



Evaluation Of Postoperative Symptom Relief With Styloidectomy Procedure For Eagle Syndrome: A Case Series From A National-Accredited Center

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University of Health Sciences Turkey, Istanbul Haseki Training and Research Hospital, Clinic of Otorhinolaryngology, Istanbul, Turkey

Abstract

Aim: Eagle syndrome is a very rare disease that manifests with a styloid process longer than normal. The aim of this study is to evaluate the characteristic features of Eagle syndrome and postoperative regression of the existing complaints.

Methods: This study is a retrospective case series study. Data of 17 patients who had styloidectomy operation due to Eagle syndrome between 2015 and 2019 in a national accredited center are presented. Radiological findings, physical examination results and medical history were evaluated retrospectively according to the medical records of the hospital. Preoperative and postoperative visual analogue scale (VAS) and verbal response scale (VRS) scoring systems were used for pain scoring.

Results: The most common symptoms were throat pain on the same side (88.2%), neck pain (82.3%), and otalgia (70.5%). Less common symptoms were jaw pain (41.1%), dysphagia (35.2%), facial pain (29.4%), headache (23.5%), and foreign body sensation (23.5%). VAS pain scoring showed that 64.7% of the patients' pain completely regressed. The results of VRS were also similar to the results of postoperative VAS and they had a significant correlation in a negative way ($p < 0.001$).

Conclusion: The procedure of styloidectomy is more successful for the regression of neck, throat, jaw, and ear pain than for the headache and facial pain for patients who had Eagle syndrome.

Keywords: Eagle syndrome, partial styloidectomy, transoral, styloid process, stylohyoid ligament

Introduction

Dr. Watt Eagle first described Eagle syndrome (ES) in 1937 as a disease which can be characterized by sore throat, dysphonia, dysphagia, otalgia and foreign body sensation (1). ES is a rare disease that occurs by elongation of the styloid process (SP) (>25 mm) with or without calcification of the stylohyoid ligament (2). This anatomical elongation causes inflammation due to traumatic irritation of the glossopharyngeal nerve (3). The prevalence of the elongated SP is approximately 4-8 cases per 10,000 people in the general population, but only 4-10% of the population has symptoms (4).

The diagnosis of ES is based on anamnesis, physical examination and radiological examination. In physical examination, palpation of the SP in the tonsillary fossa of a patient who has cervicofacial symptoms is one of

the most important findings for the elongated SP. These symptoms tend to increase after the palpation of the elongated SP (2). However, in the literature, some of the studies also indicate that if SP is shorter than 7.5 cm, it can't be palpated (5).

The most commonly used techniques for radiological imaging are panoramic radiography (PR) and computed tomography (CT). Three-dimensional reconstructive imaging computed tomography (3D-CT) is the gold standard for evaluating the anatomy of the region and deciding the type of surgery (6). In the radiological examination, the first parameter to evaluate is the length of the SP. Positional variations of SP, mediolateral angulation, anteroposterior angulation and presence of a fracture are the other radiological parameters to be considered as causing the symptoms (7,8).

Address for Correspondence: Yagmur Barcan

University of Health Sciences Turkey, Istanbul Haseki Training and Research Hospital, Clinic of Otorhinolaryngology, Istanbul, Turkey

Phone: +90 212 453 20 00 E-mail: yagmurbarcan@hotmail.com ORCID: orcid.org/0000-0002-3751-166X

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The differential diagnosis for the elongated SP includes facial neuralgia, oral diseases and temporomandibular joint problems (9,10). These diseases have many common symptoms with ES, but the main distinction is made by the radiological findings.

The aim of this study is to evaluate the data and postoperative results of the patients who have been diagnosed with ES according to the literature.

Materials and Methods

The Ethics Committee of University of Health Sciences Turkey, Haseki Training and Research Hospital approved this study with file number 2020-63 on May 14th, 2020. In this study, 17 patients who underwent surgery for ES between 2015 and 2019 were included. This study is a retrospective case series review.

Study Design

Data of the patients who underwent styloidectomy procedures were collected from the medical records of the hospital. Data about the patients like age, gender, initial symptoms, physical examination information, location (right/left), unilaterality/bilaterality, SP length, presence of fracture, prior tonsillectomy, surgical approach, postoperative complications, complaints that still remain and follow up time accessed from the patient files.

Operation Technique

The transoral approach was applied to 16 patients, and the transcervical approach was applied to 1 patient. Of the transoral surgery patients, 14 of them were operated unilaterally and 2 of them were operated bilaterally. In this study, bilateral cases were counted as a single case for the determination of demographic factors, number of operations and symptoms. All styloidectomy procedures were performed by an experienced otorhinolaryngologist.

Preoperative and Postoperative Evaluation

The intensity of pain was evaluated with the visual analogue scale (VAS) preoperatively and sixth months after surgery (11). Visual analogue scale is a 10 point visual scale which the patients choose a number according to their pain severity (0=no pain, 10=very severe-unbearable pain). Also at the sixth month after surgery, Verbal Response Scale (VRS) was applied to the patients to assess how much relief they felt after the surgery (1=no relief at all, 2=moderate relief, 3=full recovery).

Three-dimensional computed tomography was used in preoperative imaging (Figure 1). This imaging provided the predictions in terms of SP length, angulation, presence of fracture, evaluation of region anatomy and surgical planning. The SP thickness was measured in axial sections while its length was measured in sagittal sections.

Statistical Analysis

SPSS 15.0 program for Windows was used for the statistical analysis. Descriptive statistical methods were given the number and percentage for categorical variables; mean, standard deviation, minimum, maximum, interquartile range for numerical variables. Paired samples analysis was performed with Wilcoxon analysis since the differences of numerical variables did not provide normal distribution conditions. The statistical significance level was accepted as $p < 0.05$.

Results

Fifty eight point eight percent ($n=10$) of the cases were female and 41.2% ($n=7$) of the cases were male. The mean age of our patients was 45.7 (minimum-maximum: 22-66). Five point eight percent ($n=1$) of the patients were under the age of 30, 82.5% ($n=14$) of the patients were between 40-60 years old, 11.7% ($n=2$) of the patients were over 60 years old (Table 1).

According to the physical examination and scanning results, 52.9% ($n=9$) of the patients on the left side, 35.2% of the patients ($n=6$) on the right side, and 11.9% ($n=2$) of the patients on both sides had the elongated SP. The length of SP was between 25-30 mm in 23.5% ($n=4$), 30-40 mm in 47% ($n=8$), 40-50 mm in 11.7% ($n=2$), and more than 50 mm in 17.6% ($n=3$) of them (Figure 2). It was observed that 29.4% ($n=5$) of the styloid processes were fractured in the radiological examination.

Eighty eight point two percent ($n=15$) of the patients had throat pain on the same side, 82.3% ($n=14$) had neck pain, 70.5% ($n=12$) had otalgia, 41.1% ($n=7$) had

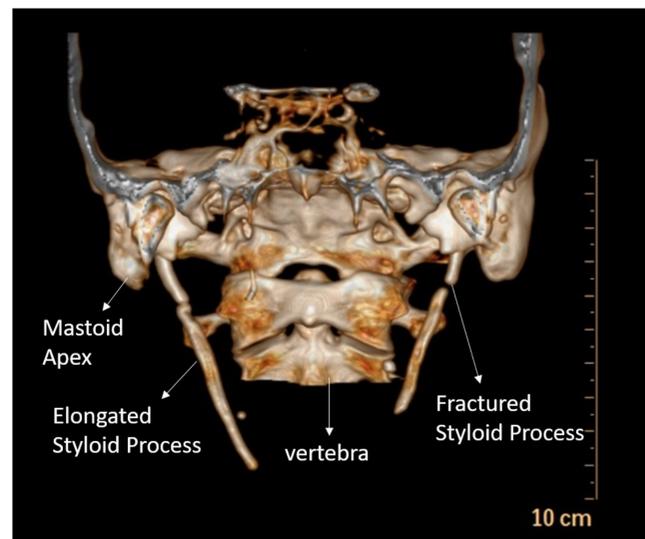


Figure 1. Preoperative 3D-CT scan being used to evaluate styloid process length, thickness and fracture features

3D-CT: Three-dimensional reconstructive imaging computed tomography

STYLOID PROCESS LENGTH OF THE PATIENTS

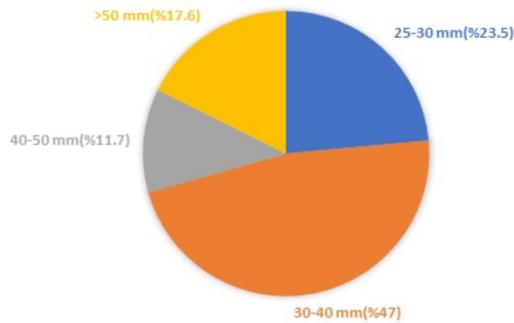


Figure 2. Styloid process length of the patients

jaw pain, 35.2% (n=6) had dysphagia, 29.4% (n=5) had facial pain, 23.5% (n=4) had headache and 23.5% (n=4) had foreign body sensation while less frequently only 1 (5.8%) patient had periorbital pain and 1 (5.8%) patient had stinging sensation in the neck (Figure 3).

During the physical examination, the severity of the pain was high in all patients with palpation of the tonsillar fossa. Seventy-six point five percent (n=13) of the patients had continuous pain while 23.5% (n=4) of them had intermittent pain. The severity of the pain increased when lying down in 29.4% (n=5) of the patients while 70.6% (n=12) of them did not describe any changes in pain with the position.

In the preoperative VAS evaluation, 47% (n=8) of the patients gave 10 points, 23.5% (n=4) of the patients gave 8 points, 17.6% (n=3) of the patients gave 7 points, and lastly, 11.7% (n=2) of the patients gave 5 points for their pain. Sixty-four point seven percent (n=11) of the patients experienced complete pain relief following surgery. Eleven percent (n=2) of the patients did not describe regression (Table 2).

In postoperative evaluation, VRS scores were 1 in 11.7% (n=2), 2 in 23.5% (n=4), 3 in 64.7% (n=11) of the patients. The results of VRS correlated with the postoperative VAS scores in a negative way (p<0.001) (Table 3).

None of the patients had a prior tonsillectomy. Three patients underwent tonsillectomy preoperatively. Five-

Table 1. Demographic features of the patients	
Age	
<30	1 (5.8%)
31-60	14 (82.5%)
>61	2 (11.7%)
Gender	
Female	10 (58.8%)
Male	7 (41.2%)

point eight percent (n=1) of the patients had postoperative wound infection in a total of 17 patients, and this patient was just treated with oral antibiotherapy without surgical drainage. Eleven percent (n=2) of the patients had mild symptoms of first bite syndrome. The symptoms of these patients disappeared in a short time. Also, 23% (n=4) of the patients had numbness in the surgical field after surgery, but these symptoms disappeared in the follow-ups.

Discussion

The styloid process is a needle-like projection of the temporal bone that has close proximity to the jugular foramen and carotid canal (12). ES can cause different symptoms depending on the irritation of the neural and vascular structures by either an elongated styloid process or a calcified stylohyoid ligament during swallowing and chewing. The most common symptoms are throat pain, otalgia, neck pain, and less often, dysphagia, tinnitus, and foreign body sensation (1,3). Many different theories have been considered for the occurrence of pain; a fracture in the ossified stylohyoid ligament with a sudden move, irritation of the glossopharyngeal nerve, trigeminal nerve or chorda tympani, degeneration and inflammation in the attachment point of the stylohyoid ligament, irritation of the pharyngeal mucosa, carotid artery compression and irritation of sympathetic nerves around the arterial wall caused by the elongated SP (13). In our study, similar to the literature, the most frequent symptoms were throat pain, otalgia, and jaw pain, and less often headache and facial pain.

There are two well-defined subgroups of ES; classical type and vascular/carotid artery syndrome type. In classical type, the irritation of the cranial nerve V (trigeminal), VII (facial), IX (glossopharyngeal) and X (vagus) by the elongated SP causes the symptoms; while the symptoms of vascular/carotid artery syndrome type arise from the affected carotid artery and sympathetic nerves (14-17). In recent studies, the internal jugular vein has also been added to these structures (14). In this second subtype, symptoms can vary from just the compression of the vessels and causing carotidynia (a headache which is spreading

Table 2. The evaluation of preoperative and postoperative pain severity

VAS	n	Mean ± SD	Min. - max.	Median (IQR)	p
Preoperative	17	8.41±1.75	5-10	8 (7-10)	0.001*
Postoperative	17	1.77±2.68	0-7	0 (0-4.5)	

*Wilcoxon Analyse
The decrease in postoperative pain scores compared to preoperative pain scores was statistically significant (p=0.001)
VAS: Visual analogue scale, SD: Standard deviation, Min: Minimum, Max: Maximum, IQR: Interquartile range

Table 3. Control of the regression in pain symptoms with VAS and VRS

Patient number	VAS scale			Verbal response scale
	Preoperative	Postoperative	Pain relief	
1	10	0	100%	3
2	8	5	37.5%	2
3	10	0	100%	3
4	10	0	100%	3
5	5	0	100%	3
6	8	5	37.5%	2
7	10	0	100%	3
8	7	4	42.8%	2
9	10	0	100%	3
10	7	7	0%	1
11	10	0	100%	3
12	7	7	0%	1
13	8	0	100%	3
14	5	0	100%	3
15	8	2	75%	2
16	10	0	100%	3
17	10	0	100%	3
Mean	8.41	1.76	76.04%	

VAS: Visual analogue scale, VRS: Verbal response scale

through the occipital pole to the eye) or dissection of the carotid artery which leads to stroke or transient ischemic attack (5,12). Sveinsson et al. (18) reported a case with ES and internal carotid artery dissection which is one of the few cases about carotid artery pathologies with ES (19).

According to the literature all patients in our study were found to be compatible with the classical type of ES.

ES is an idiopathic disease, but it was thought that prior tonsillectomy may be one of the etiological factors. The fibrosis developing after the tonsillectomy operation can cause the symptoms by compression of the neural structures beside the SP (20). However, none of the further studies supported that hypothesis (21,22). In our study, none of the patients had a prior tonsillectomy or trauma history. We accepted the etiology of all patients as idiopathic similar to the literature (23).

The main and long-lasting treatment of ES is styloidectomy operations (24). In surgical treatment, there are two different approaches: transoral and transcervical. As a third approach for some rare cases; those two procedures can be combined. Short hospital stay and absence of external scar are some of the advantages of the transoral approach, while the disadvantages of this procedure include operating in a small area with no good vision, postoperative infection risk and in some cases perioperative tonsillectomy need (25). Muderris et al. (26) compared transoral and transcervical approaches in 2014. They operated 4 patients with the transoral approach while the other 4 patients were operated with the transcervical approach. In postoperative follow-ups, all patients in the transoral group had satisfying recovery except one patient. In the transcervical group, only one patient complained about the external scar. They concluded that both of the approaches were successful and short operation time, absence of external scar and less surgical trauma were the advantages in the transoral approach (26). In our study, we preferred to apply the transoral approach mostly and the results were similar to the literature.

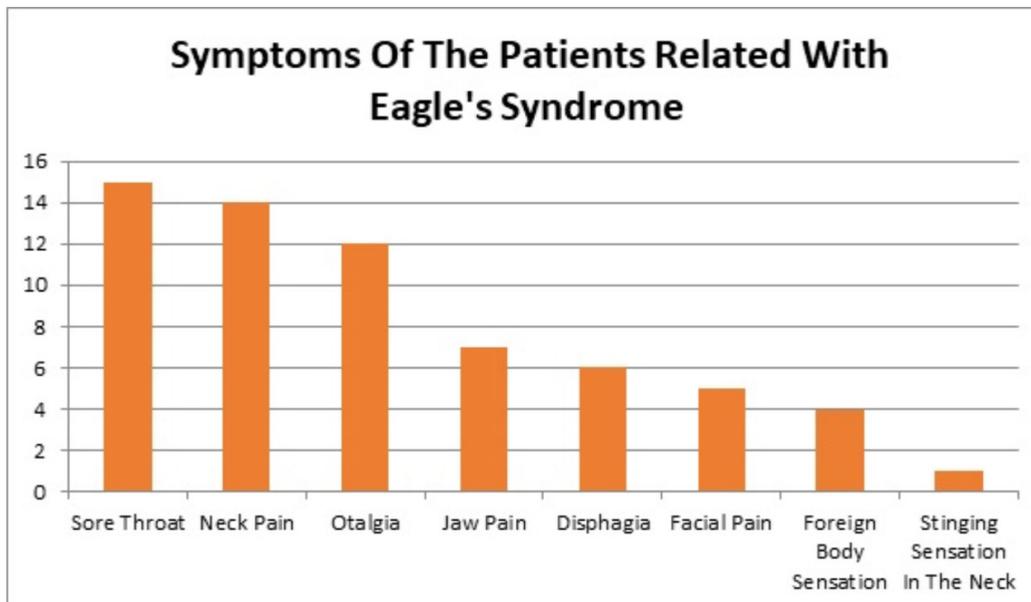


Figure 3. Symptoms of the patients related with Eagle syndrome

A new technique, called transoral robotic surgery (TORS), has been developed in recent years. Kim et al. (27) operated on four patients with this technique, and all four patients had a full recovery without any postoperative complications in follow-ups (27). Rizzo-Riera et al. (28) operated 6 patients with TORS and except one patient who had discomfort by chewing in specific moments; all of the patients had total relief in symptoms. There were no complications except one which was presented with suture dehiscence 4 days after the operation (28).

Fitzpatrick et al. (29) conducted a study with 21 patients and 6 of them were operated with TORS while the rest of the patients were operated with a transcervical approach. As a result, they didn't find a significant difference on symptom reduction between the two approaches. In the same study, there was no difference between perioperative estimated blood loss, operation duration and postoperative hospital stay. Ninety percent of their patients had symptom regression while only 55% of them had a significant recovery. In a study of 133 patients that underwent endoscopic assisted styloidectomy with retroauricular incision by Chen et al. (30), all operations were successful and there was no need to change the technique. Eighty point five percent of their patients had a full recovery, while 15% of them had moderate recovery. They suggested that this is a safe and feasible method in terms of effectiveness, few complications and advantage of cosmetic results. Mevio et al. (31) operated a patient with 3D endoscope-assisted anterior tonsillar fossa approach. They conclude that it's a good alternative for surgical management of ES in terms of high-quality magnification of tonsillar fossa and safe manipulation of the instruments which avoids damage to healthy tissues (31).

The other treatment options are anti-inflammatory, anti-convulsive, anti-psychotic and other analgesic drugs besides surgical intervention (32). However, the disadvantages of these conservative treatments are that the regression of the symptoms is not permanent and there are many possible side effects (16,33).

In our study, we mostly preferred the transoral approach in surgical treatment. The transcervical approach was only used for one patient due to the preoperative 3D-CT scan results and limited mouth opening. Unlike literature, in our study postoperative infection developed in this patient. When we evaluate the other complications in all patients, 2 patients (11.2%) developed first bite syndrome with mild symptoms. First bite syndrome is a clinical diagnosis which is a potential complication of parapharyngeal space operations causing severe pain with the first bite of the meal. There are limited studies of styloid process excision for this complication (25,34). In our study, the

prevalence of this complication is much lower compared to the former studies. The first bite syndrome is a very rare complication, but the operation of ES is related with the parapharyngeal space and always should be kept in mind. In our study, we had complications like postoperative wound infections, numbness around the operation field and first bite syndrome.

The decrease in postoperative headache and facial pain complaints was less than neck and jaw pain in the literature (25). In our study, less regression was observed in the head and face pain similar to the literature.

Study Limitations

The main limitation of the study is the small number of patients. Further studies involving multicentric, large series of cases are needed to generate more data about this disease. Also, another limitation is that there was a significant difference between the transcervical and transoral approach groups in terms of patient number. More patients who were operated with a transcervical approach must be added to further studies to be able to compare these two groups and have more reliable results. In the literature, mostly case reports are present and this study is one of the few studies about ES which is a case series.

Conclusion

The surgical excision of the symptomatic elongated styloid process is a successful treatment option in ES. Dramatic recovery is observed especially in neck, throat, jaw, and ear pain; while the regression of head and facial pain is lower.

Ethics

Ethics Committee Approval: The Ethics Committee of University of Health Sciences Turkey, Haseki Training and Research Hospital approved this study with the file number 2020-63 on 14th May 2020.

Informed Consent: Informed consent was obtained from all individual participants included in the study.

Authorship Contributions

Concept: S.U., O.U., G.G., Y.B., Design: S.U., O.U., G.G., Y.B., Data Collection or Processing: S.U., O.U., G.G., Y.B., Analysis or Interpretation: S.U., O.U., G.G., Y.B., Literature Research: O.U., G.G., Y.B., Writing: S.U., O.U., G.G.

Conflict of Interest: No conflict of interest was declared by the authors.

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