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Active surgical management of pharyngodynia in cases attributable to styloid complex

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Abstract
Objective/Hypothesis: To describe the role of active surgical management in cases of pharyngodynia attributable to styloid enlargement. The purpose of this study is to assess the postoperative recovery time and improvement in pain score.
Study design: Retrospective-Prospective analysis of a case series.
Methods: Twenty seven patients of chronic Pharyngodynia with no other discernible cause except a possible association with styloid process directly or indirectly were analysed. Nineteen patients were operated using established technique of Tonsilostyloidectomy, with slight modifications. A minimum follow-up of 12 weeks was considered. Main outcome measures were the postoperative pain score and time taken to wean off drugs.
Results: The incidence of stylgia, classical Eagles syndrome, was found to be 0.44 per 1000 ENT OPD visiting people at MMABM Hospital. The mean age at presentation was found to be 40.70 years. The Females cases were more than twice as common as males. The average duration of symptoms was 22 months at presentation, and bilateral symptoms were found in 9 out of 10 patients, with left and right predominant in 5 cases each. The patients had varied symptomatology with oropharyngeal pain as most common. The average styloid process size was found to be 3.99 cm on right and 4.29 cm on left. All patients were operated by intraoral tonsilostyloidectomy method, and 0.8 to 2.5 cm bone size removed. Three of the patients were off all drugs within 3 week time, while 9 more patients were weaned off in 6 week time and 5 more in two months. The ratings on visual analogue scale for pain changed from 7.36 +/- 1.02 to 3 +/- 1.75. Complete resolution was achieved in 17/19 patients, (89.4%).
Conclusion: A high index of suspicion and an active look out for enlarged styloid processes in cases of chronic pharyngeal pain is highly recommended. Patients diagnosed with stylgia should be actively counselled for surgical intervention. The surgical approach, i.e. intraoral tonsilostyloidectomy, is a safe and highly effective procedure.

Keywords: Pharyngodynia, Stylgia, tonsilostyloidectomy

Introduction
Chronic throat pain (not discomfort) is rare by anecdotal experience. However, some of these cases could be traced to styloid complex issues directly or indirectly. In addition to pain (as a major symptom) these patients may report other vague symptoms. On intraoral palpation hard bony mass may be palpated deep to or behind or above the tonsils (one or both sides) with the patient instantly attributing whole cause of his/her pain to the palpated area. Most of these patients are referred for digital radiography or CT scans where the size and orientation of styloid processes is addressed. The patients in whom the styloid processes are significantly enlarged are advised surgery, and others are managed medically using various kinds of analgesics or neuromodulators. While this is the norm in clinical practice, our aim is to investigate the role of active surgical intervention in all such cases. The evidence to support such an argument is sparse in literature. However, an anecdotal experiment would provide a certain justification for such a study. As a thorough clinician is aware of chronic irritating nature of the course of pharyngodynia attributable to styloid processes, or a classical 'Eagles syndrome', any possible approach to lower the cost burden of a prolonged medical treatment is heartfelt. And an unsatisfied patient leaving the clinic with a further increased dose of medication is unassuming of a surgeons practice.

Normal length of styloid process in adults can vary between 20 and 25 millimetres [1]. Styloid processes longer than 30 mm are called elongated styloid processes (ESP) [2, 3]. The incidence of the ESP is controversial (ranges between 1.4% and 30%) in the literature [1, 4]. Sizes as long as 80.4mm are recorded and even these huge styloid processes could be asymptmatic [5]. Only small percentages (between 1% and 5%) of the patients have clinical symptoms where elongated styloid process (ESP) occasionally irritates or disrupts adjacent anatomical structures, which is called Eagle syndrome [6]. ES can occur unilaterally or bilaterally and frequently results in symptoms of dysphagia, recurrent throat pain and foreign object sensation, referred otalgia, headache, pain on rotation of the neck, dizziness, pain on extension of the tongue, pain on opening mouth, discomfort during chewing, change in voice, and a sensation of hypersalivation [7]. The differential diagnosis for ES includes cervical myofacial pain syndrome, migraine, trigeminal neuralgia, glossopharyngeal neuralgia, nervus intermedius neuralgia, nasopharyngeal mass/lesion, tonsillitis, otitis, degenerative diseases causing neck pain, psychosomatic diseases, vascular compromise (atherosclerosis), pain of dental origin, and TMJ problems [8, 9].

Most frequently, a panoramic radiograph is used to determine whether the SP is elongated [10]. However, 3-D CT scan gives a better assessment of size and orientation and is mostly not operator dependent. H. Ozlunc *et al*., [6] have suggested cone beam (CB) CT as an alternative. CBCT allows images to be acquired with a low dose of radiation, shorter patient examination time and lower costs than conventional CT, which make its routine use practicable for oral and maxillofacial imaging and surgical procedures. The length of SP is measured from the caudal margin on the tympanic plate to the tip of the process. The ossification of SHL that joined SP is added to the measurement.

Nonsurgical treatments include reassurance, non-steroidal anti-inflammatory medications, analgesics, anticonvulsants, antidepressants and local infiltrations with steroids or anesthetic agents [2]. Patients who fail medical therapy may benefit from surgical removal of the elongated portion of the SP. This is generally accepted approach to stylgia. Our approach in this study has been slightly different, that is, to offer surgical intervention as the primary treatment method.

Material and methods: This study is a retrospective-prospective analysis of patients of chronic throat pain attributable to the styloid complex. The source of our patients in the study are the ENT OPD visiting patients in MMABM Hospital, Anantnag, which is a secondary referral multispeciality hospital with a fully function ENT department. All such patients were chosen after a thorough clinical examination which failed to link such cases with any other pathology like oropharyngeal ulcers, chronic infections, oropharyngeal growth, or cervical spine pain. Palpation of tonsilar fossae in any stylgia patient evoked a typical response. In doubtful cases injection of 1-2 mL of 2% Xylocaine in the tonsilar fossae was done. Patients in whom this led to a, partial or complete, temporary cessation of discomfort were further evaluated. In earlier cases in the series digital skull x-rays were ordered as first investigation. Subsequently it was felt that a non-contrast skull base CT scan with 3-D analysis of styloid processes should be ordered in all such cases. All such patients had styloid processes of sizes in excess of 2.9 cm and were offered surgical treatment in the form of tonsilostyloidectomy. After proper counselling most of these patients consented for the procedure.

The technique of the surgical procedure briefly described as: Under general anaesthesia tonsillectomy is done by diathermy aided subcapsular dissection method. Styloid process is palpated in the tonsilar bed and incision is made by cautery directly over it followed by retraction of tissues to expose the tip of styloid process from which the attachments are freed and periosteal elevation is attempted. Using a styloid curette or a sharp bone cutter the bone is broken as high as possible. The fossa is sutured using 2-0 vicryl and hemostasis is ensured. Post-operatively patients were managed with intravenous antibiotics, and progressively decreasing analgesic doses.

Patients whom we were able to completely wean off from analgesics were considered treated. The patients were seen at 3, 6 and 12 weeks, and were assessed for pain. The pain score was compared with the pre-operative visual analogue score.

Results

Between April, 2013 to October, 2017, 60750 new patients attended our ENT OPD. In this period, 27 patients with chronic throat pain (pharyngodynia) traced to elongated styoid process (ESP) as the origin were picked by us. This gives an incidence of 0.44 per 1000 OPD patients. All of these patients were counselled for surgery, but only 19 patients gave their consent. We have done a detailed analysis of these 19 patients in the present report. The remaining 8 patients are still on our follow up. (Table 1)

Table 1

Operated	Non-operated	Total
19	8	27

The youngest patient in our study is 25 years old and oldest is 55 years. (Table 2)

Table 2

Age group	No. of patients
21-30	4
31-40	4
41-50	7
51-60	4

The mean age of our patients is 40.70 years. The male to female ratio in our study group was 1:2.1, with 13 patients in study group being females. (Table 3)

Table 3

Males	Females	Total (Operated)
6	13	19

The average duration of symptoms at the presentation to our OPD is 22 months (about 2 years). Bilateral pharyngeal pain (bilateral ES) was noted in 9 out of 19 patients, with the right and left sided predominant symptoms in 5 patients each.

The presentation symptoms were intraoral pain (in all patients) with occasional burning sensations (in 7 patients), temporal region pain (noted in 4 patients), pain in

submandibular and infra-auricular region (reported by 3 patients), dizziness (in 2 cases), neck pain, earache, and pressure sensation inside the mouth (as mentioned by 1 patient each). One of the patients reported pain aggravation on chewing and swallowing, and another one due to neck movements. One of the patients also reported a few fainting episodes. (Table 4)

Table 4

Symptoms:	No. of patients
Intraoral pain	19
Intraoral burning sensation	7
Temporal region pain	4
Submandibular region pain	3
Infra-auricular pain	3
Dizziness	2
Neck pain	1
Earache	1
Pressure sensations in mouth	1

Multiple ENT doctor visits were reported by 11 patients in the study group and 7 patients had visited psychiatrists. Two patients had undergone tooth extractions for the same symptoms with no relief. One of the patients in the study group had undergone a surgery for cervical rib.

The mean styloid process size of the 38 processes measured in the study group is 4.14 cm, with the right processes 3.99 cm and left processes 4.29 cm average size. The smallest process that was operated upon in this series is 2.9 cm and the largest one is 5.19 cm.

Bilateral tonsillectomy alongwith removal of styloid processes broken as high as possible in each case was done, with removed bone size ranging from 0.8 to 2.5 cm in length.

Three of the patients were off all drugs within 3 week time, while 9 more patients were weaned off in 6 week time and 5 more in two months. But two patients continue to take analgesics off and on. (Table 5)

Table 5

No. of patients	Time required to wean off drugs
3	3 weeks
9	6 weeks
5	9 weeks
2	NA

The ratings on visual analogue scale for pain changed from 7.36 +/- 1.02 to 3 +/- 1.75. One patient showed a marginal increase in pain postoperatively with no improvement. This patient had an anomalous cervical rib also which was operated previously and she was in obvious distress. On further queries she was not able to relate the origin of the pain clearly.

Out of the 8 non-operated patients, 4 patients are on Carbamazepine 200 mg TID, 3 patients are on Gabapentin 300 mg BID and one patient is on Gabapentin plus Nortriptiline 25 mg OD.

Statistics: Visual analogue pain score was documented pre and post operatively. Our hypothesis was that postoperatively the VAS rating changes significantly. The change in the score at 3 months was the end point for calculation of average scores with standard deviations. P value was calculated using t score calculator online. (Table 6a, 6b)

Table 6a

	Pre-operative VA Rating	Post-operative VA Rating	VA Rating at 3 months		Pre-operative VA Rating	Post-operative VA Rating	VA Rating at 3 months
1	8/10	2/10	-6	11	6/10	3/10	-3
2	7/10	2/10	-5	12	8/10	4/10	-4
3	7/10	1/10	-6	13	6/10	1/10	-5
4	8/10	9/10	+1	14	7/10	2/10	-5
5	8/10	3/10	-5	15	6/10	2/10	-4
6	9/10	5/10	-4	16	6/10	3/10	-3
7	7/10	1/10	-6	17	9/10	4/10	-5
8	8/10	3/10	-5	18	6/10	2l10	-4
9	8/10	4/10	-4	19	9/10	3l10	-6
10	7/10	3/10	-4	20			

Table 6b

Preoperative Statistical Mean with Standard deviation of VAR out of 10	Postoperative Statistical Mean with Standard deviation of VAR out of 10
7.36 +/- 1.02	3 +/- 1.75

P value < 0.00001.

Since the p value is < than 0.005, it is deemed significant.

Discussion

The population catered by our hospital is poorly defined in location, so it was not possible for us to calculate the incidence and prevalence ratios appropriately. However, in terms of the OPD visiting people the incidence of symptomatic Elongated Styloid Process is roughly 0.44 per 1000 ENT OPD patients in our set-up. According to W. W. Eagle, the incidence of Eagles syndrome varies among populations, but its main incidence is 4% in general population [1]. This correlates well with other studies noting the incidence of symptomatic elongated styloid process to be 4% [2]. But overall the incidence of ESP is 1.4% to 3%, and about 1% to 5% of these are said to be symptomatic [6]. With this in mind our study has covered a lot less number of such cases and as such a lot needs to be done in emphasizing the need of active lookout of this condition in our population.

According to some researchers there is no relation to gender in such cases [11]. Others have shown higher incidence in female population [12]. In our study we found that the

females are more than twice more likely to be affected by this condition.

The incidence is reported to be higher in age group 30-40 years according to some studies, while others report it to be higher in age more than 50 years [2 13]. In the present series we found it to be almost equally distributed in 3rd, 4th and 6th decade, with a marginal increase in the 5th. It seems that the calcification of stylohoid ligament with ageing which adds to the length of styloid process is an aiding factor for the symptoms to appear.

There is no side preference in this condition, and it is most likely to be bilateral. However, symptoms could be more on one side than another (asymmetrical pattern), which is a common clinical finding. In their studies, Thot *et al*., who studied on the anatomy of styloid process had found that the average styloid process length is 1.49 cm on the right and 1.52 cm on the left [14]. Although they studied the general population, in the symptomatic cases as in the present contest it seems to correlate (3.99 cm on right vs 4.29 on left).

Non-contrast CT scan with 3D reconstruction to find about the size and orientation of the styloid processes is a sufficient investigation for the suspected cases. Recently some researchers have proposed 3-D Cone beam CT as the more preferred investigation [6]. If there is any clinical evidence of carotid compression then CT or MR angiography may be ordered [15].

Although initially patients are managed medically, but surgical management is the more accepted approach. For the conservative management, use of steroids, local anaesthesia, and drugs like carbamazepine have all been tried [16]. The most satisfactory and effective treatment is surgical shortening of the styloid process through either an intraoral or external approach [17]. For the surgical excision, both intraoral and extraoral approaches have been tried. Extraoral approach offers the advantage of a good view and that the major vascular complication chances are inhibited. But cosmetic satisfaction is low with such an approach and it takes a longer OT time. There are no such issues in intraoral approach [18]. Although tonsilostyloidectomy seems to be a commoner technique, but other novel techniques have been tried [19]. We favour a straight forward intraoral approach for the styloid excision. It has been reported to be safe as the carotid space is mostly not breached, and also the operating time is sufficiently less [20]. We did tonsillectomy in all cases to aid in better exposure and avoid any future tonsilar space infections.

We found a significant improvement in the symptoms, i.e. oropharyngeal pain, of the patients after surgery. The self-scoring of pain by the patients offered us a means of pain assessment and documentation. On the visual analogue scale all the patients, except one, reported improvement. Zinnuroglu M. *et al* reported that the visual analogue scale scores decreased significantly after the surgical excision of elongated styloid processes (from 6.7 +/- 2.3 to 2.1 +/- 1.8), and all the complaints except for headache had diminished (P<.05) [21]. In the present study the rating improved from 7.36 +/- 1.02 to 3 +/- 1.75 (p<0.005)

We do not recommend the conservative way of managing the stylgia patients, i.e. with medications like analgesics or neuromodulator drugs. In our view such an approach is illogical as it does not address the causative factor of the condition. Medical management could be prescribed for patients till they are prepared for the surgery, or it could be offered to patients who refuse surgery altogether after all the efforts to counsel the patient are exhausted. We used our own experience with earlier patients as a model and evidence. Patients were made to talk to operated ones as a further motivation. The evidence in support of active surgical management in stylgia in literature is unquestionable. And the surgical excision of styloid process is safe and not fraught with any significant complications.

Conclusion

A high index of suspicion and an active look out for enlarged styloid processes in cases of chronic pharyngeal pain is highly recommended. Patients diagnosed with stylgia should be actively counselled for surgical intervention. This is done to prevent moribund use of analgesics and other medications which does not treat the disease but only prolong the associated miseries. Besides the surgical approach, i.e. intraoral tonsilostyloidectomy is a safe and highly effective procedure.

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