

EAGLE SYNDROME: A FOLLOW-UP EXAMINATION OF TEN PATIENTS AFTER SURGICAL TREATMENT VIA TRANSORAL APPROACH

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Background: Eagle syndrome is a uncommon pain syndrome caused by elongated styloid process or an ossified stylohyoid ligament. The characteristics of syndrome are covered by large spectrum of symptoms in head and neck regions. Removing the distal part of styloid process is the most effective surgical treatment with good outcomes. **Objective:** The investigators reported ten cases with follow-up examination after shortening styloid process by surgical treatment via transoral approach. **Materials and methods:** Retrospectively collected medical records of 10 patients (7 females, 3 males) with the diagnosis of Eagle syndrome, aged from 28 to 54 years old (mean age=43.2) who underwent removing distal part of styloid process via transoral approach between January 2017 and November 2019. Additionally, the authors created a survey with clinical questionnaire and 4 visual analogue scales to assess the outcomes. **Results:** Duration of follow up ranged from 7 months to 20 months (mean 12.4 months). The mean length of styloid process was 4.71 ± 0.72 cm. All patients showed the low mean visual analogues scales in each categories: (facial pain, foreign body sensation, throat and neck pain and hypoesthesia). There was no report of recurrence of pre-operation symptoms amongst 10 patients. **Conclusion:** Surgical treatment via transoral approach for symptomatic Eagle syndrome has good efficacy and safety for patients. It is an urgent to conduct prospective studies to evaluate the benefits and outcomes of this technique.

Keywords: Eagle syndrome, styloid process, stylohyoid ligament, transoral approach, visual analogue scale.

1. INTRODUCTION

Eagle syndrome was first proposed by Watt Eagle in 1937, following his observation that a rare pain syndrome caused by long styloid process or an ossified stylohyoid ligament and later reported with a case series with over 200 patients^{1,2}. Traditionally, the syndrome contained myriad of symptoms including pain in one side of oropharynx, ear, temporal mandibular joint or face. The prevalence of elongated styloid process in adults populations is approximately 4%, however, there is a small amount of patients in this group having pain². To our knowledge, the normal length of styloid process ranges from 2.5 to 3 cm while having abnormal length if it is over 3.5 cm³. The treatment of Eagle syndrome can be divided into non-surgical treatment with conservative methods of medicine management and surgical treatment.

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However, conservative treatments shows the difficulties to solve all the symptoms in many cases while it can be achieved by surgical management completely. Surgical treatment to shorten the styloid process is divided into transoral approach and cervical approach⁴. The main goal of this study is to evaluate the early outcomes with transoral approach after screening and diagnosis of Eagle syndrome⁴.

2. MATERIALS AND METHODS

2.1. Materials. We retrospectively collected medical records of 10 patients (7 females, 3 males) with the diagnosis of Eagle syndrome, aged from 28 to 54 years old (mean age=43.2) who underwent removing distal part of styloid process via transoral approach between January 2017 and November 2019 in Hue University Hospital.

2.2. Methods. Retrospective case series study.

Variables we collected from medical records included age, sex, presenting symptoms, daily medication, length of styloid process, outcomes in terms of intraoperative and postoperative course, persistence of symptoms and surgical complications.

Preoperatively, all patients had complained of neck pain as their major presenting symptom. Mainly, the pain was located in parapharyngeal area and radiated to the ear and temporal mandible joint. Furthermore, other symptoms were presented included foreign body sensation, dysphagia, pain in mandible or facial. In clinical examination, we palpated the lateral pharyngeal wall, tonsillar fossa and the area between mandibular and mastoid apex to trigger the pain by hand. All patients were preoperatively obtained panoramic radiography and computed tomography (CT) and the technician measured the length of styloid process.

Additionally, 4 visual analogue scales (VAS) were created to assess facial pain, foreign body sensation, throat and neck pain and hypesthesia. VAS is a psychological measurement instrument can measure the intensity of pain, consisting a scale of 0 to 100 mm with 0 mm is no pain and 100 mm is unbearable pain⁵. Patients were explained and trained to estimate their intensity of pain by drawing a vertical line on a continuous line between two endpoints on paper.

Surgical technique: All patients underwent general anesthesia and tonsillectomy in the site having tonsillar fossa palpated to identify the tip of the styloid process. Tonsillar artery forceps are used to dissect through the medial pterygoid and superior constrictor muscles. In the next step, blunt dissection is used to identify styloid process tip. The dissection is then

reached the periosteum of styloid process and the ligamentous parts of the process are removed. A bone cutting forceps is used to remove the process. After following shortening styloid process, tonsillar fossa is closed by suturing.

3. RESULTS

Table 3.1. Patients Characteristics

Parameter	
Sex Male/Female	3/7
Age, (mean±SD), years	42.3±8.72
Duration of symptom, (mean±SD), months	12.4±3.94
Follow up (mean±SD), months	8.5±4.47
History of trauma	0/10
History of tonsillectomy	1/10
Pain unilateral/bilateral	10/0
Length of styloid process (mean±SD), cm	4.71±0.72
Presenting symptom	
Dysphagia	7/10
Foreign body sensation	5/10
Facial pain	2/10
Neck pain	3/10
Throat pain	7/10
Temporo-mandibular joint pain	2/10

We reviewed medical records of ten patients (3 males and 7 females) with duration of symptoms ranged from 7 to 20 months (mean 12.4±3.94 months). There was only one patient having tonsillectomy in history and no patient had trauma in history. All patients complained of presenting symptoms in one side of the throat. According to table 3.1, we witnessed the most symptoms complained by patients were throat pain and dysphagia with 7/10 patients.

There were no early complication after operation included bleeding, infection, carotid injury, post-operative airway edema. The patients showed low mean VAS score for each category (facial pain: 0.8±0.78, foreign body sensation: 0.7±0.82, throat and neck pain: 1.5±0.97, hypoesthesia: 0.7±0.67) (Fig 3.4). None of patients were symptomatic in the follow up and used any kinds of medication related to treating pain in head and neck region.

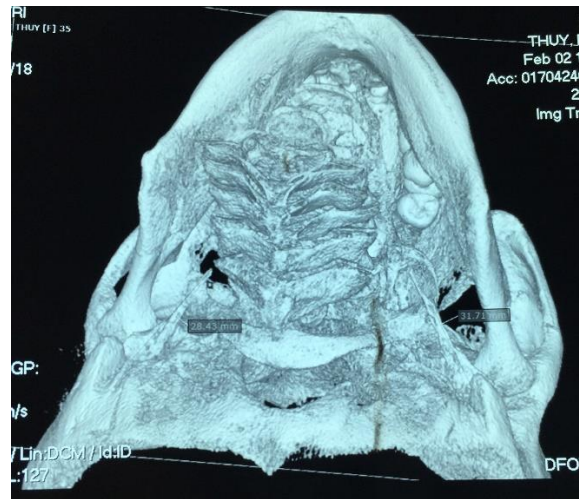


Fig 3.1. A: Preoperative a computed tomography image showing elongated styloid process on the right side. B: 3-dimensional CT showing styloid process



Fig 3.2. Intraoperative transoral exposure of the right elongated styloid process

Fig 3.3. Photograph showing the length of surgical distal end of styloid process removed during surgery

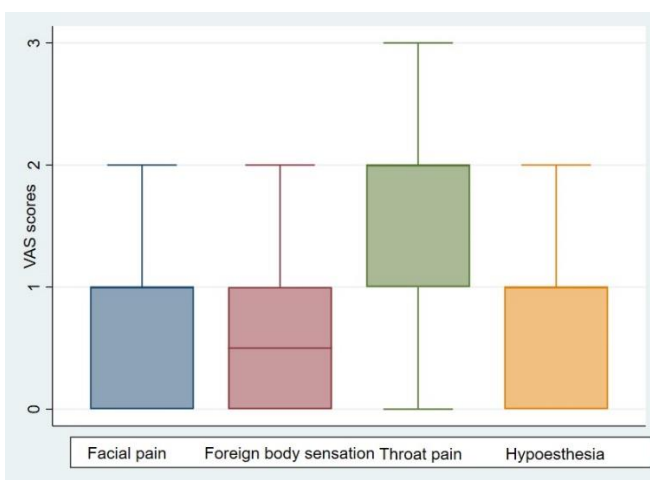


Fig 3.4. Four Postoperative VAS scores

4. DISCUSSION

According to Table 3.1 our case series included 10 patients with 7 females and 3 male, this syndrome seems to occur more often in women⁶. In reference to the aforementioned studies, an elongated styloid process or an ossified stylohyoid ligament can be caused by trauma or tonsillectomy in history but in our case series, none of the patients had history of tonsillectomy or trauma. The prevalence of a patient having an elongated styloid process is 4% and the real incidence of Eagle syndrome is 0.16%². There are many different theories that have been proposed to explain Eagle syndrome but there is no consensus of a completed and clear explanation. One theory indicates retained embryologic cartilage tissue from Reichert's cartilage, a second one proposes the calcification of the stylohyoid ligament and the last explanation is the hyperplasia of osseous tissue at the origin of the stylomandibular ligament⁷. However, a myriad of symptoms such as dysphagia, foreign body sensation, facial pain, neck pain, throat pain, temporomandibular joint pain are likely suggested by the ossified elongated styloid process or stylohyoid ligament, which has a huge impact on surrounding tissues and structures such as the carotid vessels and the carotid nerve plexus⁸.

In our case series, panoramic radiography can play a role in diagnosing and computed tomography (CT) might have further advantage especially 3-dimensional CT can be considered the radiologic test of choice in the determination of anatomic relationship, length, and angulation of the styloid process and we managed to make 3-dimensional CT as a routine radiologic test in the protocol of diagnosing Eagle syndrome⁹.

An elongated styloid process or ossified stylohyoid ligament can be treated with conservative methods and surgical options. While pharmacotherapy and local anesthesia might not solve the problem completely, surgical treatment can achieve a success rate of 80%⁴. Typically, surgical management is divided into the intraoral and cervical approaches. To our knowledge, the cervical approach has disadvantages such as extended surgical time and cervical dissection as well as a visible scar and possible paresthesia of the affected region. On the contrary, the transoral approach has the advantage of an external scar, allowing for better cosmesis, however, it does not allow for complete exposure of the styloid process. Notwithstanding, it does bring a satisfactory outcome in most cases⁴. In the light of the evidence that in our case series all ten patients were asymptomatic in the follow-up. The low mean VAS scores (facial pain: 0.8 ± 0.78 , foreign body sensation: 0.7 ± 0.82 , throat and neck pain: 1.5 ± 0.97 , hypoesthesia: 0.7 ± 0.67) in each item illustrated good condition and patient's quality of life. However,

because of lacking preoperative VAS measurement we could not compare two different mean VAS scores and our results were then not objective.

In this day and age, there has been a modified transoral technique introduced by Torres et al in term of focusing on tonsillar sparing. It starts with an incision in the anterior tonsillar pillar and retraction of the tonsil medially on a bipediculed flap. Secondly, a dissection similar to the one in the traditional approach is then carried out down to the styloid periosteum¹⁰. Al Weteid also reported his cases with the concept of using endoscopic assisting approach¹¹.

In our study, we achieved the main goal of shortening the elongated styloid process or ossified stylohyoid ligament in patient with Eagle syndrome with the successful outcome. Surgical treatment with transoral approach still proved its advantages and feasibility in performing operation.

5. CONCLUSION

Surgical treatment via transoral approach proves the efficacy and safety as treatment in patient with Eagle syndrome in the short term follow up. The limitation of this study is in term of case series study with retrospective study. Long term prospective studies with large cohort must be conducted to evaluate the outcome of this technique.

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