

A case series on Eagle syndrome. A Study.

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Abstract: Eagle syndrome is named after Watt W. Eagle an Otolaryngologist at Duke University, Durham, North Carolina who described the earliest cases in 1937¹ and published a series on elongated styloid process in 1957. It is also termed as a stylohyoid syndrome, styloid syndrome, or styloid-carotid artery syndrome². With the stylohyoid ligament and the lesser horn of the hyoid bone, the styloid process forms the stylohyoid apparatus. Eagle syndrome is characterized by recurrent pain in the oropharynx with radiation towards ear due to an elongated or disfigured styloid process or calcified stylohyoid ligament³.

Eagle defined the length of a normal styloid process at 2.5-3.0 cm⁴. The normal length of the styloid process⁵ and its morphology varies greatly⁶. About 4% of population is considered to have an elongated styloid process⁴. This study was undertaken because most of the people presenting with stylalgia and Eagle's syndrome in the local population did not have significantly enlarged styloid processes.

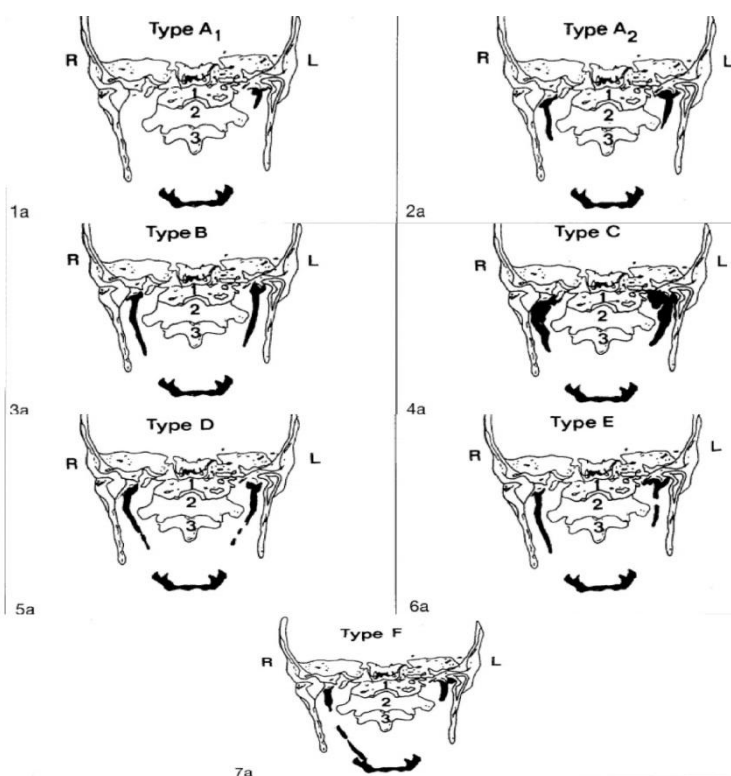
Aims:

1. To study the radiological morphology of styloid process and calculate the normal length of styloid process in local population
2. To compare the length of styloid process in people presenting with Eagle's syndrome: Radiologically and Intra-operatively.
3. To compare the length of Styloid Process in symptomatic vs asymptomatic people
4. To assess the efficacy of surgery in different morphological types and sizes of styloid process causing Eagle's syndrome.

Key words: Eagle, styloid, stylalgia, glossopharyngeal, neuralgia.

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Morphological types of styloid process⁶.

Type A1 (short) <25 mm,

Type A2 (long) 25—40 mm,

Type B: Elongated SP > 40 mm,

Type C: Bent SP,

Type D: Segmented SP+ ossified

Stylohyoid Ligament,

Type E: Pseudoarticulated,

Type F: SP with stylohyoid ligament

distally ossified and fixed to the

lesser horn of the hyoid bone

[SP= styloid process]

Patients and Methods:**Type of study:** Retrospective study

Source of data: All patients who underwent styloidectomy in Govt. E.N.T. Hospital, Andhra Medical College, Visakhapatnam with clinical diagnosis of Eagle's syndrome between July 2015 and June 2018 have been evaluated and 15 patients have been included in this study. For the purpose of calculation of normal length of styloid process, radiological lengths of styloid process of 100 people within age group of 20 to 80 years has been taken.

Inclusion Criteria-

1. Patients with pain in oral cavity not subsiding with medication with radiation to ipsilateral ear with increase in pain on palpating in tonsillar fossa.
2. Patients previously operated for tonsillectomy with a sensation of a raw throat, which the patient thinks did not heal properly after operation; a pain referred to the ear, especially noted on swallowing; painful and difficult swallowing; sensation of a foreign body in the pharynx^{3,4,5}, such as a toothbrush bristle or a fish-bone, and a persistent dull-aching type of sore throat which does not subside on analgesics and antibiotics.

Exclusion Criteria-

1. Patients who have other coexisting conditions like Eustachian tube obstruction/inflammation, Allergic rhinosinusitis and pharyngitis.
2. Patients having neuralgias due to other reasons like diabetes or cervical spondylosis etc.
3. Patients not fit for surgery or refusing surgery.

Method of collection of data:

A complete ENT examination and appropriate investigations were done.

1. Oral cavity, oropharynx, nose, nasopharynx and Ear examination.
2. DNE, Indirect and videolaryngoscopy.
3. Radiological measure were by [3DCT] of styloid process using maximum intensity projections and ADW 44 software and GE imaging systems hardware with lengths measured from base of styloid to tip of styloid process and when calcified stylohyoid ligaments were present, a note was made of morphology and length of SP with and without distal most calcification of stylohyoid ligament.
4. Surgery: 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.. Measurement of styloid process length intra-operatively was done by a silk suture thread held next to the process and thread measured by callipers ex vivo.

I. Observations And Results

1. Age and length:

Age in years	Avg. length of Styloid process [mm]
21-30	24.7
31-40	26.4
41-50	28.9
51-60	30.6
61-70	32.3
71-80	35.3

There was a progressive increase of styloid process with age.

The average length of Styloid process by 3DCT imaging in local population is 2.83 cm [+/- 0.09cm]

2. Variation of length of styloid process with sex:

Male	Female
29.2 mm	27.5 mm

Difference between average length of male and female styloid process is 1.7 mm

3 .Morphological types

The most common type of Styloid process in local population is A2 [2.5cm to 4cm] > Type E> Type D = C = A1

Type	Male %	Female%	Total%
A1	2.5	10	12.5
A2	16.25	15	31.25
B	3.75	0	3.75
C	5	7.5	12.5
D	5	7.5	12.5
E	11.25	7.5	18.75
F	0	0	0
TOTAL	50	50	100

4. Complaints and duration:

Throat pain, ear ache and foreign body sensation were the most common presenting symptoms. Otagia was bilateral in 9 patients [60%] with 3 patients [20%] complaining of more pain on one side. Two patients who had unilateral styloidectomy came back within 5 years with pain on the opposite unoperated side with an elongated styloid process. The average duration of symptoms before diagnosis was 6 months. 12 patients [80%] had previous tonsillectomy without resolution of symptoms. 4 patients [26.6%] were on analgesics for long term before surgery.

Complaint	Present in
Throat pain	15 [100%]
Referred otalgia	15 [100%]
Foreign body sensation	15 [100%]
Head ache	12 [80%]
Dysphagia	11 [73.3%]
Globus pharyngeus	8 [53.3%]
Dizziness	3 [20%]
Previous tonsillectomy	12 [80%]

5. Length & Morphology of styloid process in patients with Eagle’s syndrome:

Type	Male	Female	Total
A1	0	0	0
A2	0	0	0
B	1 [6.6%]	2 [13.3%]	3 [20%]
C	2 [13.3%]	4 [26.6%]	6 [40%]
D	1 [6.6%]	1 [6.6%]	2 [13.3%]
E	1 [6.6%]	3 [20%]	4 [26.6%]
F	0	0	0
TOTAL	5 [33.3%]	10 [66.6%]	15 [100%]

The most common type of Styloid process in People with Eagle’s syndrome in the local population is Type C [40%] [Curved/ Bent SP] > Type E [26%] [Pseudoarticulated] > Type B [20%] [Elongated SP>4cm] > Type D [13.3%] [segmented SP].

6. Patients with Eagle syndrome

We studied 15 patients with eagle syndrome. Most common age group is 41-50 years. Female : male = 2:1. The youngest patient was 38 years old and oldest patient was 60 years old. The mean age of presentation is 49.

Age	Male	Female	Total
21-30	0	1 [6.6%]	1 [6.6%]
31-40	2 [13.3%]	3 [20%]	5 [33.3%]
41-50	2 [13.3%]	4 [26.6%]	6 [40%]
51-60	1 [6.6%]	2 [13.3%]	3 [20%]
Total	5 [33.3%]	10 [66.6%]	15 [100%]

7. Intra - op findings and post - op findings:

Intra operatively the styloid process was palpated through the superior constrictor muscle but the stylohyoid ligament was not palpated in all situations. Curved styloid processes and pseudoarticulated curved styloid processes were seen impinging on glossopharyngeal nerve in 6 situations. Ossified stylohyoid ligaments which were seen on CT scans were not always palpable intra operatively and as such were not removed. All patients reported that the presenting complaint of pain subsided after surgery.

II. Discussion

1. Age and length:

There is a certain degree of inter observer variability when it comes to calculation of length of styloid process depending on the radiographer. In situations of calcification of stylohyoid ligament, it is observed that the morphology varies and Type D and Type E morphologies are considered as elongated styloid process but not as elongated styloid with stylohyoid calcification by some. The calcified stylohyoid complex can be causing pain in the tonsillar fossa or the styloid carotid artery syndrome. Ectopic calcification (EC) may have a role for the elongation of the styloid process in people with abnormal calcium (Ca), phosphorus (P), and vitamin D metabolism⁷.

The average length of Styloid process by 3DCT imaging in local population is 2.83 cm [+/- 0.09cm]. There was a progressive increase of styloid process with age with older individuals having longer styloid processes and calcified stylohyoid complex⁸. This probably explains the increased incidence at older age. Among the 15 patients with eagle syndrome that were evaluated, most common age group is 41-50 years.

2. Variation of length of styloid process with sex:

Difference between average length of male and female styloid process is 1.7 mm. There was no significant difference between average length of male and female styloid process in similar age groups but however in the cases studied, women were symptomatic at lengths at which men were not. There was a female : male ratio of 2:1. This may be explained by idea that ossification of the stylohyoid ligament might be related to endocrine disorders in women at menopause, accompanied by the ossification of ligaments elsewhere (eg, iliolumbar, thyrohyoid)⁹.

3. Morphological variations in local population and Eagle's syndrome:

The most common type of Styloid process in local population is A2 [2.5cm to 4cm] > Type E[Pseudoarticulated] > Type D [segmented SP] = C [curved / bent SP] = A1[<2.5cm].

The most common type of Styloid process in People with Eagle's syndrome in the local population is Type C [Curved/ Bent SP] > Type E [Pseudoarticulated] > Type B [Elongated SP>4cm] > Type D [segmented SP].

The abnormal length associated with abnormal angulation of the styloid process^{10,11} also called Type C SP was the most common type causing Eagle syndrome in the local population. This may be the reason why people presented with symptoms in the absence of a radiologically elongated styloid process.

The fracture and medialization of the ossified stylohyoid ligament, with incomplete repair due to continuous hyoid bone movements and formation of excessive granulation tissue¹², the ossification of muscular tendons leading to irritation of the structures nearby^{13,14} and the ossification of the stylohyoid ligament complex, causing contraction of the stylopharyngeal muscle and stretching of the XII cranial nerve⁶ may explain the symptoms in Type E, B and D styloid process in the local population.

8. Complaints and duration:

Presenting symptoms may be due to compression of the adjacent nerves^{15,16}. Pain may be due to proliferation of granulation tissue after a traumatic fracture of the styloid process causing tendinosis or impingement on the carotid vessels^{15,16}. Pain maybe due to impingement of the pharyngeal mucosa against an elongated process or impingement of the carotid vessels and their associated sympathetic chain¹⁷. Pain from an elongated styloid process is due to "constant mechanoreceptor discharge in the area of the 5th, 7th, 9th and 10th cranial nerve endings" initiated by a mechanical irritation from the styloid process¹⁷.

III. Conclusion

1. Eagle's syndrome associated with elongated styloid process is a rare clinical entity. Clinical palpation of tonsillar fossa may be complemented with a plain radiography of skull base Towne'view and 3DCT scan to make a diagnosis.

2. The length of the styloid process doesn't always correlate with the clinical signs and symptoms.

3. Understanding morphological and radiological variations of Styloid process is important in making a diagnosis.

4. Type C [Bent/Curved SP] is the most common cause of Eagle's syndrome in the local population.

5. Tonsillo-styloidectomy is the treatment of choice in all cases of Eagle's syndrome and has definitely proven to relieve the patients of styalgia.

References

- [1]. Eagle.W.W.: Elongated styloid process. Arch. Otolaryng. 25:584. May 1937.
- [2]. Bokhari MR, Bhimji SS. Eagle Syndrome. 2017 Jun.
- [3]. <https://emedicine.medscape.com/article/1447247-overview>
- [4]. Eagle W. Elongated styloid process: Further observation and a new syndrome. Arch Otolaryngol. 1948. 47:630-640.
- [5]. Eagle W. Elongated styloid process. Report of two cases. Arch Otolaryngol. 1937. 25:584-587.
- [6]. Monsour PA, Young WG. Variability of the styloid process and stylohyoid ligament in panoramic radiographs. Oral Surg Oral Med Oral Pathol. 1986 May. 61(5):522-6. [Medline].
- [7]. Gokce C, Sisman Y, Sipahioğlu M. Styloid Process Elongation or Eagle's Syndrome: Is There Any Role for Ectopic Calcification?. Eur J Dent. 2008 Jul. 2 (3):224-8.
- [8]. Bruno G, De Stefani A, Balasso P, Mazzoleni S, Gracco A. Elongated styloid process: an epidemiological study on digital panoramic radiographs. J Clin Exp Dent. 2017 Dec. 9 (12):e1446-52.
- [9]. Epifanio G. Processi stiloidei lunghi e ossificazione della catena stiloidea. Rad Prat. 1962. 12:127-132
- [10]. Baddour HM, McAnear JT, Tilson HB. Eagle's syndrome. Report of a case. Oral Surg Oral Med Oral Pathol. 1978 Oct. 46(4):486-94. [Medline].
- [11]. Frommer J. Anatomic variations in the stylohyoid chain and their possible clinical significance. Oral Surg Oral Med Oral Pathol. 1974 Nov. 38(5):659-67. [Medline].
- [12]. Moffat DA, Ramsden RT, Shaw HJ. The styloid process syndrome: aetiological factors and surgical management. J Laryngol Otol. 1977 Apr. 91(4):279-294.
- [13]. Kaufman SM, Elzay RP, Irish EF. Styloid process variation. Radiologic and clinical study. Arch Otolaryngol. 1970 May. 91(5):460-3. [Medline].
- [14]. Lindeman P. The elongated styloid process as a cause of throat discomfort. Four case reports. J Laryngol Otol. 1985 May. 99(5):505-8
- [15]. CT findings associated with Eagle syndrome. Murtagh RD, Caracciolo JT, Fernandez G. AJNR Am J Neuroradiol. 2001 Aug; 22(7):1401-2
- [16]. Eagle's syndrome (elongated styloid process). Balbuena L Jr, Hayes D, Ramirez SG, Johnson R. South Med J. 1997 Mar; 90(3):331-4
- [17]. Baddour HM, Anear JT, Tilson AB. Eagles Syndrome, Case report. J Oral Surg 1978;36(6):486 .10.1016/0030-4220(78)90378-X

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