be suspected if symptoms are triggered by any activity that requires elevation or rotation of the hyoid bone, such as head movements, chewing, speaking, swallowing, or tongue movement. The symptoms are thought to be caused by irritation or impingement of the cranial nerves VII, IX, or X, the carotid arteries, or the submandibular gland, all of which are in close proximity to the SHC. We believe that the tension in the SHC is greatly increased with these pathologic conditions, resulting in a minimally distensible SHC. This affects the ability of the hyoid bone to elevate, depress, and rotate with head and oral movements, thereby putting more pressure on the surrounding neurovascular structures with these movements and causing vague pain symptoms. Because of the vagueness of the symptoms, many patients are treated for multiple different problems by many different specialists, including dentists, neurologists, gastroenterologists, and psychiatrists.

The workup of patients with suspected SHCS should include a thorough medical history, as well as a complete examination of the head and neck to rule out other diagnoses. Care should be taken to try and reproduce symptoms. Palpation over the components of the SHC while the patient performs cervical and oral movements can reproduce and localize the pain. Palpation of the tonsillar fossa for an elongated styloid process should also be performed. Some authors suggest confirming the diagnosis with infiltration of local anesthetics into the tonsillar bed in an attempt to relieve pain symptoms. This procedure can be valuable, but it may not differentiate SHCS due to an elongated styloid process from other pathologic conditions, such as glossopharyngeal neuralgia.

A CT scan is used to confirm the diagnosis. We have used 3-dimensional (3D) CT volume-rendered reconstructions to better delineate the size of the structures in the SHCS and their relationships to surrounding neurovascular structures. We have found this technique to be invaluable in helping to identify pathologic conditions in the SHC (Figure 1 and Figure 2). Three-dimensional CT is considered by most to be the radiologic test of choice for diagnosis of SHCS because it is the most advanced technique available to definitively measure the length of the styloid process and hyoid bone. Multiplanar reconstruction of CT images has also been of use when 3D reconstructions are not available. Many of our patients had arrived with plain film imaging, most commonly panoramic radiologic images, but we find these images to be much less valuable than CT scans.

Surgical intervention aimed at the pathologic point along the SHC is the mainstay of treatment. Two traditional surgical approaches to the styloid process can be used: the transoral approach and the extraoral/cervical approach. The transoral approach is easily performed, with multiple advantages over the external cervical approach including less tissue dissection, shorter surgical time, and no external scarring (Figure 3). The recovery is the same as with a tonsillectomy. A cervical incision is the standard approach used both for ossified stylohyoid ligament and for an elongated hyoid bone and may be necessary if the styloid process is extremely large or heavily ossified (Figure 4). This approach offers the advantage of a larger exposure to the pathologic area and a clean surgical field and causes less swallowing discomfort than the transoral approach. However, it requires more dissection and slightly more recovery time for the patient. Medical treatment with medications aimed at neuropathic pain, such as anti-inflammatory or anti-epileptic drugs, are of limited benefit but may be of use when a patient is not a surgical candidate.
No complications cited in the literature of persistent pain, trismus, bleeding, infection, or neurovascular injury were encountered in our study within the follow-up period. One patient who underwent transoral resection of bilateral elongated styloid processes required overnight observation with intravenous hydration due to swallowing discomfort.

All patients experienced eventual resolution of their pain symptoms within the follow-up period, with most patients achieving relief within 2 weeks of surgery. Cure rates in the literature range from approximately 80% to 100% following surgical intervention. No patients reported difficulty with swallowing function following resection of the hyoid.

In conclusion, we advocate the term stylohyoid complex syndrome to include all conditions characterized by lateral orocervicofacial pain resulting from an elongated styloid process, ossified stylohyoid ligament, or elongated hyoid bone. These particular pathologic conditions all result in reduced distensibility of the SHC, inhibiting normal elevation and depression of the hyoid bone with neck and oral movements, resulting in lateral orocervical pain from compression and irritation of the neurovascular structures around the SHC. Surgical intervention directed to disrupt this complex relieves tension and offers patients relief of symptoms.

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REFERENCES