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BILATERAL EAGLE SYNDROME PRESENTING WITH CERVICO-FACIAL PAIN: A RARE CASE REPORT

***[1]Dr. Amit Sinha, [2]Dr. Rumit Jha, [3]Dr. Tek Nath Yogi**

*[1]MD (Senior Resident), [2]MD (Junior Resident), [3]MBBS
Department of Radiodiagnosis and Imaging, BP Koirala Institute of Health Sciences (BPKIHS), Nepal.

***Corresponding Author: Dr. Amit Sinha**
MD (Senior Resident), Department of Radiodiagnosis and Imaging, BP Koirala Institute of Health Sciences (BPKIHS), Nepal.
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ABSRACT

Background: Eagle syndrome is a rare condition caused by elongation of the styloid process or calcification of the stylohyoid ligament, leading to irritation or compression of adjacent neurovascular structures. It has two variants: the classic type and the stylocarotid artery syndrome, a vascular subtype where carotid artery compression or irritation of the sympathetic plexus results in neurological and vascular symptoms. **Case Presentation:** An 18-year-old male presented with a three-month history of dull, persistent pain and mild swelling over the left mandibular angle, worsened by mastication and wide mouth opening. Clinical examination showed localized tenderness without infection or lymphadenopathy. Orthopantomogram revealed bilateral elongated styloid processes. 3D-CT confirmed elongation measuring 6.5 cm on the right and 7.6 cm on the left, with segmented calcification. The left styloid process was notably close to neurovascular structures, correlating with unilateral symptoms. A transoral styloidectomy was performed under general anesthesia. The patient had an uneventful recovery and was symptom-free at the 3-month follow-up. **Discussion:** Clinical and imaging findings were consistent with stylocarotid-type Eagle syndrome. 3D-CT played a pivotal role in diagnosis and surgical planning. Transoral styloidectomy, though cosmetically favorable, requires precision due to limited access and proximity to neurovascular structures. **Conclusion:** Eagle syndrome should be considered in patients with unexplained cervicofacial pain. Timely diagnosis and appropriate surgical management can lead to excellent outcomes.

KEYWORDS: Eagle syndrome, Elongated styloid process, Stylocarotid artery syndrome, 3D CT, Transoral styloidectomy.

Highlights

- Eagle syndrome is caused by an elongated styloid process or calcified stylohyoid ligament.
- This case reports a rare bilateral presentation in an 18-year-old male.
- The patient had unilateral cervicofacial pain without classic symptoms.
- Diagnosis was confirmed with 3D-CT; surgery led to complete relief.
- Clinicians should consider Eagle syndrome in persistent facial pain cases

INTRODUCTION

Eagle syndrome is defined as a rare clinical condition characterized by neuropathic and vascular occlusive symptoms caused most likely by calcification of the stylohyoid ligament or temporal styloid process elongation (length, >2.5 cm),[1,2] which was first defined in 1937.[3] The styloid process (SP) is a protrusion of the temporal bone, located between the angle of the mandible and the mastoid process, posterior to the tonsillar fossa and laterally to the pharynx.[1] The SP originates from the second branchial arch and is a slender bone projecting immediately below the ear which extends downward and forward from the inferior aspect of the temporal bone posterior to the mastoid apex. The

tip of the SP is attached to the ipsilateral lesser cornu of the hyoid bone via stylohyoid ligament. The SP can sometimes elongate enough to cause symptoms owing to compression of surrounding the vital anatomical structures. Eagle described homonymous syndrome, which is characterized by an ossified stylohyoid ligament or elongated SP.[4] It has an anatomic relationship with the carotid arteries, internal jugular vein, and facial, glossopharyngeal, vagus, and hypoglossal nerves.[1] There are two types of Eagle's syndrome: classic Eagle's and stylocarotid artery syndrome.[5] In the Classic Stylohyoid Syndrome, the following clinical findings are distinctive: post-tonsillectomy pain in the operation site, stinging pain, dysphagia, painful swallowing, and foreign body sensation. In the Stylocarotid Syndrome, the elongated styloid process deviated laterally or medially, regardless of tonsillectomy, causes compression on the internal and external carotid arteries and perivascular sympathetic fibres.[2,3,6]

In 1937, the otolaryngologist Watt Weems Eagle described the syndrome, having reported >200 cases. Its incidence ranges from 1.4% to 30%, affecting more women than men, with ages ranging from 20 to 40 years. Bilateral stretching is common, but bilateral symptomatology is uncommon.[1] Its etiology is not totally known, and some hypotheses have been accepted, including congenital origin, partial/ complete calcification of the stylohyoid ligament, trauma followed by reactive hyperplasia, metaplasia of the remnants of the second branchial arch.[1]

Symptoms can be bilateral or, more frequently, unilateral[7] and nonspecific, such as pharyngeal globus sensation (55%), recurrent sore throat (40%), bilateral reflex otalgia (40%), headache (25%), carotidynia (20%), reduction of cervical mobility, and pain when opening the mouth. The symptoms appear or worsen on swallowing, chewing, tongue movements, rotation of the head, or palpation of the tonsillar fossa.[1] Eagle syndrome is a rare and unusual cause of head and neck pain that might be confused with oropharyngeal, maxillofacial disorders, myofascial pain, dental origin, temporomandibular joint and particularly with cranial neuralgias but with appropriate clinical history and radiography, Eagle syndrome can easily be identified and treated.[1,8] The diagnosis is based on the patient's detailed medical history,[1] afterwhich radiographic imaging will be typically done to evaluate the length and angulation of the styloid process, often supported by clinical palpation of the tonsillar fossa for confirmatory diagnosis.[5] In propaedeutics, radiography is a limited resource owing to the possibility of overlapping images. The gold standard is computed tomography with three dimensional (3-D) reconstruction, which provides detailed anatomy of the region, enabling surgical programming. Another form of diagnostic suspicion is the temporary improvement of pain after local anesthesia, with return of the symptomatology after the end of the anesthetic effect.[1,2,6,7] Different approaches are available for the treatment of the syndrome, including pharmacological or surgical approaches, or a combination of both.[1] The differential diagnosis of ES should include all conditions that may cause cervicofacial pain.[9]

Conservative treatment can be performed with physiotherapy, analgesics, antidepressants, anticonvulsants, corticosteroids, and even local infiltration with an anesthetic associated with hydrocortisone. However, some patients require surgical intervention to improve the condition, which can be performed either intraorally or transcervically.[1] Finally, the follow-up duration is not yet well established.[1] They range from weekly to monthly consultations, with progressive spacing between them.[1]

CASE PRESENTATION

An 18-year-old male presented to the outpatient department of a tertiary healthcare center with a primary complaint of persistent pain and swelling over the angle of the left side of his mandible for the past three months. The patient reported that the pain was dull and non-radiating in nature, with intermittent episodes of sharp exacerbations particularly noted during mastication, yawning, and wide mouth opening. Over this period, the patient had been self-medicating with over-the-counter analgesics, which provided only temporary relief. However, the pain persisted and gradually worsened, prompting multiple visits to local healthcare facilities without significant improvement. Eventually, he was referred to our center for further evaluation. He denied any prior trauma to the head or neck region, recent dental procedures, or signs suggestive of odontogenic infection. There were no symptoms of fever, trismus, dysphagia, odynophagia, or constitutional complaints such as weight loss or fatigue.

The patient's past medical and surgical history was unremarkable. He was not on any regular medications and had no known allergies. There was no history of similar complaints in the past, nor a family history of head and neck conditions or genetic syndromes. The patient was a non-smoker and did not consume alcohol or use recreational drugs. On general examination, the patient was alert, oriented, and hemodynamically stable. Vital signs were within normal limits. Local examination of the face revealed a mild, poorly demarcated swelling over the angle of the mandible on the left side **(as shown in figure 1A).** The swelling was non-fluctuant, firm to palpation, and mildly tender. There was no erythema or increased warmth over the area. Palpation did not elicit crepitus or discharge. No cervical lymphadenopathy was appreciated. Intraoral examination revealed no dental caries, gingival inflammation, or signs of dental abscess. The floor of the mouth, tongue, palate, and buccal mucosa appeared normal. No tonsillar hypertrophy, peritonsillar mass, or oropharyngeal abnormalities were observed. Mouth opening was within normal range, but the patient reported discomfort on full mouth opening

and during lateral mandibular movements. Cranial nerve examination was grossly normal. There was no evidence of facial asymmetry, neurological deficit, or signs of temporomandibular joint (TMJ) dysfunction. Systemic examination revealed no abnormalities.

In view of the non-specific nature of the symptoms and the location of pain and swelling, radiological investigations were undertaken to evaluate underlying bony pathology. Xray revealed an elongated radio opaque shadow likely styloid process **(as shown in figure 1B**) A panoramic radiograph (orthopantomogram) was obtained as the initial imaging modality. The orthopantomogram revealed bilateral elongation of the styloid processes, more prominent on the left side. The measured length exceeded the normal anatomical range (>30 mm), which raised the suspicion of Eagle syndrome. To confirm the diagnosis and assess the spatial orientation of the elongated styloid processes in relation to adjacent anatomical structures, a computed tomography (CT) scan of the head and neck was performed. The CT scan, with three-dimensional reconstruction, demonstrated elongated astyloid processes bilaterally, measuring approximately 6.5cm on the right and 7.6cm on the left, reaching up to the body of hyoid bone **(as shown in the figure 2**,**4)**. The styloid processes appeared to extend toward the tonsillar fossa region, with closer proximity to neurovascular structures on the left side, correlating with the patient's unilateral symptoms. On 3D reconstructed CT imaging, the styloid processes demonstrated a segmented pattern of calcification with few distinct segments on both sides **(as shown in the figure 3).** Both processes exhibited a knobby or scalloped appearance, with areas of partial and complete calcification interspersed with central radiolucent zones. These radiological features were indicative of a bilaterally nodular complex calcification pattern. Based on the clinical presentation and radiological findings, a diagnosis of bilateral Eagle syndrome, more symptomatic on the left, was made. The patient was counseled about the condition, including the benign nature of the anatomical variation, possible symptomatic management options (such as analgesics, physiotherapy, and corticosteroid injections).

A transoral styloidectomy was performed under general anesthesia, chosen for its cosmetic benefit of avoiding external neck incisions. The styloid process was accessed intraorally through an incision near the ascending mandibular ramus, followed by careful dissection to expose and resect the elongated process using a bone rongeur. The site was irrigated and closed with absorbable sutures. Postoperatively, the patient was managed with antibiotics and analgesics. Recovery was uneventful, and the patient remained symptom-free with improved quality of life at the 3-month follow-up.

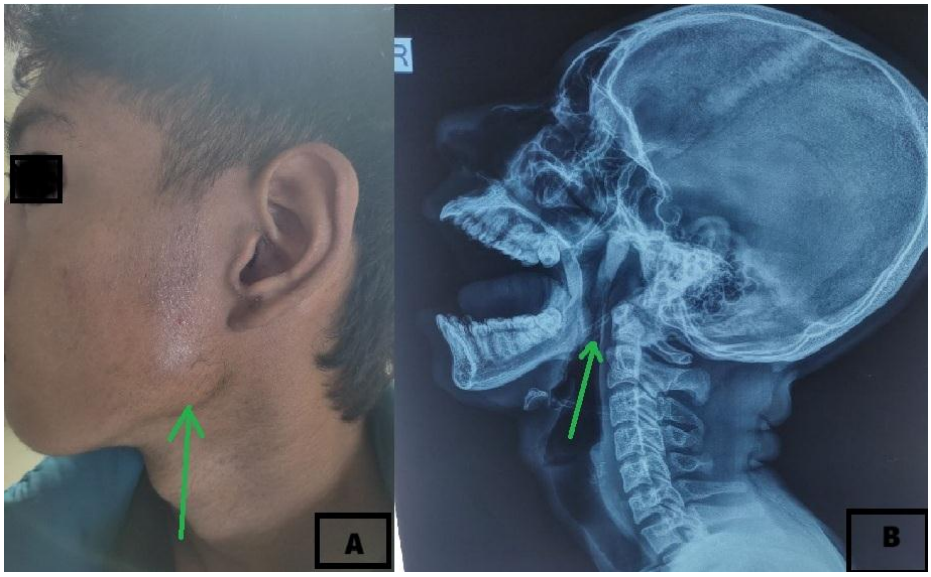

Figure 1: (A). Local examination of the face revealed a mild, poorly demarcated swelling over the angle of the mandible on the left side;(B). Xray lateral neck revealed an elongated radio opaque shadow likely styloid process.

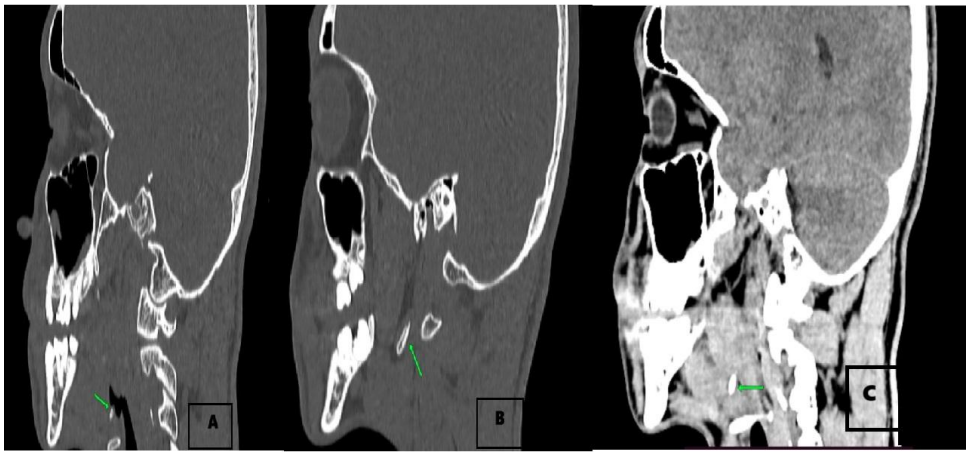

Figure 2: Sagittal CT images showing bilateral elongated styloid processes. (A) Left side measuring 7.6 cm; (B) Right side measuring 6.5 cm, C)Sagittal section of neck region in soft tissue window, the marked arrow shows right styliod process at level of oropharynx superior to hyoid bone (green arrows).

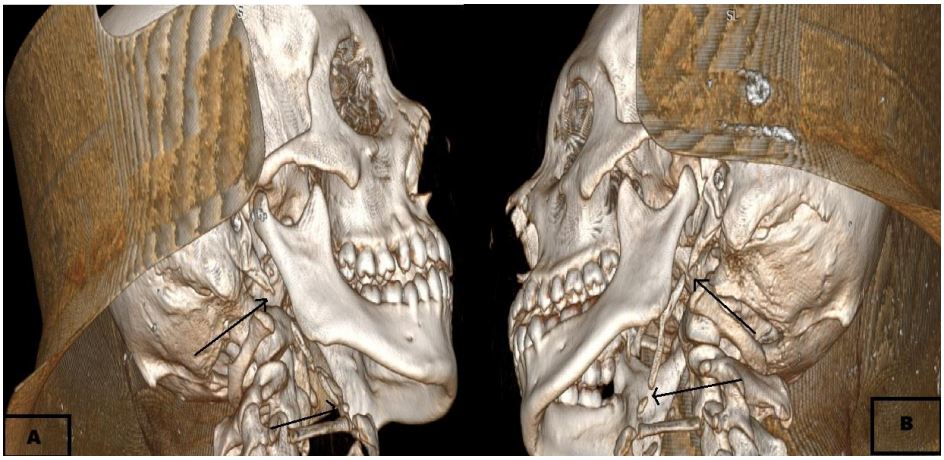

Figure 3: Volume rendered image of skull and neck, arrow shows entire course of styloid process starting from mastoid and reaching just above of hyoid bone with non ossifiedtranshyoid ligament at places.(A) Right Side;(B) Left Side(black arrows).

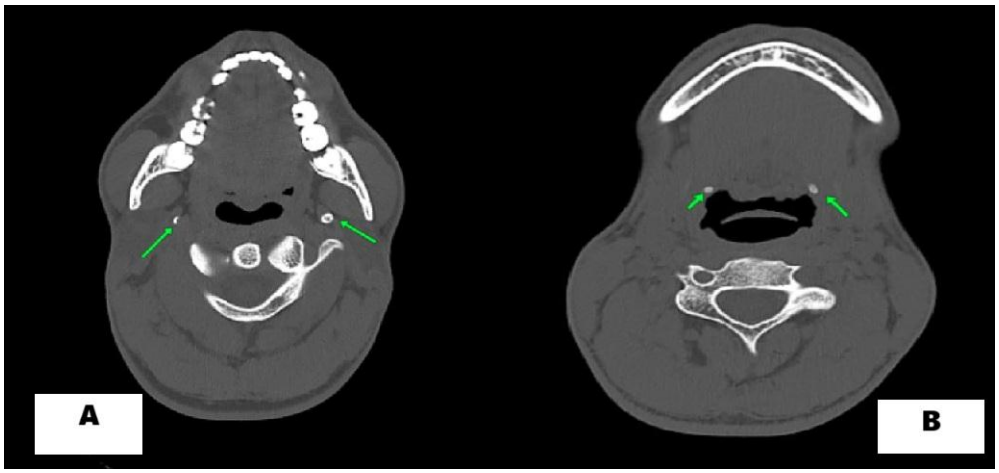

Figure 4: (A); Axial section of skull base bone window at C1-C2 level, the marked arrow shows styloid process on bilateral side in paraphayngeal space region medial to masseter muscle,(B); Axial section of neck bone window at C3 level, the marked arrow shows bilateral styloid process at level just posterior to body of hyoid bone(green arrows).

DISCUSSION

The Eagle syndrome is a rare condition with many uncertainties, which makes most patients seek several proessionals, as the patient reported, before the diagnosis is established.[1] This same condition had happened to our patient, who had been self-medicating with over-the-

counter analgesics, which provided only temporary relief. However, the pain persisted and gradually worsened, prompting multiple visits to local healthcare facilities without significant improvement. Eventually, he was referred to our center for further evaluation.

The styloid process normally has a mean length of 21–29.5 mm. As a result, a styloid process greater than 30 mm in length increased the likelihood of developing Eagle syndrome.[10] While in our case, the CT scan, with three-dimensional reconstruction, demonstrated elongated styloid processes bilaterally, measuring approximately 6.5cm(65 mm) on the right and 7.6cm (76 mm) on the left, reaching up to the body of hyoid bone, well beyond the normal range (>2.1–2.95cm).

During its description, Eagle observed two types of syndrome as follows:[1] classical syndrome, characterized by pharyngeal globus, dysphagia, odynophagia, ipsilateral reflex otalgia, or retromandibular pain, usually initiated after a tonsillectomy secondary to local trauma; and[2] carotid artery syndrome, which occurs independently of previous surgeries in the region, caused by local mechanical irritation, with consequent stimulation of the sympathetic plexus of the carotid arteries. Although two syndromic spectra are defined, in some cases, like the present case, overlapping spectra may be observed, which are often erroneously diagnosed as head and neck disorders. Our case appears to be a carotid-type Eagle syndrome as the patient did not have a history of tonsil surgery, which is usually linked to the classical type. He experienced pain on one side of his lower jaw, specifically around the retromandibular area. Imaging showed that his styloid process was abnormally long and positioned close to important blood vessels on the affected side. He did not show typical throat-related symptoms like difficulty swallowing or a sensation of something stuck in the throat. Since these classical features were absent, and the pain is likely caused by the elongated bone irritating nearby structures, the diagnosis points to carotid-type Eagle syndrome.

The clinical picture is composed of recurrent throat and neck pain, radiating into the ear and dysphagia and Symptoms can be bilateral or, more frequently, unilateral. In this case persistent, dull pain and mild swelling over the left angle of the mandible, with intermittent sharp exacerbations, particularly triggered by mastication, yawning, and wide mouth opening. Physical examination and clinical history are useful diagnostic tools. An elongated styloid process may be palpated during intraoral examination and may provoke the pain.[7] On examination, the swelling was poorly demarcated, firm, non-fluctuant, and mildly tender, without signs of inflammation or discharge. Although mouth opening was within normal range, the patient experienced discomfort on full opening and during lateral mandibular movements.

Correct indication to radiological investigation, dedicated technology and experience are needed in order to reach a diagnosis. Although plain skull radiographs might be sufficient to reveal the anatomical abnormality, CT of the head/neck and especially 3D-CT scan is considered as the gold standard for visualisation of the anatomically complex styloid process, as it avoids the problems of obscured overlapping anatomy.[7] While in our case Initial imaging with a panoramic radiograph (orthopantomogram) revealed bilateral elongation of the styloid processes, with the left side being more prominent. The length of the styloid processes exceeded the normal anatomical range, raising suspicion for Eagle syndrome. To further evaluate the anatomy and confirm the diagnosis, a CT scan with three-dimensional reconstruction was performed. It showed elongated styloid processes extending toward the tonsillar fossa and reaching up to the body of the hyoid bone. The left styloid process was found to be in closer proximity to neurovascular structures, correlating with the patient's unilateral symptoms. Both styloid processes exhibited a segmented, knobby or scalloped pattern of calcification, with alternating areas of partial and complete ossification and central radiolucent zones, consistent with a nodular complex calcification pattern. These imaging features confirmed the diagnosis of bilateral Eagle syndrome, more symptomatic on the left.

However, the surgical approachin patients with a diagnosis of Eagle's syndrome is quite conclusive: surgical styloidectomy has, in fact, a cure rate of 80%.[7] When a surgical technique is opted for, it may be performed intraorally or transcervically.[1]

In this case atransoralstyloidectomy was adopted which was performed under general anesthesia, chosen for its cosmetic benefit of avoiding external neck incisions knowing that transoral styloidectomy approach proved effective, it carries risks such as limited visualization, potential nerve injury, and complications, making it unsuitable for all cases, particularly those with complex anatomy or bilateral involvement. The styloid process was accessed intraorally through an incision near the ascending mandibular ramus, followed by careful dissection to expose and resect the elongated process using a bone rongeur after which the site was irrigated and closed with absorbable sutures.

Finally, the follow-up duration is still variable. Some authors perform weekly follow-up for 2 months, biweekly for the next 2 months, and monthly for other months, while others perform follow-up only in the first 2 months, with discharge after this period. Our patient remained symptom free with improved quality of life at the 3-month follow-up.

CONCLUSIONS

In conclusion, Eagle syndrome should be considered in patients presenting with nonspecific head and neck discomfort, particularly after common causes such as

temporomandibular joint (TMJ) disorders or dental problems have been ruled out. This case illustrates that Eagle syndrome, especially in its carotid-type variation, can present with atypical symptoms, such as jaw pain and swelling, without the usual signs of otalgia or dysphagia. Early and accurate diagnosis relies heavily on radiological imaging, with 3D-CT scans offering the most detailed view to confirm elongation of the styloid process and assess its relationship with adjacent structures. Trans-oral styloidectomy, can be chosen for its cosmetic benefit of avoiding external neck incisions but for more complex cases, a transcervical approach may be preferred to improve visualization and reduce risks of potential nerve injury and other complications. Early and accurate diagnosis of Eagle syndrome is crucial and relies heavily on radiological imaging, with 3D-CT scans offering the most detailed view to confirm elongation of the styloid process and assess its relationship with adjacent structures. This comprehensive imaging approach helps confirm the diagnosis and informs the treatment plan. Additionally, a multidisciplinary team, including radiologists, surgeons, and other specialists, ensures timely diagnosis and optimal management. Long-term follow-up is essential, although the frequency and duration of visits can vary. Monitoring ensures proper recovery and helps identify any complications early on.

Consent

Written informed consent was obtained from the patient's parents/legal guardian for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

The study is exempt from ethical approval in our institution.

Sources of funding

None available.

Conflict of interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Research registration

N/A.

Provenance and peer review

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Table 1: Comparison of typical symptoms of Stylocarotid Eagle's syndrome and the case presented.

Typical symptoms of Stylocarotid Eagle's syndrome	Presented case
Sympathetic plexus irritation can cause neck pain, headache, eye pain, tinnitus, or dizziness.	Head and neck discomfort is presnt.
Neck pain, particularly along the carotid artery	Dull, non-radiating pain over the left angle of the mandible.
Pain triggered by head/neck rotation or movement	Pain exacerbated by mastication, yawning, and wide mouth opening
Symptoms typically unilateral despite bilateral elongation	Unilateral symptoms (left-sided), despite bilateral elongation
Arterial impingement possibly seen on CT angiogram	No CT angiogram performed; 3D CT showed proximity of left styloid to neurovascular structures
Confirmed elongated styloid process (>3 cm)	Bilateral elongation confirmed: 6.5 cm (right) and 7.6 cm (left)
Calcified stylohyoid ligament may be visible	Nodular, segmented calcification pattern on both styloid processes
Management may require surgical resection	Managed with transoral styloidectomy; postoperative recovery uneventful and symptom-free at 3 months

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