STYLALGIA - GLOSSOPHARYNGEAL NEURALGIA: OUTCOME OF MANAGEMENT

Shedje Swapna A ^{*1}, Havle Abhay D ², Roy Priya P ³, Shedje Ajay J ⁴.

^{*1} Assistant Professor Department of Anatomy, KIMS University, Karad, Maharashtra, India.

² Professor & Head, Department of Otorhinolaryngology, KIMS University, Karad, Maharashtra, India.

³ Associate Professor Department of Anatomy, KIMS University, Karad, Maharashtra, India.

⁴ Amaltas Institute of Medical Sciences, Dewas, MP India.

ABSTRACT

Introduction: Elongated styloid may pose either Eagle's or Stylocarotid syndrome leading to oro-facial pain. However, in patients of oro-facial pain, the investigation about the elongation of styloid process (SP) is not made routinely. Consequently, SP elongation is usually overlooked as possible cause of symptom referral.

Materials and Methods: In our study the various presenting symptoms were throat pain, ipsilateral otalgia and foreign body sensation in throat. Digital palpation of styloid process and accentuation of pain in tonsillar fossa, positive lignocaine test along with radiological demonstration of the elongation of styloid process confirm the diagnosis. Total of 30 patients were treated surgically by transoral Styloidectomy for oro-facial pain. All resected tonsils were subjected to histopathological examination. This is a prospective study conducted at the Krishna Hospital, Karad, Maharashtra, India.

Results: Twenty eight (93%) of 30 cases of oro-facial pain became asymptomatic after surgery. 1 had considerable improvement to the extent that the subject did not require any analgesics subsequently. 21 (70 %) out of 30 resected tonsils revealed fibrous variety. There were no serious surgical complications.

Conclusion: Tonsillo-styloidectomy is the treatment of choice for Eagle's syndrome with a high success rate. Usefulness of medical (non surgical) treatment remains doubtful.

KEY WORDS: Eagle's syndrome, Stylocarotid syndrome, Oro-facial pain, Digital Palpation, Female, Transoral Styloidectomy.

Address for Correspondence: Dr. Shedje Swapna A, Assistant Professor, Department of Anatomy, KIMS University, Karad, Maharashtra, 415110, India. Mobile- (91) 9850816966

E-Mail: swapna.shedje@gmail.com

Access this Article online

Quick Response code

DOI: 10.16965/ijar.2017.227

Web site: International Journal of Anatomy and Research
ISSN 2321-4287
www.ijmhr.org/ijar.htm

Received: 21 Apr 2017
Peer Review: 22 Apr 2017
Revised: None

Accepted: 23 May 2017
Published (O): 31 Jun 2017
Published (P): 30 Jun 2017

INTRODUCTION

Embryologically origin of styloid process is from Reichert cartilage of the second branchial arch. It arises from the inferior surface of the temporal bone at the junction of its petrous and tympanic bones. Styloid process with the stylohyoid

ligament and the lesser cornua of the hyoid bone, forms the stylohyoid apparatus. The word Styloideidos is derived from Greek- stylos i.e. 'pillar' and eidos i.e. 'like' meaning pillar-like process.

An abnormally elongated styloid process or

ossification of stylohyoid ligament may produce cluster of symptoms in few giving rise to "Eagle's syndrome" also known as Stylohyoid syndrome, which is characterized by craniofacial or cervical pain [1-3].

In 1937 Eagle primarily described two syndromes:

Classic Styloid syndrome: It occurs mainly after tonsillectomy and is characterised by isolated pharyngalgia in the tonsillar fossa. Its symptoms include dysphagia odynophagia, a sensation of increased salivation, and a sensation of foreign in the pharynx.

Stylocarotid syndrome: The "second form" of the syndrome ("stylocarotid syndrome") is characterized by the compression of the internal or external carotid artery (with their peri-vascular sympathetic fibers) by a laterally or medially deviated SP.¹⁰ It is related to a pain along the distribution of the artery, which is provoked and exacerbated by rotation and compression of the neck. It's not correlated with tonsillectomy. In case of impingement of the internal carotid artery, patients often refer supraorbital pain and parietal headache. In case of external carotid artery irritation, the pain radiates to the infraorbital region.(57th)

Eagle defined the length of a normal styloid process less than 25mm [1-3].

The styloid process may vary from 5 to 50 mm in length and the stylohyoid ligament may ossify from its origin at the styloid process to its attachment at the hyoid bone.

Elongated SP is seen in about 4% of general population, while only 4% of these patients remain symptomatic; the true incidence is 0.16% with a female predominance of 3:1 [4,5].

Eagle's syndrome can be treated conservatively or surgically or both.

Conservative treatment: Transpharyngeal injection of steroids and lidocaine, nonsteroidal anti-inflammatory drugs, antidepressant diazepam transpharyngeal manipulation with manual fracturing of the SP [6]. However failing this attempt, surgery remains a viable alternative.

Surgical Treatment: The most satisfactory and effective treatment is surgical shortening of the

SP through either an intraoral approach via tonsillar fossa or external approach from neck [8-12]. The intraoral approach via tonsillar fossa is advocated by Eagle (1949) [13,7].

The transoral approach involving resection of the SP is relatively easy to perform and leaves no external scar. Both the operation and recovery times of this procedure are short. However, the rare disadvantages of the transoral approach are possible deep cervical infection, poor visualization of the surgical field and possible temporary edema at the operative site [10].

Aims & objectives: To determine the incidence of elongated styloid process among symptomatic patients visiting ENT out patient.

To assess the effectiveness of management of Symptomatic elongated styloid process (Eagle's syndrome).

MATERIALS AND METHODS

A prospective time bound study of probable etiology in cases of stylalgia was carried out from December 2014 to December 2016 in a tertiary care hospital. Total of 30 cases were studied based on the clinical presentation, radiological findings and histopathology of all resected tonsils.

Inclusion criteria: All cases irrespective of their age and gender presenting with typical symptom of throat pain, accompanied unilateral ear ache, cervico-facial pain on swallowing and or head turning movements, and not responding to initial conservative medical treatment were included in the study. Also the symptomatic cases with clinically palpable styloid, positive lignocaine test and elongated (>25mm) styloid process on radiological imaging were included.

Exclusion criteria: All cases presenting with typical symptom and having history of tonsillectomy, pregnancy, on breast feeding and diabetes irrespective of its control were excluded in this study.

In all cases subjective symptom of pain was assessed by using visual analog scale (VAS). Severity of pain experienced by an individual was recorded in the form of numbers ranging from 0-10 where 0=no pain and 10=worst imaginable pain. Depending on the range of analog scores

the results were further grouped into three as mild=1-3, moderate=4-6 and severe=7-10. All patients were treated with tonsillectomy and transoral styloidectomy. In all cases the resected tonsillar tissues were subjected to histopathological examination. Pain was assessed by Questionnaire method using Visual Analogue scale. Each patient was asked to describe the level of pain experienced by them in the form of numbers ranging from 1-10 or by using Visual analogue scale.

Fig. 1: Visual analogue scale.

Statistical analysis (SPSS version 18): Unpaired t test to measure the Quantitative data like length of Styloid process, χ^2 test to measure the Qualitative data like sex, ANOVA to analysis the variations within the groups.

Table 1: Distribution of study group.

Distribution of patient according to age	
Group	Age(yrs)
I	≤ 30
II	31-40
III	41-50
IV	≥ 51

Fig. 2: Digital palpation of SP.

RESULTS

Incidence: A total of 45,570 patients visited ENT opd during the study period, among them 24,390

patients were male and 21,182 patients were females during the study period. A total of 30 cases of ES were studied. The overall incidence of ES in patients visiting ENT opd was found to be 0.65%

Sex distribution and ratio: Study group consisted of 9(30%) males and 21(70%) females with M: F ratio being 1:2.33.

Age distribution: Majority of patients (5 males, 14 females) belonged to age group 31-40 yrs(63%). Youngest patient was 27 yrs old while oldest was 60 yrs.

Mean length of the left and right SP in two sexes: Mean length of the left and right SP in males was 32.4 ± 5.5 and 32.2 ± 7.1 mm while in females it was 32.5 ± 8.0 mm and 31.6 ± 7.0 mm. No significant difference was found in mean length of SP among male and female on both sides.

Fig. 3: Plain radiograph for Styloid process.

Fig. 4: 3D CT showing B/L ESP.(R-45mm,L-36mm).

Table 2: Mean length of SP in different age groups.

Age group (years)	N (%)	Average length of SP (mm)	
		Left side	Right side
30	3(10)	26 ± 2.0	24.6 ± 1.1
31-40	19(63.3)	30.26 ± 4.49	30.4 ± 3.8
41-50	6(20)	35 ± 8.27	38 ± 8.3
50	2(6.6)	46 ± 8.48	48 ± 2.8
ANOVA	$F=12.83, p<0.000$		$F=6.73, p<0.002$

Highly significant difference in average length of SP among different age groups on both sides. Among 30 patients 15(50%) had U/L ESP(L=7,R=8) and 15(50%) had B/L ESP.

Table 3: Major complaints.

Symptoms	Present(%)
Isilateral throat pain	25(83)
Ipsilateral otalgia	22(73)
F B sensation	18(60)
Cervical pain	17(56)
Facial pain	11(36)
Headache	08(26)

50% of patients presented with moderate pain and 33% patients were having mild pain while remaining 16% had severe pain according to VAS. Frequency distribution of number of patients with pain according to VAS on first visit among both the sexes shows more number of females suffering from moderate to severe pain.

Assessment of pain on 1st follow-up 3 patients suffering from moderate pain on 1st visit were found to have mild pain after 3 weeks of medical treatment, where as there was no change in pain category for other patients after medical treatment.

Table 4: Assessment of pain on all visit.

Pain VAS	1 st visit	After med T/t	After surg (2nd f/ up)	After surg (3rd f/ up)
No pain	0	0	20	28
Mild (0-3)	10	13	6	2
Moderate(4- 6)	15	12	4	0
Severe (7-10)	5	5	0	0
X2 test	X2=0.7246, p=0.69			
	X2=53.06, p<0.0001			

Fig. 6: Transoral Styloidectomy

Fig. 7: Resected Styloid process.

Length of SP removed: A total of 35 SP were removed, 13 removed SP had length less than 10mm, 17 SP between 10-20 mm and rest 05 had length greater than 20 mm.

DISCUSSION

Eagle's Syndrome or Stylalgia caused by elongated SP is an uncommon and under diagnosed clinical entity. It is a rare disease. 0.04-0.08% of population is suffering from it, whereas 1.5 – 3.0 % of adults have some of the complaints due to the pathology of this apparatus [14]. According to Wong ML and Chase et.al. 4% of general population an elongated SP occurs, while only about 4% of these patients are symptomatic; thus the true incidence is 0.16% with a female predominance of 3:1 [15,16]. Studies performed by anatomists Keur et al., Milner et al (1996) [17], have proven that 2% to 4% of the overall population have either an elongated or a calcified SP, but only 0.04% of them are symptomatic. Baddour HM 2004 [18] reported 0.04-0.08 % incidence.

Table 4: Comparison of sex ratio in Eagle syndrome.

Study	Male	Female	Ratio(M:F)
Sikanjic and Vlak et al.(2008) [19]	38	64	01:01.7
Alper Ceylan et al. (2008) [23]	19	42	01:02.2
Y. K. Maru et al. (2003) [20]	140	192	01:01.4
Albinas Gervickas et al.(2002) [24]	32	65	01:02.0
Samar Yadav et al. (2001) [25]	5	35	1:07
Present study	9	21	01:02.3

Our study goes in favour with study done by Sikanjic and Vlak et al.(2008) [19], during their 2008 study on 448 X-rays, 102 of which presented with Eagle's syndrome. Of the 102 patients with Eagle's syndrome, 66(64%) were females. Y. K. Maru and Kusum Patidar(2003) [20] in their clinical experience of 332 cases of ES included 140(42%) males and 192(57%) females.

As to the incidence in the overall population, Grossman & Tarsitano(1977) [21] reported that the syndrome usually affects adults, with no difference between sexes. However, Neville et al. [22]

observed that the syndrome affects mostly women in their 20s and 30s.

The present study goes in favour of study by Ceylan Alper et al.(2008) [23] in which pain on swallowing was the most common complaint (85%). Otolgia(77%), foreign body sensation in throat (60%), cervical pain (57%), facial pain (36%).

In the present study the average length of right SP was 31.8 ± 6.9 mm while average length of left SP was 32 ± 7.3 mm respectively. There was no statistical difference between the lengths of the two sides.

In the present study 15 (50%) of subjects were having B/L ESP which comprised of 5 (33%) male and 10(66%) females. Among remaining 15(50%) patients 8 (53.4%) [1 male (12.5%) and 7 female (87.5 %)] of them were having ESP on Rt. side and 7(46 %) [3 male 42.8%) and 4 female (57.1 %)] were having ESP on Lt. side. Frequency laterality of ESP according to sex and side was not found statistically significant.

In another study conducted by Sarika S Naik et al.(2011) [26] on 15 cases of ES found bilaterality in 12(80%) case and unilaterality in 3(20%) cases. The study by Alper Ceylan et al.(2008) [23] on 61 case of ES 23 (37.7%) had bilateral symptoms, 38 patients (62.3%) had unilateral symptoms.

In the present study only 3(10%) patients out of 30(100%) were benefitted to some extent by medical treatment but was not acceptable to the patients. In a similar study by Sanjeev Mohanty et al.(2009) [27] on 28 patients of ES, 17 were females and 39 were male. All these patients underwent a trial of the drug carbamazepine, taken orally at a dose of 200 mg TDS for 3 weeks, but without any relief in symptoms.

All patients underwent tonsillo-styloidectomy by trans-oral approach. A total of 35 SP were removed. In 10 patients B/L tonsillo-styloidectomy was done, while in case U/L cases the symptomatic ESP was removed. There was no intra-operative or postoperative complication.

CONCLUSION

The present study has laid to the conclusion that: ES is a rare, definitive, often misdiagnosed clinical entity with elongated SP as the underlying etiology. The analysis of clinical symptoms allows not only to diagnose the syndrome, but also to foresee a possible disturbance of the SP or the SHL. But the basics in pathogenesis are not yet clearly understood, so also are the factors that encourage calcification at SHL. Radiological analysis is the basic method for the localization of the stylohyoid complex disturbance and the diagnostics of its nature. Medical treatment did not seem effective, and we feel that every patient with an enlarged, misdirected and fractured symptomatic SP must be offered surgical treatment preferably in the form of intraoral complete excision of SP. An excision of SP by the intraoral route under general anesthesia is easy and most appropriate answer to the nagging, disturbing pain syndromes in the head and neck which appears to be due to enlarged SP. Fibrous tonsillitis could be considered as one of the other etiological factors in Eagle Syndrome.

ACKNOWLEDGEMENTS

Authors conveying their heartfelt thanks to Dr Avinash , ENT surgeon and Dr M A Doshi , HOD Anatomy KIMS Karad.

Conflicts of Interests: None

REFERENCES

- [1]. Eagle WW. Elongated Styloid process. Arch Otolaryngol 1937;25:584-587.
- [2]. Eagle WW. Elongated styloid process; symptoms and treatment. AMA Arch Otolaryngol 1958. Feb;67(2):172-176. [PubMed]
- [3]. Eagle WW. The symptoms, diagnosis and treatment of the elongated styloid process. Am Surg 1962;28:1-5.
- [4]. Mendelsohn AH, Berke GS, Chhetri DK. Heterogeneity in the clinical presentation of Eagle's syndrome. Otolaryngol Head Neck Surgery. 2006; 134:389-393.
- [5]. Linden R WF. Indications for esophageal reconstruction of corrosive Structures. Acta Oto-laryngol .1966;62:27-32.
- [6]. Glogoff MR, Baum SM, Cheifetz I. Diagnosis and treatment of Eagle's syndrome. J Oral Surg 1981;39:941-4.
- [7]. Eagle WW. Elongated SP. Further observations and a new syndrome. Archives of Otolaryngology. 1948;47(5):630-640.

- [8]. Ernest EA. The Ernest Syndrome: An Insertion Tendinosis of the Stylomandibular Ligament. Journal of Neurology and Orthopedic Medicin and Surgery. 1986;7(4):427-438.
- [9]. Wong ML, Rossi MD, Groff W, Castro S, Powell J. Physical therapy management of a patient with Eagle syndrome. Physiotherapy Theory Pract 2011;27:319-27.
- [10]. Chase DC, Zarmen A, Bigelow WC, McCoy JM. Eagle's syndrome: a comparison of intraoral versus extraoral surgical approaches. Oral Surg Oral Med Oral Pathol 1986;62:625-629.
- [11]. Mendelsohn AH, Gerald S, Dinesh KC. Heterogeneity in the Clinical Presentation of Eagle's Syndrome. Am Otolaryngology - Head and Neck Surgery. 2006;134:389-393.
- [12]. Camarda AJ, Deschamps C, Forest DI. Stylohyoid chain ossification: A discussion of etiology. Oral Surg Oral Med Oral Pathol. 1989;67:508-14.
- [13]. Kaufman SM, Elzay RP, Irish EF. SP variation. Radiologic and clinical study. Arch Otolaryngol. May 1970;91(5):460-3.
- [14]. Restrepo, Palacios SE, Rojas R. Eagle's Syndrome. Ear, Nose & Throat Journal 2000;81(10):700-701.
- [15]. Mendelsohn AH, Berke GS, Chhetri DK. Heterogeneity in the clinical presentation of Eagle's syndrome. Otolaryngol Head Neck Surgery. 2006;134:389-393.
- [16]. Linden R WF. Indications for esophageal reconstruction of corrosive Structures. Acta Otolaryngol. 1966;62:27-32.
- [17]. Keur JJ, Milner, Campbell JP et al. The clinical significance of the elongated SP. Oral Surg Oral Med Oral Pathol. 1986;61:399-404.
- [18]. Baddour HM, Mc Anear JT, Tilson AB. Eagle's syndrome Report of case. J Oral Surg. 1978;36(6):486.
- [19]. Sikanjic, P.R., and Vlak, D. Elongated SP in Late Medieval Skeletons from Uzdolje- Grablje, Croatia. International Journal of Osteoarchaeology. 2008;20(2):248-252.
- [20]. Maru YK, Patidar Kusum. Stylalgia and its surgical management by intra oral route- clinical experience of 332 cases. Indian Journal of Otolaryngology and Head and Neck Surgery. April - June 2003;55:2.
- [21]. Grossman JR & Tarsitano JJ. The styloid-stylohyoid syndrome. J. Oral Surg. 1977;35(7):555-60.
- [22]. Neville B, Dam DD, Allen CM, Bouquot J. Patologia oral maxillofacial. Rio de Janeiro Elsevier. 2009;3: 972.
- [23]. Ceylan Alper, Sabri Uslu. Surgical Treatment of Elongated SP: Experience of 61 Cases Skull Base. 2008 July;18(5):289-295.
- [24]. Albinas Gervickas, Ricardas Kubilius, Gintautas Sabalys. Clinic, Diagnostics and Treatment Peculiarities of Eagle's syndrome, Stomatologija, Baltic Dental and Maxillofacial Journal. 2004;6:11-13.
- [25]. Yadav Samar P.S. Stylalgia: An Indian Perspective The Journal of Otolaryngology. October 2001;30(5):304-305.
- [26]. Naik SM, Naik SS. Tonsillo-Styloidectomy for eagle's syndrome: a review of 15 cases in KVG medical college Sullia. Oman Med J. 2011 March;26(2):122-126.
- [27]. Mohanty S. Significance of styloidectomy in Eagle's syndrome: an analysis. Indian J Otolaryngol Head Neck Surg. October-December 2009;61:262-265.

How to cite this article:

Shedje Swapna A, Havle Abhay D, Roy Priya P, Shedje Ajay J. STYLALGIA - GLOSSOPHARYNGEAL NEURALGIA: OUTCOME OF MANAGEMENT. Int J Anat Res 2017;5(2.3):3952-3957. DOI: 10.16965/ijar.2017.227