Case Report
Oral Surgery

Transoral endoscopic-assisted styloidectomy: How should Eagle syndrome be managed surgically?

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Abstract. Eagle syndrome, or calcification of the stylohyoid ligament, is a rare condition that may present a clinical diagnostic dilemma for those unfamiliar with its existence and its typical presenting signs and symptoms. Management of this disease process may involve either non-surgical or surgical treatment options. When surgery is indicated, the choice of a specific surgical modality is highly variable and is generally dependent upon individual surgeon preference and experience, since the location of the styloid process is consistent between patients, and the required surgical access is also similar depending upon the specific surgical plan. This paper reports a case of Eagle syndrome managed with a transoral endoscopic-assisted approach, explores the advantages and disadvantages of each surgical approach, and reviews the literature regarding surgical management options for Eagle syndrome.

The styloid process is a thin bony protuberance that emerges from the skull base. It has several attachments including the stylomastoid foramen, the jugular foramen, the mastoid process, and importantly, the carotid canal.[1] Typical styloid process lengths range between 1.0 and 3.0 cm, and styloid processes of more than 3.0 cm are defined as excessively long, although in the articulated, or pseudo-articulated cases, each segment of ossified stylohyoid ligament may be less than 1.0 cm. Bilateral styloid process elongation is the most common presentation, but

interestingly, clinical symptoms are usually present only unilaterally.[2] The stylohyoid ligament is a fibrous band that attaches to the most caudal aspect of the styloid process and the cranial aspect of the hyoid bone (lesser cornu). Mineralization of the stylohyoid ligament occurs infrequently and is usually only an incidental finding noted on routine radiographic examination. Eagle syndrome is diagnosed only when the facial or neck pain originates from a stylohyoid ligament that is calcified. This clinical presentation was identified in 1937 by W.W. Eagle.[1]

Eagle syndrome is divided into two subtypes: classic and carotid. The classic type exists when a calcified stylohyoid ligament results in facial, neck, and throat pain, possible dysphagia, globus sensation, and tinnitus. The pain may radiate to the ipsilateral ear, and turning the head to the opposite side while simultaneously swallowing can reproduce the painful symptoms. These symptoms certainly evoke a wide differential diagnosis and this may make the diagnosis of Eagle syndrome very challenging from a clinical perspective. Also, due to the proximity of

the stylohyoid ligament to the internal carotid artery, a vascular form of Eagle syndrome (carotid type) may occur, with carotid artery compression on head turning, peri-orbital pain, and symptoms of transient ischaemic attacks or stroke (dizziness, headache, or syncope).

The clinical patient examination may allow palpation of an elongated styloid process, or bimanual palpation transorally and transfacially may localize the pain to the retromandibular region. If the pain is reproduced with turning the head to the contralateral side and swallowing simultaneously, there should be a heightened index of suspicion for the presence of Eagle syndrome. From a radiographic perspective, Eagle syndrome may be a common incidental finding on routine panoramic radiographs, although only a very small percentage of patients with calcified stylohyoid ligaments present with clinically significant symptoms. Currently, the use of computed tomography (CT) or cone beam CT (CBCT) scanning, with or without three-dimensional reconstruction, is essential to establish an Eagle syndrome diagnosis and to appropriately plan individual patient management.

Once the diagnosis of Eagle syndrome has been established, there seems to be little consensus regarding the appropriate treatment algorithm to follow.[3–14] Most clinicians will begin with non-surgical options for management using pharmacological therapy with a variety of medications, including non-steroidal anti-inflammatory agents, glucocorticoids, anticonvulsants, and local anaesthetic injections for symptomatic relief of the painful symptoms. Once the decision has been made to treat the patient surgically, there is controversy regarding the most appropriate approach to consider. In general, the position of the calcified and elongated stylomandibular ligament is consistent between patients, therefore the anatomical relationships do not vary a great deal and an individualized approach is not necessary based upon anatomical location alone.

Since there is no consensus in the literature regarding the most appropriate surgical approach to manage Eagle syndrome, the purpose of this study was to review the literature in order to determine whether there is a preference for a transoral or extraoral approach to the elongated stylohyoid ligament in Eagle syndrome, and whether this preference is based upon the specific surgical specialty group involved. In addition, a novel approach to this controversial topic is presented: the case of a patient with Eagle syndrome

treated using an endoscopic-assisted transoral approach is reported; this maximizes the advantages (e.g. the use of a familiar surgical approach) and minimizes the disadvantages (e.g. limited surgical access) of the transoral approach, and minimizes the potential untoward consequences of an extraoral transcervical approach (e.g. unaesthetic facial scar and facial nerve injury).

Materials and methods

A 33-year old Caucasian woman presented to the Department of Oral and Maxillofacial Surgery of the University of Illinois complaining of pain in her left submandibular region that had started 5 years previously, with a history of left-sided neck pain on head movement and swallowing. The patient had consulted several head and neck specialists, including dentists, otolaryngologists, and oral and maxillofacial surgeons, in an attempt to determine the cause of her symptoms as well as to resolve her complaints. Although none of the specialists could determine the aetiology of her pain despite the presence of classic signs and symptoms, as well as radiographic evidence on prior panoramic radiographs and CT scans of the presence of calcified stylohyoid ligaments, she was diagnosed with myofascial pain by one specialist and treated with non-steroidal anti-inflammatory drugs and physical therapy without effect. She then underwent a tonsillectomy and adenoidectomy 2 years later by another specialist in an attempt to alleviate her pain, without any effect. The patient described the pain as a chronic, vague, generalized pain and a sensation similar to a ''...toothpick under my jaw bone with pain that radiates to my left ear.'' In addition, she reported mild dysphagia, sore throat, and functional limitations in her neck movement with resultant pain. She denied any discomfort or symptoms from her right side. In addition, she did not have a history of bruxism, which could theoretically lead to calcification of the stylohyoid ligaments. On physical examination, the painful area was localized to the retromandibular region with bimanual palpation transorally and transfacially, and the calcified stylohyoid ligament was palpable deep in the left tonsillar fossa. The pain was exacerbated by turning the head to the contralateral (right) side and simultaneously swallowing.

A CBCT scan demonstrated nearly complete calcification, without pseudo-articulation, of the left stylohyoid ligament, almost to the level of the hyoid bone, as

well as partial calcification of the stylohyoid ligament on the right side (Fig. 1).

Based upon the history and clinical examination, the decision was made to manage this case of Eagle syndrome surgically with a transoral approach, since prior non-surgical management with non-steroidal anti-inflammatory drugs had been ineffective and since the styloid process was palpable transorally. The patient was taken to the operating room for transoral resection of the elongated calcified stylohyoid ligament. An intraoral incision was made (left lateral posterior buccal vestibular approach, similar to a sagittal split osteotomy incision) and a sub-periosteal dissection was then carried out on the medial aspect of the mandible with blunt dissection (Fig. 2A). The medial pterygoid muscle was identified and bluntly dissected. Once this dissection was completed, the stylohyoid ligament was identified posteromedially (Fig. 2B). Careful blunt dissection was continued using a 4.0-mm zero-degree endoscope to expose the calcified stylohyoid ligament. Two vessel loops were passed around the ligament to retract the superior and inferior tissues and to isolate the ligament. After complete and clear identification of the entire calcified ligament, the ligament was excised from its superior aspect, near the skull base, with a sharp rongeur (Fig. 2C). Dissection was then carried out inferiorly to detach all soft tissues from the caudal aspect of the ligament, and the calcified ligament was then removed in to (Fig. 2D). The length of the resected portion of the ligament was approximately 4.0 cm, and the removal was documented with a postoperative CBCT scan (Fig. 3).

Results

Postoperatively, the patient experienced an immediate relief of the symptoms she had had prior to surgery, including complete resolution of the left-sided submandibular pain as well as the left-sided neck pain during swallowing and turning of her head to the opposite side. Considering these clinical findings, this procedure of endoscopic-assisted transoral resection of the styloid process resulted in a clinical success. At the 6-month follow-up visit, the patient reported no recurrence of her pre-surgical symptoms.

Discussion

Eagle syndrome was documented by W.W. Eagle,[15–19] and is defined as symptomatic mineralization and elongation
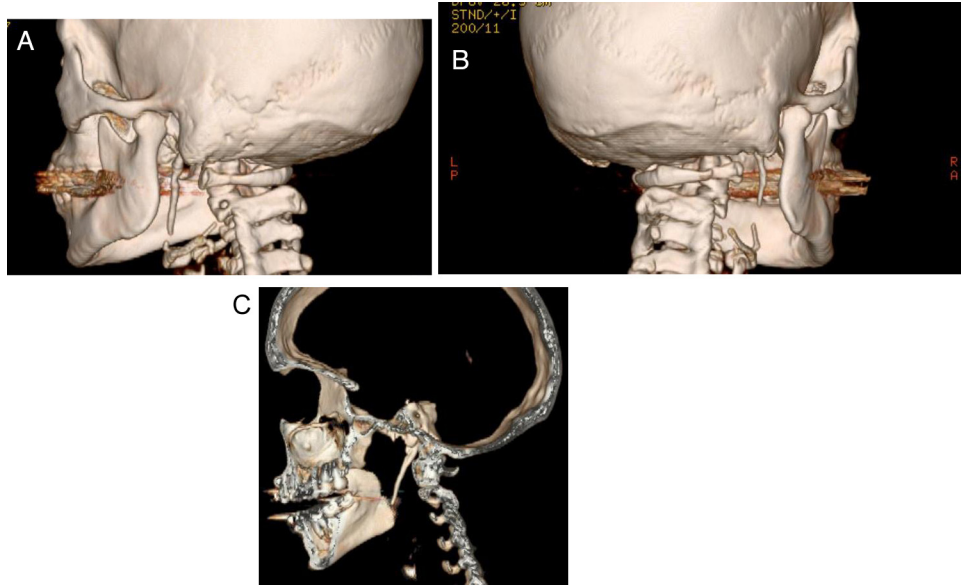
Fig. 1. Three-dimensional reconstruction and sagittal CBCT images showing the elongated calcified stylohyoid ligament: (A) left styloid process, (B) right styloid process, (C) sagittal of left styloid process.
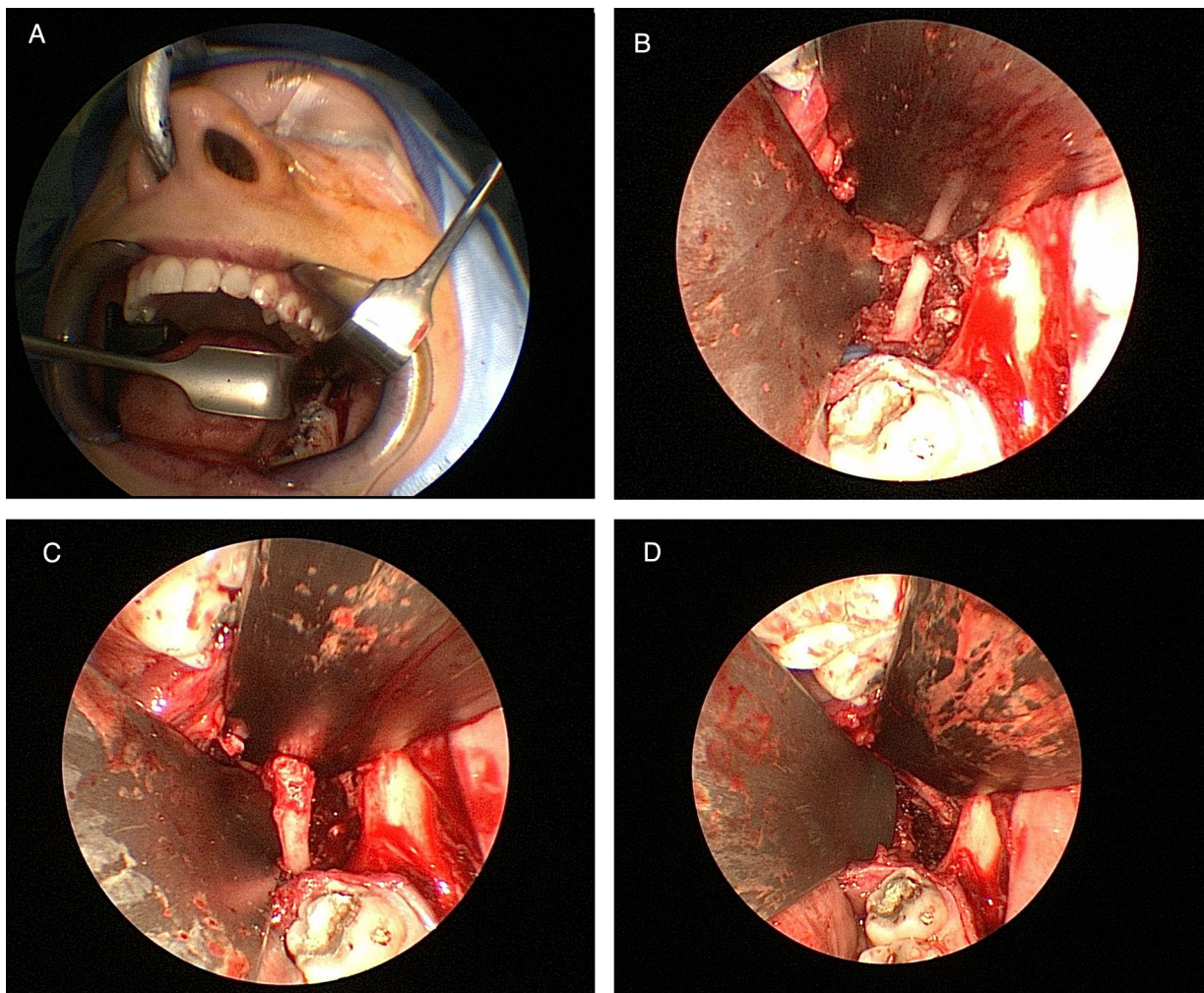

Fig. 2. Intraoperative endoscopic views: (A) the incision, (B) identification of the calcified ligament, (C) resection of the cranial portion of the ligament, and (D) completed removal of the elongated calcified stylohyoid ligament.
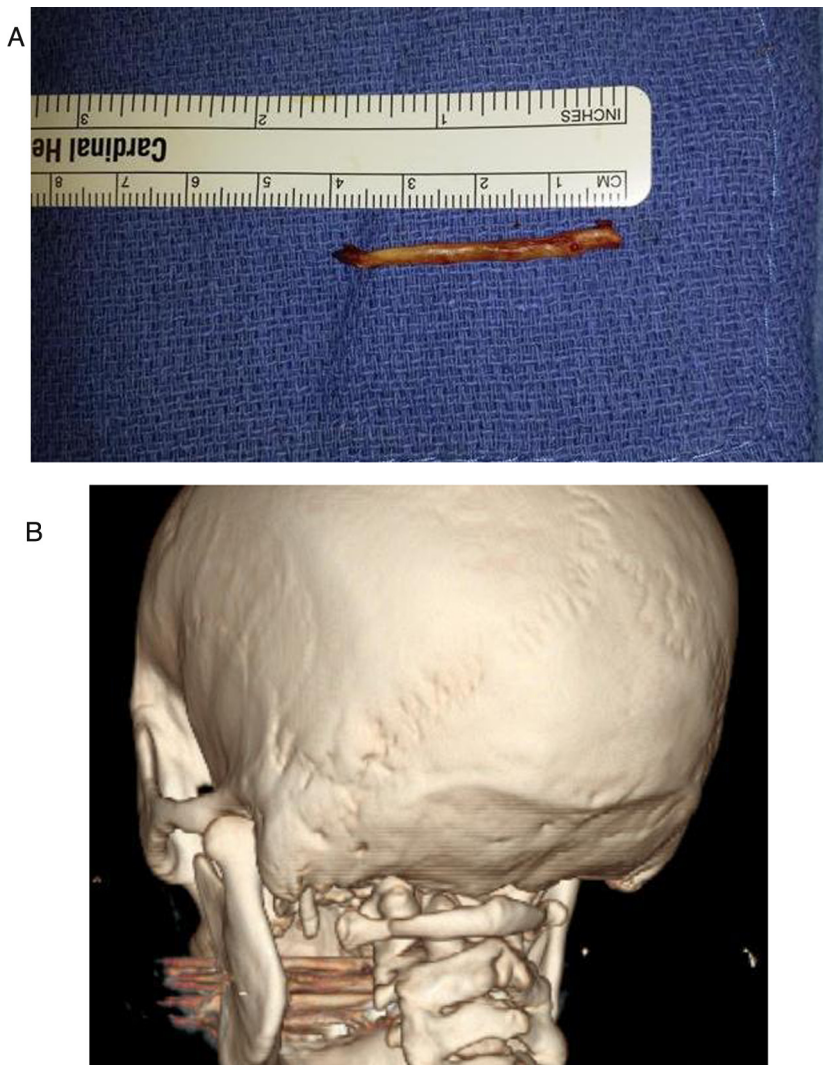
Fig. 3. (A) Specimen of the calcified stylohyoid ligament. (B) Postoperative three-dimensional reconstruction showing the resected stylohyoid ligament with a small portion remaining at the skull base.

One common regimen is to use hydrocortisone 25 mg diluted with 1 ml 0.25% lidocaine injected in close proximity to the styloid process or painful area. Antiepileptic preparations are also used, and 400 mg of carbamazepine per day is used initially, with increased dosage as tolerated for effect for 2–3 weeks. As an alternative to carbamazepine, and without the adverse side effects, gabapentin 300 mg per day, tianeptine 1.5 mg per day, and tramadol hydrochloride 37.5 mg/acetaminophen 325 mg 3 tablets per day has been used.[26] Also, a combination of steroid and local anaesthetic agent (triamcinolone–mepivacaine) can be injected in the tender regions. Subsequently, a stellate ganglion block may be performed once per week for approximately 4 weeks.[26] Other non-surgical treatment methods include pain reduction through physical therapy via relaxation of the associated muscles in the area that has been stretched, constricted, or in spasm as a result of stimulation of the stylohyoid ligament by the calcification process.[25]

The surgical treatment methods involve either fracture or excision of the elongated styloid process via an intraoral or extraoral approach. The two most common surgical approaches described in the literature include either transoral manual fracture of the calcified stylohyoid ligament, or resection of the stylohyoid ligament via a transoral or transfacial surgical approach. The rationale for inducing a fracture of the ligament and allowing retraction either in a cranial or caudal direction is that there is immediate relief of the pressure on the remaining areas of the ligament, which will act to relieve the painful symptoms. Regarding the two surgical approaches (transoral and transcervical), there are advantages and disadvantages of each surgical technique (Table 1).

Although many incisions may be used for the transoral approach, a sagittal split osteotomy incision is useful since it is a well-known approach for most oral and maxillofacial surgeons, and it avoids a blind incision and dissection into an area with vital neural and vascular structures. A

(ossification, calcification, or pseudo-articulation) of the stylohyoid ligament. In fact, Eagle syndrome was first described in 1652 by an Italian surgeon, Pietro Marchetti, and the clinical signs and symptoms were defined by Eagle.[1] The first surgical procedure done for the relief of symptoms associated with styloid process elongation was in 1872.[3] From a demographic standpoint, patients with Eagle syndrome are generally in the fourth to sixth decades of life, although it can occur in adolescents and the elderly. Eagle syndrome is twice as common in women as men.

Both non-surgical and surgical treatment methods exist for the management of Eagle syndrome. For non-surgical treatment, oral medication with gabapentin, antidepressants (e.g. amitriptyline), and anticonvulsants (e.g. valproic acid, carbamazepine) can be used, and corticosteroid or long-acting local anaesthetic injections may be used in the tonsillar region or individual clinically symptomatic areas.[20–25] One of the main components of treatment is the use of glucocorticosteroid hormones, which remain a mainstay of non-surgical therapy for these patients in order to decrease the inflammation of the ligament leading to the painful symptoms.

Table 1. Comparison of the intraoral and extraoral surgical approaches to the styloid process for Eagle syndrome.

Intraoral approach	Extraoral approach
• No external scar	• Cutaneous scar
• Shorter operative time	• Increased operative time
• Restricted operative field	• Risk of facial nerve injury
• Increased risk of deep cervical infection (clean-contaminated wound)	• Better visualization of the surgical field
	• Decreased risk of deep space neck infection (clean wound)

sub-periosteal dissection is carried to the medial pterygoid muscle, which is either retracted or divided to access the styloid process, and this dissection proceeds proximally and distally along the stylohyoid ligament, with surgical resection of the majority of the elongated calcified stylohyoid ligament, which helps to prevent recurrence.

The intraoral approach is favoured from an aesthetic standpoint since it avoids an external incision and resulting neck scar. In addition, the transoral approach may result in the potential for shorter operative times due to accelerated exposure and wound closure. One of the potential disadvantages of the transoral route is exposure of the pharyngeal spaces to the intraoral clean-contaminated environment which elevates the risk of infection, and, therefore, perioperative antibiotics are used routinely. Further potential disadvantages of the transoral access include difficulty with carotid artery exposure and control in the event of iatrogenic intraoperative injury, oropharyngeal and hypopharyngeal airway oedema, and postoperative trismus. With the endoscopic-assisted approach, if significant bleeding is encountered, initial treatment should involve pressure and the cautious use of the endoscope to identify a vessel at the point of severance, and either suture ligation or electrocoagulation, if possible. A contingency plan should include wider surgical access either with extension of the transoral incision, or an additional transcervical incision to gain vascular access. If transoral access alone is not successful, then the most appropriate management of a significant vascular bleed encountered during an endoscopic-assisted approach is to obtain pressure hemostasis and utilize interventional radiology with arteriography to identify the bleeding site, with embolization of the vessel(s) involved. In addition, with the likelihood of mild to moderate airway oedema from a transoral approach, bilateral procedures should not be performed simultaneously. Lastly, full exposure and excision of the entire calcified stylohyoid ligament may not be possible, but excision of even a small component of the ligament (or one or more of the pseudo-articulations) is generally sufficient to relieve the symptoms of Eagle syndrome. The wound is typically closed with resorbable mucosal sutures only. It should be noted that none of the reported studies have described neurovascular injury, deep cervical infection, or long-term complications, or failure to complete the procedure from an intraoral approach.

On the other hand, with the external approach, an incision is made in a skin crease midway between the mandibular angle and the mastoid bone. The sternocleidomastoid muscle is retracted posteriorly, as well as the parotid gland superiorly, and the digastric muscle (posterior belly) inferiorly. This surgical approach may require more time than a transoral approach, with dissection through the fascial layers of the neck and with possible intraoperative facial nerve monitoring, since this approach certainly places the facial nerve at risk of injury, and transient paresthesia may be expected from neurapraxic traction injury. The styloid process is identified and isolated and freed from surrounding soft tissue attachments and distal connections to the hyoid bone. The styloid process is resected partially or completely, and the wound is closed in a layered fashion, with a resulting cutaneous neck scar. The advantages of this external approach include an improved operative field exposure and decreased risk of infection since it is considered a clean wound.

Table 2 shows the results of an evaluation of the literature from 2001 to 2014 regarding the surgical management of Eagle syndrome with a transoral or transcervical approach.[27–47,23,48,5,49,50,22,51–54] Of the 31 articles identified and 92 patients treated, there was a slight preference for the transoral approach (50 cases) over the transcervical approach (42 cases). Only one study reported a complication of the transcervical approach in two of five patients who experienced a transient weakness of the facial nerve (marginal mandibular branch).[22] This review supports the fact that the decision regarding the use of a transoral or transcervical approach is surgeon-dependent and not necessarily based upon a clearly identifiable risk–benefit ratio. This literature review does not assist in the decision-making process for the use of a transoral or transfacial approach, since there is a near even preference and there are very few reported complications. Additionally, we were not able to identify a specialty-based preference (Otorhinolaryngology

Table 2. Intraoral vs. extraoral approach for Eagle syndrome (literature 2001–2014).

	Author [Ref.]	Number	Transoral/ transcervical	Complications
1	Torres 2014 [27]	11	Transoral	–
2	Bertossi 2014 [28]	1	Transcervical	–
3	Dong 2014 [29]	1	Transcervical	–
4	Moon 2014 [30]	1	Transcervical	–
5	Bensoussan 2014 [31]	1	Transcervical	–
6	Ferreira 2014 [32]	1	Transcervical	–
7	Kim 2014 [33]	3	2 Transoral	–
			1 Transcervical	–
8	Kar 2013 [34]	1	Transcervical	–
9	Hoffmann 2013 [35]	1	Transcervical	–
10	Thotappa 2012 [36]	1	Transoral	–
11	Matsumoto 2012 [37]	1	Transoral	–
12	Bahgat 2012 [38]	1	Transoral	–
13	Todo 2012 [39]	2	Transcervical	–
14	Valerio 2012 [40]	1	Transcervical	–
15	Koivumäki 2012 [41]	1	Transoral	–
16	Santini 2012 [42]	1	Transoral	–
17	Mayrink 2012 [43]	1	Transcervical	–
18	Nakagawa 2011 [44]	1	Transcervical	–
19	Yavuz 2011 [45]	27	Transoral	–
20	Dao 2011 [46]	1	Transcervical	–
21	Raychowdhury 2011 [47]	1	Transoral	–
22	Hossein 2010 [23]	1	Transoral	–
23	Shin 2009 [48]	5	Transcervical	–
24	Farhat 2009 [5]	1	Transcervical	–
25	de Souza 2009 [49]	1	Transoral	–
26	Zinnuroglu 2008 [50]	14	Transcervical	–
27	Martin 2008 [22]	6	1 Transoral	–
			5 Transcervical	2/5 VII nerve (marginal mandibular branch) transient weakness
28	Cernea 2007 [51]	1	Transcervical	First-bite syndrome
29	Renzi 2005 [52]	1	Transoral	–
30	Slavin 2002 [53]	1	Transcervical	–
31	Murtagh 2001 [54]	1	Transcervical	–

(ENT), Plastic Surgery, and Oral and Maxillofacial Surgery) for a specific surgical approach. We believe that the use of an endoscopic-assisted approach allows for all of the benefits of a transoral approach with mitigation of several of the negative sequelae of the transcervical approach (e.g. facial scar, increased operative time, facial nerve injury risk), as well as improved visualization of a confined operative site with the use of an illuminated and magnified field of view, which may, in fact, provide a better exposure than a direct transcervical approach.

Eagle syndrome, or mineralization and elongation of the stylohyoid ligament, may be appropriately diagnosed via a detailed history, physical examination, and radiological evaluation. This disease process can be confused with many other conditions that must be excluded in the differential diagnosis and allow appropriate prompt patient management. Based upon the literature, both transoral and transfacial surgical approaches have been used to manage Eagle syndrome. In a review of the published literature from 2001 to 2014, there were 50 transoral and 42 transcervical approaches reported, with rare complications from either approach, mostly due to transient facial nerve marginal mandibular branch weakness. As a result of an evaluation of the advantages and disadvantages of the surgical access options, partial or complete transoral surgical removal of the elongated stylohyoid ligament is the preferred treatment option, since, as opposed to the transfacial technique, it provides a safe approach with decreased operative times and without a facial scar and potential facial nerve injury. In order to overcome the potential disadvantage of limited exposure and visibility, an endoscopic-assisted transoral approach may be employed.

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Competing interests

None.

Ethical approval

Exempt, per Institutional Review Board, University of Illinois at Chicago.

Patient consent

Written patient consent was obtained.

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