Correlation between clinical presentation and treatment outcomes in Eagle syndrome: a cross-sectional study

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Background: Eagle syndrome (ES) is characterized by neck or oropharyngeal pain due to an elongated styloid process and/or calcified stylohyoid ligament(s). Our aim was to evaluate the clinical presentation and associations of ES as well as the outcomes of various treatments.

Methods: A cross-sectional study was performed on electronic medical records from 2012 to 2021 were searched for the diagnosis of ES made by the treating physician using standard criteria of styloid process measuring ≥3 cm and/or calcification of the stylohyoid ligament. Demographics, symptomology, medical and surgical treatments, and clinical course data were collected retrospectively and analyzed. Styloid length was also measured by two reviewers on prior imaging and analyzed. Continuous variables were analyzed using *t*-tests and categorical variables were analyzed with Chi-square analyses.

Results: Forty-seven patients (25 females, 22 males) with symptomatic ES were identified (mean age 47.4 years; range, 18–86 years), a majority (71.1%) presented with unilateral symptoms. Eleven patients with asymptomatic ES were also identified. A minority of symptomatic patients had a history of prior tonsillectomy (18/47; 38.3%) which was not significantly different from asymptomatic patients (1/11; 9.1%) (P=0.06). Styloid length did not correlate with the presence of symptoms (P=0.54). Laterality of symptoms did not correlate with laterality of radiological findings (P=0.37). A minority of patients (7/25; 28.0%) had improvement in symptoms with medical management. Following surgical treatment, most patients had improvement in symptoms (22/25; 88.0%).

Conclusions: ES can have a varied clinical presentation and is not necessarily related to a history of tonsillectomy as previously described. Surgical treatment seem to have low morbidity and may be effective in improving clinical symptoms in the short term.

Keywords: Eagle syndrome (ES); stylohyoid syndrome; tonsillectomy; surgical outcomes; styloidectomy

Received: 12 May 2023; Accepted: 18 March 2024. Published online: 11 June 2024.
doi: 10.21037/amj-23-80
View this article at: https://dx.doi.org/10.21037/amj-23-80

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Introduction

Background

Eagle syndrome (ES), also known as stylohyoid syndrome, is a rare condition characterized by recurrent neck or oropharyngeal pain due to an elongated styloid process and/or calcified stylohyoid ligament. The most commonly cited incidence of elongated styloid processes is 4%; however, some studies report that the prevalence may be as high as 30% (1,2). In 1937, Watt W. Eagle first characterized stylohyoid syndrome describing two distinct classifications (3,4). "Classic" ES is the elongation of the styloid process resulting in oropharyngeal and neck pain, which may also be referred to the ear or jaw (5). Vascular ES, or styloid process-carotid syndrome and jugular ES, describes the impingement of the internal or external carotid artery or jugular vein by an elongated styloid process, leading to pain and symptoms of carotid/jugular compression (5). Rarely, vascular ES has been known to cause syncope, strokes, and/or carotid artery dissection (6-8).

The exact cause of ES is unknown but it is theorized that it may be the result of retained embryologic cartilage from Reichert's cartilage, which develops into the stylohyoid complex, versus calcification/ossification of the stylomandibular ligament (6). In adults, the normal styloid process length is approximately 2.5 cm and an elongated styloid process is commonly defined as greater than 3 cm which is typically identified on imaging (9). Steinmann proposed several theories regarding the mechanism of ES, including the theory of reactive hyperplasia and metaplasia potentially triggered by a traumatic event such as a tonsillectomy, which has been an implicated cause since Eagle's first characterization of the syndrome (5,10). Both surgical trauma (e.g., dental extraction, tonsillectomy) and accidental trauma (e.g., blunt neck injury, motor vehicle accidents) have been implicated as a possible cause of ES (11); however, it is not clear what impact tonsillectomy or other trauma plays on the development of ES. The pain associated with ES is thought to be a consequence of stretching or compression of cranial nerves (CNs), most commonly CN XII, and nerve endings in tonsillar fossa (12). Furthermore, it is unclear what causes some patients with elongated styloid processes or calcified stylohyoid ligaments to be symptomatic while others are not. Styloid length, angulation, or distance from the tonsillar fossa have been suspected as potential causes (13,14).

Treatment for ES consists of either medical management or surgical intervention. Medical management includes non-steroidal anti-inflammatory drugs (NSAIDs), local steroid injections, neuromodulators (e.g., gabapentin), locally injected or topical anesthetics, or a combination of these therapies (6). Some of these conservative options, notably the neuromodulator medications, demonstrated an 80% decrease in symptoms following treatment for 6 months (6). There are two primary surgical approaches, intraoral or transcervical. The intraoral approach allows for better cosmesis in regards to scarring, while the cervical approach permits for better exposure of the styloid process (6). Prior studies have shown little difference in the efficacy of the two approaches (6,15).

Rationale and knowledge gap

Despite considerable research progress on the ES, significant gaps persist in comprehending its precise etiology, the variability and symptomatic presentation among individuals, and the optimal treatment strategies. The factors determining symptomatic expression and the impact of trauma or anatomical characteristics are not fully elucidated. Additionally, a comprehensive understanding of treatment outcomes and the factors influencing response to medical or surgical interventions is lacking.

Highlight box

Key findings

- Patients with Eagle syndrome (ES) may not have a history of tonsillectomy/trauma and the length of the styloid process is not likely the sole indicator of pathology or risk factor for symptomatology.

What is known and what is new?

- ES is a rare condition characterized by recurrent neck or oropharyngeal pain due to an elongated styloid process and/or calcified stylohyoid ligament.
- This study suggests that history of tonsillectomy does not influence the development of ES and laterality of radiological findings and styloid length does not correlate with location or presence symptoms respectively.

What is the implication, and what should change now?

- Careful correlation of symptoms with imaging as well as assessment of response to attempted interventions assists in appropriate diagnosis. Surgical treatment seems to have a low morbidity and may be effective in improving clinical symptoms.

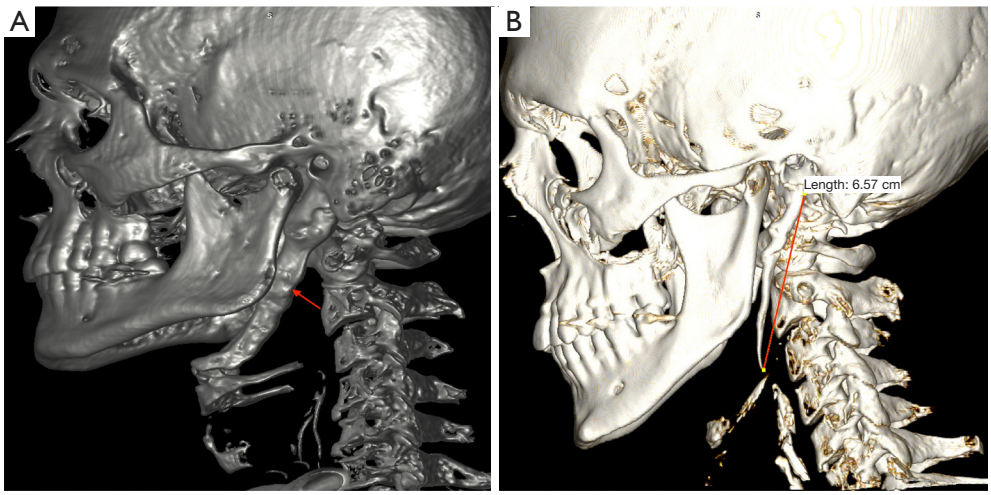

Figure 1 Three-dimensional CT reconstruction of patients with Eagle's syndrome. One with a calcified stylohyoid ligament indicated by the red arrow (A) and one with an elongated styloid process with a demonstrated measurement along the sagittal plane (B). CT, computed tomography.

Objective

The aim of this study was to evaluate the clinical presentation and associations of ES as well as the outcomes of various treatment modalities. Secondarily, we assessed the differences in potential risk factors for symptomatic ES compared to asymptomatic styloid elongation or stylohyoid ligament calcification. We present this article in accordance with the STROBE reporting checklist (available at https://amj.amegroups.com/article/view/10.21037/amj-23-80/rc).

Methods

The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by the Institutional Review Board of the Medical University of South Carolina (IORG 0000020) and individual consent for this retrospective analysis was waived. A cross-section study was conducted on electronic medical records from 2012 to 2021 at our tertiary care institution that contained relevant diagnoses during patient visits were retrospectively collected and analyzed in 2022. Because ES is not an International Classification of Diagnosis (ICD) condition, similar diagnoses of Eagle symptomology were used to search records including those recommended by the American Academy of Otolaryngology (16). The following ICD 10 codes, and their corresponding ICD 9 codes, were used for the initial search: M24.20 (disorder of ligament, unspecified site), J02.9 (acute pharyngitis, unspecified), R13.10 (dysphagia, unspecified), G50.1 (atypical facial pain), R07.0 (pain in throat), M54.2 (cervicalgia), R13.19 (other dysphagia), M62.89 (other specified disorders of muscle), M89.8X8 (other specified disorders of bone, other sites). Patients were included during chart review if a clinical diagnosis of ES was made by the treating physician using standard criteria of styloid process measuring ≥3 cm and/or calcification of the stylohyoid ligament with symptoms reasonably attributable to the syndrome. Example 3D reconstructions of styloid elongation and stylohyoid ligament calcification made using Horos open-source software (version 3.3.6) are shown in *Figure 1*, Videos S1,S2. In patients with isolated styloid elongation, the length was measured by two blinded raters, a clinical research fellow (R.K.W.) and a laryngologist (A.K.O.R.) on 2D sagittal sections of neck computed tomography (CT) imaging using Impax software (AFGA Corporation, Mortsel, Belgium). Data was retrospectively extracted for identified patients, including demographics, symptomology, medical and surgical treatment, and clinical course. Patients with a history of tonsillectomy were compared to assess for differences in clinical presentation, treatments, or outcomes. Symptom improvement was judged to be significant, partial or none based on patient report at follow up evaluation. Significant improvement was defined as complete or nearly complete resolution of symptoms and partial improvement was defined as a noticeable alleviation in symptoms but still present. Asymptomatic patients with incidental findings of

elongation of the styloid or calcification of the stylohyoid ligament were also evaluated. Assessment of calcification of the stylohyoid ligament, styloid length, demographic data, and tonsillectomy history was performed on these patients. Missing data was included in analysis and listed as not reported or not specified.

Surgical technique

For those who underwent surgery, the technique was recorded as either transoral or transcervical. The transoral approach started with incision of the left or right posterolateral pharyngeal wall, lateral or superior to the tonsillar fossa with dissection, skeletonization, and then resection of the distal styloid process or calcified stylohyoid ligament. The cervical approach was completed via an incision posterior to the angle of the mandible. Dissection identified the sternocleidomastoid muscle and then the posterior belly of the digastric muscle. The digastric muscle was dissected posterosuperiorly to the mastoid tip, beyond which the styloid process tip was identified and resected. The approach used was based on patient/surgeon preference.

Statistical analysis

All statistical analyses were performed using SPSS version 27.0.1.0 (IBM Corporation, Armonk, NY, USA). Categorical variables (gender, race, ethnicity, treatments, etc.) were summarized by frequency (n) and percentage (%). Continuous variables (age, pack years, etc.) were assessed for normality using the Shapiro-Wilkes test and summarized by mean ± standard deviation (SD) or median and interquartile range (25–75 IQR) as appropriate. Comparison of categorical variables was performed using a Chi-square with Yates correction or Fisher's exact test. Comparison of continuous variables was performed using either a *t*-test or Mann Whitney rank sum test as appropriate. Finally, intraclass correlation coefficients (K), which are a measurement of the homogeneity of the raters' measurements of styloid process, were also performed. K is an index for the reliability of different raters averaged together. The K value can be interpreted as follows: poor, less than 0.20; fair, 0.21 to 0.40; moderate, 0.41 to 0.60; good, 0.61 to 0.80; and very good, 0.81 to 1.00 (17). A P value of <0.05 was considered significant for all statistical tests. All included P values are of two-sided testing.

Results

Forty-seven symptomatic patients (25 females, 22 males) were identified as having an ES diagnosis at our institution during the study time period. Eleven asymptomatic patients who had an incidental finding of elongated styloid processes and/or calcified stylohyoid ligaments on imaging were also identified from our search. The demographic data for included patients is shown in *Table 1*.

Clinical symptomology

The presenting symptoms of patients with symptomatic ES were throat pain (29/47; 61.7%), neck pain (19/47; 40.4%), dysphagia (18/47; 38.3%), otalgia (15/47; 31.9%), globus sensation (15/47; 31.9%), odynophagia (9/47; 19.1%), dysphonia (9/47; 19.1%), and headache (8/47; 17.0%). Symptoms were typically unilateral (32/45; 71.1%), noting that laterality was not specified in two patients. Median symptom duration prior to presentation was 12 months (range, 1–132 months) and median time to surgery was 4 months (range, 0.4–78.9 months). Additional information regarding clinical symptomology is shown in *Table 2*. Laterality of radiological findings (styloid elongation and stylohyoid ligament calcification) did not correlate with laterality of symptoms as shown in *Table 3*.

Imaging

A CT scan of the neck was predominantly used for evaluation of the styloid process (44/47 symptomatic patients, 93.6%; 11/11 asymptomatic patients, 100.0%). Other radiologic tests included CT angiography of the neck (2/47, 4.3%), neck plain radiographs (2/47, 4.3%), neck MRI (2/47, 4.3%), and panoramic radiograph (1/47, 2.1%; 1/11, 9.1%). Some patients had more than one imaging study performed (6/47, 12.8%; 1/11, 9.1%). Imaging identified either an elongated styloid process (20/47, 42.6%; 5/11, 45.5%), calcification of the stylohyoid ligament (16/47, 34.0%; 5/11, 45.5%) or both (11/47, 23.4%; 1/11, 9.1%). The mean length of the styloid processes among symptomatic patients with isolated elongation was 3.94±0.92 and 4.12±1.29 cm in asymptomatic patients. Most patients (17/20, 85.0%; 3/5, 60.0%) had bilateral elongation, 3 cm or greater, of the styloid processes with the remainder having unilateral elongation (3/20, 15.0%; 2/5, 40.0%). Mean length in symptomatic patients was 3.96 cm on the

Table 1 Demographic data and imaging findings of patients with symptomatic Eagle syndrome and asymptomatic elongation/calcification of the styloid process/stylohyoid ligament

Characteristics	Symptomatic Eagle syndrome patients	Asymptomatic patients	P value [95% CI]
Total	47	11	–
Age (years)	47.4±15.0, 45 [18–86]	58.3±14.18, 56 [30–84]	0.38 [–20.31 to –1.49]
Sex			0.31 [–0.25 to 0.79]
Male	22 (46.8)	7 (63.6)	
Female	25 (53.2)	4 (36.4)	
Race			0.40 [–0.29 to 0.74]
Caucasian	39 (83.0)	6 (54.5)	
African American	6 (12.8)	5 (45.5)	
Other	2 (4.3)	–	
Ethnicity			0.35 [–0.27 to 0.76]
Non-Hispanic	46 (97.9)	10 (90.9)	
Hispanic	1 (2.1)	1 (9.1)	
Alcohol use			0.67 [–0.40 to 0.63]
Yes	19 (40.4)	3 (27.3)	
No	24 (51.1)	7 (63.6)	
History of alcohol abuse	2 (4.3)	1 (9.1)	
Unknown	2 (4.3)	–	
Smoking status			0.24 [–0.21 to 0.83]
Yes	2 (4.3)	2 (18.2)	
No	30 (63.8)	5 (45.5)	
Former smoker	12 (25.5)	4 (36.4)	
Unknown	3 (6.4)	–	
History of tonsillectomy			0.06 [–0.02 to 1.04]
Yes	18 (38.3)	1 (9.1)	
No	29 (61.7)	10 (90.9)	
Imaging findings			0.54 [–0.35 to 0.68]
Calcification of stylohyoid ligament	16 (34.0)	5 (45.5)	
Elongation of styloid process	20 (42.6)	5 (45.5)	
Both	11 (23.4)	1 (9.1)	
Average styloid length in isolated styloid process elongation (cm)			0.54 [–1.15 to 0.79]
Total	3.94±0.92	4.12±1.29	
Left (ICC =0.97)	3.96±1.05	4.15±1.18	
Right (ICC =0.98)	3.91±0.81	3.48±1.00	

Data are presented as n (%), mean ± SD, or median [range]. CI, confidence interval; ICC, intraclass correlation coefficient; SD, standard deviation.

Table 2 Clinical symptomology and medical treatments of patients with symptomatic Eagle syndrome

Characteristic	Values
Total	47
Clinical symptoms	
Throat pain	29 (61.7)
Difficulty swallowing	18 (38.3)
Odynophagia	9 (19.1)
Globus	15 (31.9)
Voice changes	9 (19.1)
Otalgia	15 (31.9)
Facial pain	4 (8.5)
Jaw pain	4 (8.5)
Neck pain	19 (40.4)
Headache	8 (17.0)
Symptom laterality	
Unilateral	32 (68.1)
Bilateral	13 (27.7)
Not specified	2 (4.3)
Duration of symptoms prior to initial presentation (months)	21.8±39.5, 12.0 [6–22]
Medical treatments	
NSAIDs	11 (23.4)
Significant improvement	1 (9.1)
Oral neuromodulators	8 (17.0)
Partial improvement	1 (12.5)
Steroids	3 (6.4)
Partial improvement	3 (100.0)
Opiates	3 (6.4)
Significant improvement	1 (33.3)
Partial improvement	1 (33.3)
No improvement	1 (33.3)

Data are presented as n, n (%), mean ± SD, or median [IQR]. NSAIDS, nonsteroidal anti-inflammatory drugs; SD, standard deviation; IQR, interquartile range.

left and 3.91 cm on the right. For asymptomatic patients the mean length was 4.15 cm on the left and 3.91 cm on the right. Intraclass correlation coefficients for measurement of styloid lengths demonstrated very good interrater reliability (left 0.97, right 0.98). Differences in styloid process length between symptomatic and asymptomatic patients was not significant [P=0.54; 95% confidence interval (CI): –1.15 to 0.79].

Medical treatment

Medical treatments included NSAIDs (11/47, 23.4%), oral neuromodulators (gabapentin and amitriptyline) (8/47, 17.0%), oral steroids (3/47, 6.4%), and opiates (3/47, 6.4%). The oral steroids used were either prednisone or methylprednisolone. Opiates prescribed included hydrocodone-acetaminophen, tramadol, and a topical narcotic not otherwise specified (further data regarding this medication was not available). Significant improvement was identified in one patient with NSAIDs and one patient with opiates. Partial improvement was identified in one patient with neuromodulators, all patients with steroids, and one patient with opiates.

Surgical treatment

The results of surgical treatment and outcomes for patients with ES are summarized in *Table 4*. None of the asymptomatic patients underwent surgery. One patient underwent surgery; however, the styloid process was not removed due to poor visualization. Surgical complications included wound dehiscence (2/31, 6.5%), postoperative bleed/hematoma (2/31, 6.5%), and transient tongue weakness with asymmetric palate elevation (1/31, 3.2%). The patient that developed transient tongue weakness and the other that developed a hematoma both underwent transcervical resection. The patient with transient tongue weakness and asymmetric palate elevation had a negative stroke evaluation and findings were ultimately attributed to a peripheral process secondary to post-surgical swelling. The remaining complications occurred in patients who underwent the transoral approach. The transoral patient with post-operative bleeding was on anticoagulation (warfarin) with increased international normalized ratio (INR) (9.3). Frequency of complications compared between the two surgical approaches was not significantly different and is summarized in *Table 5*. Five patients were admitted post operatively; two were admitted due to surgeon preference, two due to the complications discussed above, and one due to a previous recent carotid dissection.

Three patients had additional or alternative procedures performed. One patient's neck and shoulder pain initially improved after styloidectomy but returned after 1.5 months

Table 3 Comparison of the laterality of symptoms versus radiological findings

Characteristics	Unilateral radiological findings, n (%)	Bilateral radiological findings, n (%)	P value [95% CI]
Total (N=45)	11 (24.4)	34 (75.6)	–
Symptom laterality			0.37 [–0.32 to 0.85]
Unilateral symptoms	9 (81.8)	23 (67.6)	
Bilateral symptoms	2 (18.2)	11 (32.4)	

Radiological findings include either or a combination of elongation of the styloid ligament and calcification of the styloid hyoid ligament. CI, confidence interval.

Table 4 Surgical treatment and outcomes of patients with symptomatic Eagle syndrome

Characteristics	Values
Total patients the received surgery	31
Time from presentation to surgery (months)	10.8±16.7, 4.1 [2.7–7.2]
Surgical approach	
Transoral	27 (87.1)
Transcervical	4 (12.9)
Resection laterality	
Unilateral	17 (54.8)
Bilateral	14 (45.2)
Complications	
None	26 (83.9)
Wound dehiscence	2 (6.5)
Postoperative bleeding/hematoma	2 (6.5)
Transient tongue weakness/edema	1 (3.2)
Admission	
Yes	5 (16.1)
No	26 (83.9)
Symptoms at follow up after surgery (n=24)	
No improvement	2 (8.3)
Partial improvement	9 (37.5)
Significant improvement	13 (54.2)
Lost to follow up after surgery	7 (22.6)
Appointment for same pain after surgery (n=24)	
Yes	3 (12.5)
No	21 (87.5)
Follow-up duration (months)	2.8±3.1, 1.75 [1.0–3.0]

Data are presented as n, n (%), mean ± SD, or median [IQR]. SD, standard deviation; IQR, interquartile range.

with impingement of the paraspinal muscles by the greater horn of the hyoid, completely resolved following a hyoid myotomy. One patient with a long history of dysphonia diagnosed with ES complained of ear and neck pain with speaking which improved after styloidectomy; however, vocal fold injection augmentation was subsequently completed to improve vocal projection and fatigue.

Follow up data was available for 24/31 (77.4%) of surgical patients, with the remaining nine not pursuing post-operative follow-up at our institution. There was no significant difference in outcomes between the transoral *vs.* transcervical approach or bilateral *vs.* unilateral styloidectomy. Three patients (3/24; 12.5%) were noted to have recurrence of their symptoms after surgery. Median follow-up duration was 1.8 months (range, 0.5–13 months).

Association with tonsillectomy

Eighteen out of 47 patients with symptomatic ES had history of prior tonsillectomy (38.3%). Clinical symptomology was not statistically different between the groups, nor were diagnoses (calcification *vs.* elongation), surgical approaches or outcomes (*Table 6*). Only one asymptomatic patient had a history of tonsillectomy (9.1%) which was not significantly different from the number of symptomatic patients that had a history of tonsillectomy (P=0.06; 95% CI: –0.02 to 1.04).

Vascular ES

Two patients presented with symptomology suggestive of vascular ES. The first patient presented with Horner's syndrome and a prior history of possible carotid artery dissection. CT imaging demonstrated an elongated styloid process near his mid-internal carotid artery which has been previously classified as a Type I vascular ES (18). The patient underwent a unilateral transcervical styloidectomy;

Table 5 Surgical complications of styloidectomy by surgical approach

Surgical characteristics	Transoral approach, n (%)	Transcervical approach, n (%)	P value [95% CI]
Total patients that underwent surgery	27 (87.1)	4 (12.9)	–
Surgical complication			0.23 [–0.80 to 3.35]
Wound dehiscence	2 (7.4)	0 (0.0)	
Postoperative bleeding/hematoma	1 (3.7)	1 (25.0)	
Transient tongue weakness	0 (0.0)	1 (25.0)	

CI, confidence interval.

however, follow-up data was unavailable as the patient returned home to a foreign country shortly after surgery. The second patient presented with recurrent headaches, tinnitus, peripheral paresthesia, and mild residual right leg weakness following a stroke one month prior to presentation. CT angiography revealed elongated styloid processes with compression of bilateral jugular veins, worse on the left side, classifying it as a Type II vascular ES (18). Patient underwent bilateral transoral styloidectomy and had post-operative bleeding with a high INR attributable to the patient taking warfarin for antiphospholipid syndrome. He did not note improvement in symptoms in follow-up visit after surgery.

Common misdiagnoses or alternative diagnoses

Common diagnoses prior to diagnosis of ES included glossopharyngeal neuralgia (4/47, 8.5%), temporomandibular joint (TMJ) dysfunction, (4/47, 8.5%) laryngopharyngeal reflux or gastroesophageal reflux disease (4/47, 8.5%), trigeminal neuralgia (2/47, 4.3%), hyoid syndrome (1/47, 2.1%) and myofascial pain syndrome (1/47, 2.1%). After failure of medical treatment for the initial diagnosis, subsequent imaging led to the diagnosis of ES in these patients.

Discussion

ES is an uncommon phenomenon and may be under recognized as an etiology of oropharyngeal, facial, and neck pain. Many of the patients identified in this study had pain for several years prior to diagnosis. The median time to diagnosis was 12 months and some patients experienced pain for up to 11 years. The differential for the classic symptomology of ES is broad, including but not limited to, temporal arteritis, glossopharyngeal and trigeminal neuralgia, TMJ dysfunction, cluster headache, migraine (12). Several of the patients in our study were originally evaluated for other etiologies and often imaging was obtained without a high initial suspicion for ES. While it is important to thoroughly explore other etiologies, ES should be considered, particularly when pain is unilateral, not responsive to analgesics, and reproduced on palpation of the tonsillar fossa (19). Glossopharyngeal neuralgia, which can present very similarly to ES, is more frequently characterized by sudden severe oropharyngeal pain of short duration triggered by hot or cold foods, liquids, or tongue movement (5). Imaging is recommended to evaluate for ES prior to diagnosis of idiopathic glossopharyngeal neuralgia (20).

Key findings

Our key findings highlight that a history of tonsillectomy was not significantly prevalent among symptomatic ES patients, and it did not exhibit any correlation with symptom presence or treatment outcomes. Additionally, factors like the length of the styloid process and disease laterality did not align with the severity of symptoms. Surgical intervention, however, demonstrated a positive impact on alleviating clinical symptoms in symptomatic ES patients, with a low occurrence of complications. The study population revealed a slightly higher representation of women compared to men among ES patients which deviates from the literature.

Strengths and limitations

Limitations of this study are inherent to those of retrospective reviews, including relying on the available data in the electronic medical record. However, records from outside institutions were accessed through CareEverywhere whenever possible. Most results were extracted from our tertiary institution which may limited the generalizability of our data. Our data extraction of

Table 6 Comparison of tonsillectomy and non-tonsillectomy groups

Characteristics	Tonsillectomy, n (%)	Non-tonsillectomy, n (%)	P value [95% CI]
Total	18 (38.3)	29 (61.7)	–
Clinical presentation			0.63 [–0.25 to 0.43]
Throat pain	15 (83.3)	14 (48.3)	
Difficulty swallowing	7 (38.9)	11 (37.9)	
Odynophagia	2 (11.1)	7 (24.1)	
Globus	6 (33.3)	9 (31.0)	
Voice changes	2 (11.1)	7 (24.1)	
Otalgia	6 (33.3)	9 (31.0)	
Facial pain	1 (5.6)	3 (10.3)	
Jaw pain	2 (11.1)	2 (6.9)	
Neck pain	7 (38.9)	12 (41.4)	
Headache	1 (5.6)	7 (24.1)	
Laterality			0.54 [–0.40 to 0.77]
Unilateral symptoms	13 (76.5)	19 (67.9)	
Bilateral symptoms	4 (23.5)	9 (32.1)	
Diagnosis			0.29 [–0.27 to 0.89]
Stylohyoid calcification	9 (50.0)	11 (37.9)	
Styloid elongation	7 (38.9)	9 (31.0)	
Both	2 (11.1)	9 (31.0)	
Patients undergoing styloidectomy			
Total	11 (61.1)	20 (69.0)	0.58 [–0.51 to 0.91]
Transoral approach	11 (100.0)	16 (80.0)	0.11 [–0.14 to 1.33]
Transcervical approach	0 (0.0)	4 (20.0)	
Unilateral excision	6 (54.5)	11 (55.0)	0.98 [–0.69 to 0.71]
Bilateral excision	5 (45.5)	9 (45.0)	
Surgical outcomes			
Total	10 (55.6)	14 (48.3)	–
Significant improvement	6 (60.0)	7 (50.0)	0.46 [–0.35 to 0.77]
Partial improvement	4 (40.0)	5 (35.7)	
No improvement	0 (0.0)	2 (14.3)	

Chi-square (with Yates correction) and Fisher's exact tests were used to compare groups. Laterality was not specified in two patients. CI, confidence interval.

symptom improvement relied on language reported in the patient chart. Additionally, given the lack of dedicated ICD-10 and current procedural terminology (CPT) codes for ES and styloidectomy respectively, we may have failed to capture some patients who were diagnosed with ES at our institution. We attempted to mitigate this by using a broad range of related ICD codes published by the American Academy of Otolaryngology-Head and Neck Surgery

and thorough chart review. Evaluation of previous minor traumas to the tonsillar fossa and surrounding structures was also limited. While the focus of our comparison was on tonsillectomy, we were unable to assess for previous traumas as they seemed to be infrequently reported in the medical record. Furthermore, patient outcomes were evaluated subjectively by patient report rather than a validated measurement as no such tool currently exists. Some patients whose pain did not improve may have been missed if they chose not to follow up at our institution. Lastly, measurement of the styloid process was completed using sagittal CT sections as 3D reconstructions were not readily available. This could evaluate the anterior to posterior angulation but does not account for the smaller mediolateral differentiation.

Comparison with the literature and explanation of findings

Historically, ES has also been thought to be more common in females (20,21). Interestingly, our study had only slightly more females than males (53% female). Bokhari *et al.* also had a similar gender distribution (54% female), which contradicts the 3:1 female:male ratio that is commonly cited (12).

A systematic review examining the association between trauma and the development of ES found that out of 294 articles describing ES, only 13 focused on traumatic events prior to symptom onset (11). This review suggested that a traumatic event could fracture an already elongated styloid process or calcified stylohyoid ligament, or the trauma itself may trigger pathophysiologic mechanisms that lead to lengthening of the styloid process or calcification of the stylohyoid ligament (11). However, the literature remains unclear on how frequently tonsillectomy precedes ES and if the symptomology is different compared to patients without prior history of oropharyngeal trauma. Our study found that <40% of symptomatic patients with ES and <10% of asymptomatic patients had a history of tonsillectomy. There was no statistically significant difference in the clinical symptomatology between patients with or without a history of tonsillectomy. The treatment outcomes were not significantly different between the two groups with greater than 80% of patients in both groups reporting at least some improvement in their symptoms. There was also no significant difference in tonsillectomy history between symptomatic and asymptomatic patients.

Two patients included in our study had features suggestive of vascular ES. One patient exhibited compression of the internal jugular vein by an elongated styloid process which has been reported in the literature and is classified as a Type II vascular ES (18,22,23). The second patient demonstrated an elongated styloid process in close proximity to the mid-internal carotid artery which has been defined as a Type I vascular ES (18). Both forms of vascular ES are rare with limited data on surgical outcomes in the literature. While our patient with Type I ES was lost to follow up, the patient with Type II ES did not demonstrate improvement in symptoms following surgical intervention.

Because styloid process elongation and calcification of the stylohyoid ligament can be identified in asymptomatic individuals, it is important to distinguish ES from asymptomatic styloid elongation or calcification. Our study found that there was no significant difference in the average length of the styloid process between symptomatic and asymptomatic patients. Similarly, prior studies found that the length of the styloid process was not statistically significant between individuals with ES, glossopharyngeal neuralgia, and asymptomatic controls; however, ES patients had a styloid process that was significantly closer to the tonsillar fossa compared to the asymptomatic controls (13). Pokharel *et al.* also found that medial angulation of the styloid process was significant for more severe symptomology (14). These studies demonstrate that the location and orientation of the styloid process should also be considered in addition to the length of the styloid process when evaluating a patient with potential ES. Interestingly, there was no significant correlation between laterality of symptoms and laterality of the styloid process abnormality seen on imaging in our cohort. This means that some patients that had unilateral pathology experienced bilateral symptoms or some with bilateral pathology experienced unilateral symptoms. While those with bilateral pathology and unilateral symptoms may be explained by more severe involvement on the symptomatic side, unilateral pathology with bilateral symptoms, while less frequent, may be explained by compression or irritation of surrounding nerves or muscles and the mechanisms of referred pain and central sensitization (24).

In this study, 32 patients (68%) underwent surgical treatment. This procedure was commonly performed transorally, with preservation of the palatine tonsil. Only four cases were performed transcervically, either due to surgeon/patient preference or concern regarding the proximity of the styloid process to the carotid artery. Surgical treatment was largely successful in relieving clinical symptoms with a low rate of complications, which supports findings in other studies (15,25). Studies evaluating

the complication rate of transoral versus transcervical approaches found that patients who the transcervical approach resulted in a higher likelihood of facial nerve paresis, cervical hematoma, and first bite syndrome (25,26). In our study, the only patients experiencing post-operative hematoma and tongue weakness were those who underwent transcervical surgery which is in concordance with previous findings.

Conclusions

In contrast to previous literature, patients in this study with ES were not more likely to have a history of tonsillectomy and the length of the styloid process did not correlate with symptomatology. Therefore, careful correlation of symptoms with imaging as well as assessment of response to attempted interventions assists in appropriate diagnosis. Surgical treatment seems to have low morbidity and may be effective in improving clinical symptoms in the short term, within three months of surgery.

Acknowledgments

Funding: None.

Footnote

Reporting Checklist: The authors have completed the STROBE reporting checklist. Available at https://amj.amegroups.com/article/view/10.21037/amj-23-80/rc

Data Sharing Statement: Available at https://amj.amegroups.com/article/view/10.21037/amj-23-80/dss

Peer Review File: Available at https://amj.amegroups.com/article/view/10.21037/amj-23-80/prf

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://amj.amegroups.com/article/view/10.21037/amj-23-80/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by the Institutional Review Board of the Medical University of South Carolina (IORG 0000020) and individual consent for this retrospective analysis was waived.

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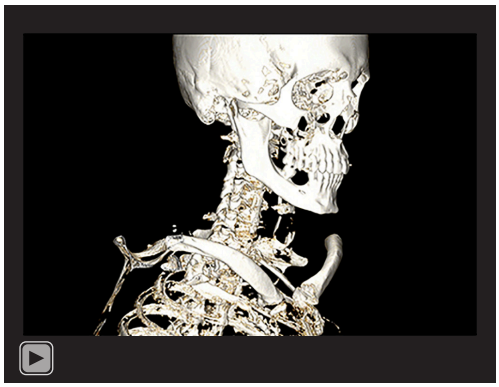
doi: 10.21037/amj-23-80

Cite this article as: Walters RK, Gudipudi R, Nguyen SA, O'Rourke AK. Correlation between clinical presentation and treatment outcomes in Eagle syndrome: a cross-sectional study. AME Med J 2024;9:32.

Supplementary



Video S1 Three-dimensional computed tomography reconstruction of an elongated styloid process.

Video S2 Three-dimensional computed tomography reconstruction of a calcified stylohyoid ligament.

https://dx.doi.org/10.21037/amj-23-80