



# Styloid-stylohyoid syndrome: a rare cause of cranio-facial pain—a retrospective case series of 12 patients

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## Abstract

**Purpose** Elongated styloid process results in severe cranio-facial/cervico-facial pain. The purpose of this study is to determine the efficacy of treatment outcomes using transcervical approach in the management of styloid-stylohyoid syndrome/styloid syndrome.

**Study design** This is a retrospective cohort study. Subjects were enrolled from out-patient clinics reporting between the periods Jan 2016–Jan 2018.

**Methods & methods** Twelve patients diagnosed with styloid syndrome based on history, thorough clinical workup, and assessment were included in the study. The primary outcomes—improvement in pain and regression of chief complaints following surgery were assessed. Elongated styloid was resected using transcervical approach under general anesthesia.

**Results** The intra-operative lengths of the styloid process varied from 40 to 43 mm. No post-operative complications were encountered and the symptoms regressed completely after surgery in all the patients at follow-up of 6 months.

**Conclusions** Transcervical styloidectomy is an effective treatment for exposure and resection of the styloid process with minimal complications in patients with stylo-stylohyoid syndrome.

**Keywords** Styloid-stylohyoid syndrome · Cranio-facial · Styloidectomy · Transcervical

## Introduction

Elongation of the styloid process and/or calcification of the stylohyoid ligament as described by Watt Eagle in 1937 is referred to as Eagle's syndrome/stylo-stylohyoid syndrome/styloid syndrome/stylagia [1]. Of patients with elongated styloid process, 4–10% report with vague symptoms such as cranio-facial pain due to its impingement on adjacent arteries and cranial nerves [2]. Medial elongation could result in interference with tonsillar fossa and more severe symptoms of cranio-facial pain [3, 4]. The frequency of this syndrome is estimated to be 4–8 per 10,000 people [5]. Patients often

present with vague symptoms such as craniofacial pain,odynophagia, dysphagia, tinnitus, cervicofacial pain, and globus sensation [6]. The etiology of pain can be conceptually explained by the impingement of styloid process at diverse angulations on various cranial nerves (V, VII, IX, and X) and adjacent vessels. Lateral deviation may cause impingement on external carotid artery. Medial angulation often interferes with the tonsillar fossa whereas posterior angulation may entrap the last four cranial nerves between elongated styloid and transverse process of atlas [7, 8]. Post-tonsillectomy fibrosis at the styloid apex that can compress neighboring nervous structures is also a known cause of stylo-stylohyoid/styloid syndrome [9].

Eagle's syndrome is a diagnosis of exclusion that can be corroborated by a thorough history and presentation with typical symptoms, physical exam that reveals a palpable styloid process in the tonsillar fossa, and a neck CT scan or panorex that reveals consistent anatomy [10]. A lidocaine infiltration test in the tonsillar fossa at the site of elongated styloid is often diagnostic. The diagnosis is confirmed if the symptoms subside temporarily. While the literature does address the surgical management of Eagle's syndrome, there is no definitive

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consensus on whether the transoral or transcervical approach is superior. Furthermore, the indications for surgical intervention are not standardized; thus, surgical treatment of Eagle's syndrome has led to mixed results. The purpose of this study was to evaluate treatment outcomes in patients diagnosed with stylo-stylohyoid syndrome treated surgically using transcervical approach at our tertiary care institution.

## Materials and methods

We conducted a retrospective cohort study of 12 patients who were diagnosed with Eagle's syndrome (Fig. 1) and treated with surgical resection of the styloid process at a single tertiary care institution between January 2016 and 2018. Patients were evaluated for a spectrum of complaints including pain in the neck, throat, or ear; headache; and dysphagia. Complete data were collected for all 12 patients. Follow-up of the patients varied from 6 months to 1 year. Patients were diagnosed with Eagle's syndrome based on thorough clinical workup and assessment, including history, physical exam, and imaging (CT and panorex films). Diagnosis was formulated based on the patient symptoms, palpation of elongated styloid process overlying the tonsillar pillar or temporary relief of symptoms following administration of local anesthetic. Transcervical approach was used in all cases (Fig. 2). A 5-cm horizontal incision was made below the angle of the mandible. The deep cervical fascia was incised and the stylohyoid muscle and posterior belly of digastric are identified overlying the carotid



Fig. 1 3-D CT scan showing elongated styloid process



Fig. 2 Transcervical incision to show elongated styloid process

arteries. The styloid process was identified by retracting the muscles postero-inferiorly and the mandible anteriorly, carefully dissecting in the periosteal plane to the base of skull (Fig. 3). Data for these patients were extracted from the electronic medical records and used to characterize the population based upon an array of variables, such as chief complaint (severity and location of pain in particular), surgical approach, and postoperative outcomes. The outcome of the surgery which is improvement in craniofacial pain with complete resolution of symptoms was thoroughly assessed at follow-ups.

## Results

A total of 12 patients were identified as having been diagnosed with and surgically treated for Eagle's syndrome



Fig. 3 Graphic illustration of intra-operative view of elongated styloid process

**Table 1** Distribution of age, gender, chief complaints, and remission status of the patient

Patient	Age/sex	Clinical features	Treatment done	Remission status (after 6 months)
1	42/M	Rt. temporal headache, foreign body sensation inside neck, atypical pain in neck radiating to the side of face and head	Transcervical styloidectomy Rt side	Complete remission
2	49/F	Pain on movement of head towards Rt direction, bilateral head ache located at the temporal region. Radiating pain towards the back side of neck	Transcervical styloidectomy Rt side	Complete remission
3	51/M	Retromandibular pain bilaterally, radiating to temporal region and behind the eyes.	Transcervical bilateral styloidectomy	Complete remission
4	47/M	Foreign body sensation inside throat, pain while movement of head	Transcervical bilateral styloidectomy	Complete remission
5	28/M	Continuous burning sensation and pain inside neck, dizziness	Transcervical bilateral styloidectomy	Complete remission
6	32/F	Headache, dizziness, and tenderness at the greater cornua of hyoid bone region	Transcervical bilateral styloidectomy	Complete remission
7	55/M	Ipsilateral burning sensation inside the throat, glossodynia, and pain in tonsillar fossa	Transcervical bilateral styloidectomy	Complete remission
8	32/M	Temporal head ache, tenderness of lateral side of head, dizziness and fainting	Transcervical bilateral styloidectomy	Complete remission
9	43/F	Atypical facial pain and head ache, burning sensation inside throat, relief with corticosteroids for a short period	Transcervical bilateral styloidectomy	Complete remission
10	29/M	Bilateral head and neck pain, tenderness of scalp at the temporal region, occasional sharp shooting pain in throat	Transcervical bilateral styloidectomy	Complete remission
11	39/M	B/L temporal headache, excruciating pain in the throat	Transcervical bilateral styloidectomy	Complete remission
12	29/M	Headache, tenderness of scalp, irritation in the throat Rt side	Transcervical styloidectomy Rt side	Complete remission

during the enrollment period (Table 1). The median age among these patients was 39.6 years, while there was a male pre-ponderance (75% males). The two types of Eagle's syndrome that have been described are classic and carotid artery. Pain in the classic type is believed to be the result of stimulation of cranial nerves V (trigeminal), VII (facial), IX (glossopharyngeal), and X (vagus). In contrast, the carotid artery type is characterized by the styloid process adversely affecting the carotid nerve plexus. All of the patients in this data set were diagnosed with the stylo-carotid type of Eagle's syndrome with no carotid nerve plexus involvement. No patient had previous history of tonsillectomy.

Right-sided pain and bilateral was more common than left-sided pain. The most common locations of pain were the neck (80%), head (62%), and ear (48%), while the jaw, face, and throat were less common (29% each). Other less common symptoms included dysphagia (24%) and odynophagia (24%). The median duration of symptoms prior to surgery was 2 years, and none of the patients had a history of prior tonsillectomy. During surgery, the transcervical approach was done in all cases. The styloid process along with the stylohyoid ligament was resected in every case (100%) (Fig. 4). The median length

of resection was 3.0 cm. The median length of procedure was 81 min, with the transcervical approach. Nearly all patients experienced improvement in their pain (90%), while 62% of patients experienced our definition of a clinically meaningful difference with a complete resolution of their pain. For those patients experiencing a

**Fig. 4** Resected styloid process

positive response to surgery, the median time to response was 18 days; median follow-up time was 3.0 months.

## Discussion

Classical Eagle's syndrome generally follows a traumatic event such as tonsillectomy, which further stimulates the styloid process resulting in overgrowth and pharyngeal symptoms such as dysphagia and odynophagia, with pain being referred to the ipsilateral ear; however, it is believed that this trauma is not responsible for the ossification of the stylohyoid apparatus [11]. The second syndrome, the stylocarotid syndrome, is unrelated to tonsillectomy. Mechanical irritation due to impingement and stimulation of the sympathetic plexus in the walls of the external and/or internal carotid arteries by the styloid tip or ossified ligament provide a diverse array of symptoms such as referred pain in the respective region of vascularization along with vague symptoms such as tinnitus and otalgia often headache which may confuse the clinician leading to misdiagnosis or inadequate treatment. The pain associated with TMJ, atypical cranio-facial pain, or tooth related pain may also lead to mis-diagnosis of Eagle's syndrome [12].

Such misdiagnosis/inadequate treatment of these patients with Eagle's syndrome may lead to various unnecessary surgical treatments that may have a negative psychosocial impact on the patient [11]. An unresolved craniofacial pain or cervicofacial pain treated with various dental procedures must prompt the clinician to think of stylo-stylohyoid syndrome. The aim of this study was to present our treatment protocol using transcervical approach in the management of patients with stylo-stylohyoid syndrome.

CT scan is a useful aid in defining the length, angulation, and anatomic relationship of the styloid process [13]. An orthopantomograph and CT scan were obtained in all cases in our study; however, the medical history was the main guide in the diagnosis of the syndrome. Relief of pain following lidocaine infiltration confirmed the diagnosis and confirmed the need of surgical procedure.

There are certain alternative treatments such as transpharyngeal infiltration of lidocaine or steroids and manual transpharyngeal fracture of the styloid process. None of the patients in our series had any previous history of any pharmacological alternative treatments. Surgical treatment is definitive and remains the mainstay in the management of elongated styloid process.

Various surgical approaches have been described in the literature on the management of elongated styloid process. The intra-oral approach is often fraught and is associated with higher risk of damage to neuro-vascular structures due to the restricted operative field [14, 15]. Extraoral

approach allows complete exposure of the styloid process and facilitates better and complete resection and reduced chances of deep neck space infections [16]. The transcervical approach allows excellent access to the styloid process offering adequate field of operation and assessment of vital structures, hence minimal complications. Layer by layer dissection was carried out to reach the lower border of the mandible and the elongated tip of the styloid was identified using digital palpation and the ligaments attached to the styloid tip were carefully separated from the process and the elongated styloid process was completely detached from the temporal bone. Transcervical approach overcomes the limitations of the intraoral approach and provides easy accessibility and safe resection of the styloid process.

## Compliance with ethical standards

**Conflict of interest** The authors declare that they have no conflict of interest.

**Informed consent** Yes.

**Ethical approval** Ethical approval was obtained from the institutional ethical committee.

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