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Original Article

Tonsillo - styloidectomy for Eagle's syndrome: review of 20 cases

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Abstract:

Objective: To study the clinical, radiological along with the management outcome of Eagle's syndrome.

Study design: Prospective.

Setting: Department of Otolaryngology - Head and Neck Surgery, Ibn Sina Medical College Hospital and Ibn Sina Hospital, Dhaka, Bangladesh.

Methods: This study included twenty patients with Eagle's syndrome presented to Ibn Sina Medical College, Dhaka and Ibn Sina Hospital, Dhaka, from April 2009 to January 2012. The data of each patient included age, sex, presenting symptoms and signs, radiological investigations, operative notes and state at follow up.

Results: Out of twenty patients, twelve (60%) were female and eight (40%) were male. Maximum nine patients (45%) belonged to the fourth decade. The mean age of presentation was 42.5 years. Maximum patients (100%) presented with pain and foreign body sensation throat. In all cases, a sharp prick was felt and pain was increased on palpation of the upper part of tonsillar fossa. In fourteen cases (70%) elongated styloid process was bilateral and in six cases (30%) it was unilateral. X-ray Towne's view, lateral view of skull base & neck and CT scan were excellent diagnostic tools as well as to measure the length of styloid process. In all cases partial styloidectomy was done via intra-oral approach. Eighteen patients (90%) were symptom free in three months follow up. Two patients (10%) had pain in throat and other symptoms even after three months follow up and were treated with oral carbamazepine.

Conclusion: Eagle's syndrome associated with elongated styloid process is a rare clinical entity. The diagnosis can easily be made with clinical examination and radiological findings. Awareness of this syndrome is important to all ENT practitioners and related specialty involved in diagnosis and treatment of Head and Neck pain.

Key words: *Eagle's syndrome; styloidectomy; temporal bone; ossification*

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Introduction:

In 1937, Watt W. Eagle, an ENT surgeon at Duke University, described the first cases defined as "stylalgia" secondary to elongated styloid process or calcification of the stylohyoid ligament or to mineralization of the stylohyoid ligament[1]. Eagle's syndrome is defined as secondary glossopharyngeal neuralgia due to elongated styloid process, resulting from abnormal stylohyoid chain ossification[2]. Usually asymptomatic, it occurs in adult patients ranged from 30 to 50 years[3]. The stylohyoid complex is made of styloid process, stylohyoid ligament and the small cornus of the hyoid bone. All these structures are derivate from Reichert's cartilage of the second branchial arch. The styloid process is an elongated conical projection of the temporal bone that lies anteriorly to the mastoid process, between the internal and external carotid arteries and laterally the tonsillar fossa. In this space, the internal carotid artery, the internal jugular vein, the facial, glossopharyngeal, vagus and hypoglossal nerves are located. From the styloid process, the stylohyoid, the styloglossal and the stylopharyngeal muscles and the stylohyoid and the stylomandibular ligaments originate. The symptoms of Eagle's syndrome are foreign-body sensation in the throat, dysphagia, intermittent facial pain, tinnitus, neck pain and otalgia[4].

There are lots of other symptoms also. Eagle described a group of patients who had symptoms of intermittent and nagging pain in the pharynx that radiated to the mastoid region, a foreign-body sensation in the throat, dysphagia and taste disturbance[5]. His original patients had a history of tonsillectomy that resulted in scar tissue in the tonsillar fossa. Eagle considered tonsillectomy responsible for the formation of scar tissue around the styloid apex with consequent compression or stretching of the vascular and nervous structures contained in the retrostyloid compartment (in particular, the glossopharyngeal nerve and perivascular carotid sympathetic fibers)[5].

However, Eagle had also discovered in patients who have never been subjected to tonsillectomy[5]. One should have a high level of suspicion when neurological symptoms occur upon head rotation. Symptoms tend to be worsened on bimanual palpation of the styloid through the tonsillar bed. An elongated styloid process occurs in about 4% of the general population, while only a small percentage (between 4-10.3%) of these patients is symptomatic. So the true incidence is about 0.16%[6], with a female-to-male predominance of 3:1[6].

Bilateral involvement is quite common but does not always involve bilateral symptoms[6]. No significant difference is detectable between the right and left sides. More important than the elongation of the styloid process and the calcification of the stylohyoid ligament is the thickening or ossification of those structures[7]. The cause of stylohyoid calcification is not well understood, but it might be related to congenital factors such as persistence of a cartilaginous analog or an embryonic precursor to the styloid process. Other possible causes include previous trauma or an inflammatory process that produces a proliferation of granulation tissue and results in calcification or ossification[7]. Calcification can lead to compression of the adjacent structures that are innervated by the glossopharyngeal, trigeminal and the chorda tympani nerve. There might also be impingement of the plexus of the carotid sheath that produces irritation of the sympathetic nerves[7]. Diagnosis is made both radiographically and by physical examination. Palpation of the styloid process in the tonsillar fossa is indicative of elongated styloid process in that,

styloid process of normal length is not normally palpable[8]. Palpation of the tip of the styloid should exacerbate existing symptoms.

If highly suspicious for Eagle's syndrome, confirmation can be made by radiographic studies. Most frequently, X-ray Towne's view and lateral view of the skull and upper neck are used to determine whether the styloid process is elongated[9]. CT scan is very important to measure the length of styloid process[10]. The normal length of the styloid process is individually variable but it is approximately 20-30mm[10]. The styloid process elongation can be assumed if its length is more than 30mm[10]. Although Eagle's syndrome is thought to be caused by an elongated styloid process or calcified stylohyoid ligament, the presence of an elongated styloid process is not pathognomonic for Eagle's syndrome because many patients with incidental findings of an elongated styloid process are asymptomatic. Lateral view radiographs of the skull and an anteroposterior view radiograph can be obtained to determine whether there is any lateral deviation of the styloid[9].

Medical treatment includes analgesics, antidepressant and anticonvulsants. Partial styloidectomy is the treatment of choice[11]. Styloidectomy can be performed by an intra- or an extra oral approach[12]. The intra oral approach may result in a restricted operative field, in the possibility of an incomplete control over many important vascular and nervous structures and in the risk of deep cervical infections. On the other hand, external surgical approach results in skin scars, longer hospitalization and risks of facial nerve injuries[12]. The choice of treatment usually depends on the experience of the surgeon.

Materials:

This study included twenty patients with Eagle's syndrome presented to Ibn Sina Medical College, Dhaka and Ibn Sina Hospital, Dhaka, from April 2009 to January 2012. The data of each patient included age, sex, presenting symptoms and signs, radiological investigations, operative notes and state at follow up. X-ray Towne's view, lateral view of skull & upper neck and CT scan were done in all cases. Postoperative follow up has been done up to 3 months in each case.

Results:

Out of 20 cases, 12(60%) were females and 8 (40%) were males 6 (30%) patients belonged to the third, 9(45%) patients to the fourth, 4 (20%) patients to the fifth and 1(5%) patient to the sixth decade. The youngest patient was 28 years and the oldest was 55 years. The mean age was 42.5 years. All patients presented with pain and foreign body sensation of throat. Other common presentations were otalgia, headache, dysphagia, pain on opening mouth and globus pharyngeus (Table-I).No patient had history of cervicopharyngeal trauma. The patient with 45 mm long styloid process had the maximum symptoms, while the patients with 32 mm had the minimum symptoms. The mean duration of symptoms was 14 months.

Table-I
Presenting symptoms.

Symptoms	No. of patients	Perchantage (%)
Pain in throat	20	100%
Foreign body sensation throat	20	100%
Otalgia	14	70%
Headache	8	40%
Dysphagia	4	20%
Pain on opening mouth	3	15%
Globus pharyngeus	2	10%

N.B.: More than one symptom may present in same patient.

Table-II
Involved side/s

Side(s)	No. of patients	Percentage (%)
Unilateral	6	30%
Bilateral	14	70%

Table-III
Mean length of styloid process.

Sex	Mean length of styloid process
Male	35.3 mm
Female	32.5 mm

Table-IV
Post-operative results after 3 months.

	No. of patients	Percentage (%)
Symptom free	18	90%
Symptom persists	2	10%

Examination of the oropharynx revealed signs of chronic tonsillitis in 2 cases. These 2 cases were diagnosed as Eagle's syndrome simultaneously, because pain didn't coincide with symptoms of chronic tonsillitis. On palpation of the upper part of the tonsillar fossa, a sharp prick was felt and pain was increased. In these 3 cases tonsillectomy has been done previously by other surgeon. In these three cases pain started during the first 2 weeks of tonsillectomy. We have differentiated these cases from the classic post-tonsillectomy pain. Post-tonsillectomy pain becomes progressively less during the first week following surgery, whereas in Eagle's syndrome pain remains at the same intensity level. In all cases styloid process has been palpated and found to be elongated. Table-I shows, in 14 cases elongated styloid process was bilateral and in 6 cases it was unilateral. Diagnosis was made with radiograph. X-ray Towne's view and lateral view of the skull base and upper neck were done in all cases. CT scan has been done in all cases. Measurement of styloid process has been done in CT scan and was tabulated. Mean length of styloid process in male and female was 35.3 and 32.5 mm respectively (Table-III). Medical treatment (carbamazepine for 3 months) has been failed in all cases. Final diagnosis was made on the operating table following tonsillectomy. In all 20 cases, intra-oral approach has been used. In all cases, tonsillectomy followed by partial styloidectomy (bilateral/ unilateral) was done except for 3 cases where only partial styloidectomy was done as tonsillectomy has been done previously by other surgeon. In 14 cases bilateral styloidectomy and in 6 cases unilateral styloidectomy has been done. One patient had secondary haemorrhage 7 days after operation which has been medically treated. Postoperatively 18 patients were symptom free in three months follow up. 2 patients didn't relieve of pain and was treated with oral carbamazepine (Table-IV).

Discussion:

Eagle's Syndrome or stylalgia caused by elongated styloid process is an uncommon and under diagnosed clinical entity. Eagle described it as a syndrome complex mainly in two varieties[5]. The classical variety presents as pain in the throat, referred to as otalgia and foreign body sensation in the throat[5]. A second variety is styloid process compressing the carotid artery presenting as carotodynia, headache and dizziness[5]. He found that these patients were relieved of symptoms by shortening the styloid process. Literature shows that females are affected more than the males[6]. In our study, out of 20 patients, 12 (60%) were females and 8 (40%) were males. Older patients are more commonly affected[3]. In the present

study, 6(30%) patients belonged to the third, 9(45%) patients to the fourth and 4 (20%) patients to the fifth and 1 (5%) patient to the sixth decade. The youngest patient was 28 years and the oldest was 55 years. The mean age was 42.5 years.

The clinical symptom with which the patient presents is due to compression of the adjacent nerves, mainly the glosso-pharyngeal, lower branch of trigeminal and the chorda tympani. Symptoms of styalgia may be due to previous trauma or an inflammatory process that proliferate the granulation tissue, resulting in the calcification or ossification of the stylohyoid ligament[7]. Eagle's Syndrome that follows a tonsillectomy procedure is characterized by symptoms of dysphagia, pain, referred otalgia and foreign body sensation in the throat[13]. Healing tonsillectomy scar tightens the mucosa across the tip of the elongated styloid process. Upon normal function such as yawning, eating and swallowing, the movements of this mucosa across the styloid process give rise to symptoms. In this study, 100% patients presented with pain and foreign body sensation of throat. Other common presentations were otalgia, headache, dysphagia, pain on opening mouth and globus pharyngeus. No patient had history of cervicopharyngeal trauma. The length of styloid process correlates with the clinical signs and symptoms[14]. In the present study, the patient with 45 mm long styloid process had the maximum symptoms, while the patients with 32 mm had the minimum symptoms.

In all cases styloid process has been palpated and found to be elongated. In 14 cases elongated styloid process was bilateral and in 6 cases it was unilateral. Imaging is important and is diagnostic. X-ray Towne's view and lateral view of the skull base and upper neck were done in all cases. CT scan has been done in all cases. Visualizing the styloid process on a CT scan with 3D reconstruction is the suggested imaging technique[15]. The enlarged styloid may be visible on an orthopantogram or a lateral soft tissue X- ray of the neck. The normal length of the styloid process varies greatly, as follows:

- From 1.52-4.77 cm, according to Moffat et al (1977)
- Less than 3 cm, according to Kaufman et al (1970)
- From 2-3 cm, according to Lindeman (1985)
- Less than 2.5 cm, according to Correl et al (1979), Langlais et al (1986), and Montalbetti et al (1995)
- Less than 3 cm, according to Monsour and Young (1986)
- According to Balcioglu (2009), the mean length of the styloid processes of the subjects reporting Eagle syndrome is reported to be 25 +/- 4.72 mm.

In the study, mean length of styloid process in male and female was 35.3 and 32.5 mm respectively. The length of the styloid process is better demonstrated on lateral views because there is less superimposition[9]. Despite the striking radiographic appearance of an ossified and elongated stylohyoid process, the significance of this abnormality has not been appreciated except by otolaryngolists who are familiar with the symptom complex[9].

Treatment is mainly surgical, where the elongated styloid process is shortened by trans-tonsillar or by external approach[11]. Eagle initially described tonsillo-styloidectomy by the intra-oral route, where a trans-pharyngeal approach was used to shorten the styloid process after tonsillectomy. There is no external scarring

with this approach and low post operative morbidity and complications. The surgeon locates the styloid process by digital palpation of the tonsillar fossa. After the incision and the identification of the styloid process, it is necessary to split the muscles, to elevate the mucoperiosteum, and finally, to fracture and excise the styloid process. If the pharyngeal tonsil is present, performing tonsillectomy first during the same operation is necessary. The advantages of the intra oral approach are that the method is safe, simple, less time consuming and an external scar is avoided. The disadvantages are possible infection of deep neck spaces, risk of injury to major vessels and poor visualization. Styloidectomy can also be performed by the extra-oral approach through an incision that extends from the mastoid process along the sternocleidomastoid to the level of the hyoid, then up across the neck to the midline of the chin. This approach is aesthetically less pleasing with more morbidity compared to the intra-oral route[12]. In the present study, in all 20 cases, intra-oral approach has been used. In all cases, tonsillectomy followed by partial styloidectomy (bilateral/ unilateral) was done except for 3 cases where only partial styloidectomy was done as tonsillectomy has been done previously. In 14 cases bilateral styloidectomy and in 6 cases unilateral styloidectomy has been done. Literature shows success rate of partial styloidectomy in Eagle's syndrome is 80-90%[16, 17]. In our study, 18 patients (90%) were symptom free in three months follow up. 2 patients didn't relieve of pain and was medically treated.

Conclusion:

Eagle's syndrome describes a syndrome in which there is elongation of stylohyoid process with associated symptoms. Diagnosis can easily be made with clinical examination and radiographic imaging. Tonsillo-styloidectomy is the treatment of choice and has proven to relieve the patients of styalgia. Awareness of this syndrome is important to all ENT practitioners and related specialty involved in diagnosis and treatment of Head and Neck pain.

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