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CASE REPORT Open Access

An unusual case of excessive angulation of styloid process and ossified stylohyoid complex causing Eagle's syndrome: a case report

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Abstract

Background Eagle's syndrome or stylalgia is a relatively rare condition that usually presents with throat pain and dysphagia. Most cases are asymptomatic, and diagnosis is difficult owing to the vague nature of the symptoms. There is a female preponderance. In most cases, the bony tip of the styloid process (SP) is palpable upon careful palpation of the ipsilateral tonsillar fossa. X-ray (Towne's view) or an orthopantomogram indicates the length of the styloid process or the ossified stylohyoid complex (OSHC). Computerized tomography (CT) of the skull base, apart from measuring length, provides additional details such as medial or ventral angulation and relation to the surrounding neurovascular structures.

Case presentation A 40-year-old female patient who presented to our hospital with left-sided throat pain and pricking sensation. After workup, she was diagnosed with left-sided stylalgia. X-ray (Towne's view) showed that the left styloid process was 27 mm long, and the right styloid process was 24 mm long. CT of the skull base measured the length and angulation of the styloid process on both sides (sagittal and transverse angles), which were found to be 60° and 61° on the left and 64° and 60° on the right side, respectively. The patient underwent left styloidectomy under general anesthesia. Postoperatively, the symptoms were completely relieved by day 7.

Conclusion Stylalgia is usually diagnosed on the basis of the elongated styloid process. Solely using the length of the styloid process as a parameter for diagnosis leads to misdiagnosis of cases that present with normal length of the styloid process. This article highlights the importance of CT scans in measuring the angulation of the ossified stylohyoid complex in such cases for accurate diagnosis.

Keywords Elongated styloid process, Computerized tomography, Eagle's syndrome, 3D reconstruction

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Background

The styloid process is a bony projection directed anteroinferiorly from the petrous part of the temporal bone, developing from the second brachial arch. The stylohyoid complex consists of the styloid process, with the attachments of the styloglossus, stylopharyngeus, and stylohyoid muscles, and the stylomandibular and stylohyoid ligaments [1]. The normal length of the styloid process is between 20 and 30 mm. Various morphological variants of the styloid process are described based on CT findings: normal, elongated, pseudoarticulated, and segmented [2, 3].

Eagle's syndrome is characterized by an elongation of the styloid process above 30 mm and/or ossification of the stylohyoid complex, usually presenting with a radiating type of neck pain. It may be unilateral or bilateral and is usually idiopathic (Fig. 1). In rare cases, it can occur secondary to the derangement of normal calcium and phosphorous metabolism [3]. As proposed by Steinmann [4, 5], the three mechanisms, which explain the ossification, are reactive hyperplasia, reactive metaplasia, and anatomical variance. Camarda et al. [6] proposed a fourth mechanism of aging developmental anomaly in which there is no ossification seen radiographically.

The clinical presentation can involve the lower cranial nerves, causing cervicofacial pain, dysphagia, otalgia and foreign body sensation in the throat, or compression of the carotid arteries causing syncope. Since the patients usually present with nonspecific symptoms, the differential diagnosis is varied, and thus, many cases are misdiagnosed or undiagnosed despite being symptomatic. Trigeminal neuralgia, temporal arteritis, cervical arthritis, migraine, and myofascial pain dysfunction are the differentials to be ruled out, which can be carried out by careful history taking, clinical examination, and relevant investigations [7].

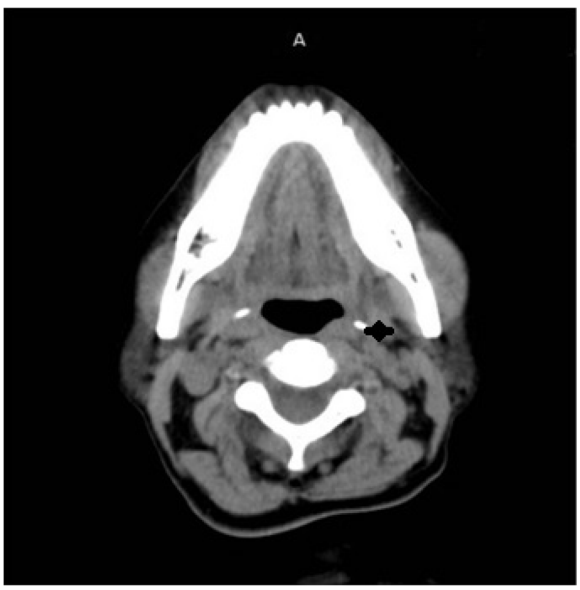

Fig. 1 CT (axial) showing bilateral styloid process (black arrow marking the left styloid bone)

In some cases, despite the length being normal, symptoms appear due to the angulation of the styloid process, especially the medial angulation of the tip, which brings it in close relation to the tonsil medially. A study done by Ondas and workers on 100 patients concluded that there is a statistically significant correlation between length and angulation of the SP, as it shows a great deal of morphological variation [8]. 3D reconstructed CT scan is the gold standard investigation for this disease, as it accurately measures the angulation and relation of the styloid process to surrounding structures. In our case, the presenting symptom was throat pain, and on further evaluation, the styloid process was excessively angulated toward the tonsillar fossa, causing the symptoms.

Case presentation

A 40-year-old female patient presented to ENT OPD with complaints of left-sided throat pain for 1 year, which was insidious in onset and gradually progressive, continuous, shooting type, and of moderate grade. The pain was aggravated upon swallowing and turning the head to the right side, no relieving factors. The patient also complained of foreign body sensation and left-sided pricking sensation in the throat while swallowing. She had a history of constitutional symptoms such as headache and generalized weakness. There was no history of fever, difficulty in swallowing liquids and solids. Her past surgical history was significant for appendicectomy 10 years ago. No other comorbidities were present.

On examination of the throat, mouth opening was adequate, and the lips, gums, buccal mucosa, tongue, anterior pillar, and posterior pillar were normal. There was bilateral grade 1 tonsillar hypertrophy. On palpation, a bony projection was felt in the left tonsillar fossa, and tenderness was present, which mimicked the sharp shooting pain that the patient complained of. The right tonsillar fossa palpation was unremarkable (Fig. 2). The neck, ear, and nose examination was normal. Lab investigations revealed anemia, and all other parameters were within normal limits. X-ray (Towne's view) was taken, and a CT scan was performed (Fig. 3). The length, anterior–posterior, and medial–lateral angulation were measured. The left styloid process was found to be pseudo-articulated (Table 1) (Fig. 4).

As the patient was symptomatic even after optimal medical management, she was taken for surgery (Fig. 5). Left-sided tonsillectomy with styloidectomy

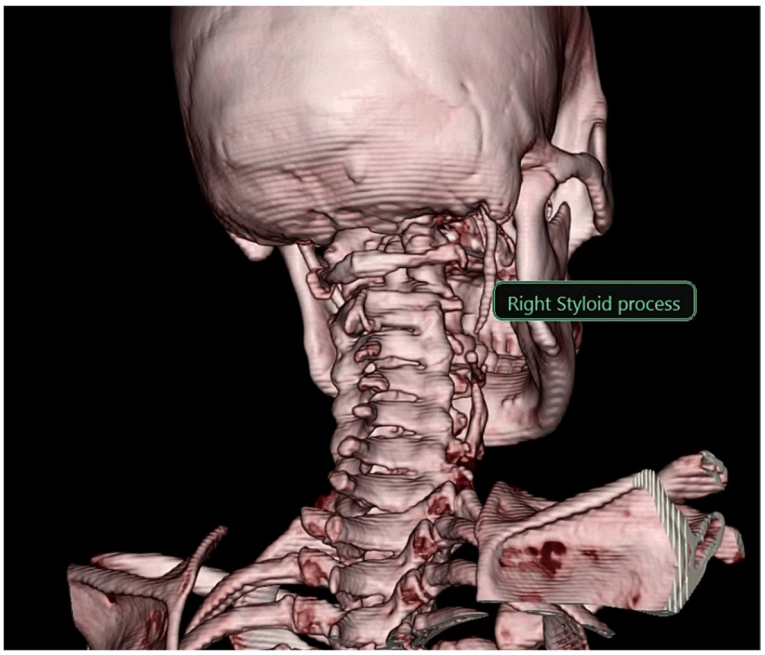

Fig. 2 3D reconstruction of right styloid process

Fig. 3 X-ray—Towne's view showing elongated styloid process

was performed with no intraoperative or postoperative complications. The distal pseudo-articulated styloid was removed and measured 20 mm (Fig. 6). The patient was relieved of her symptoms at the first follow-up after 7 days.

Table 1 Anterior–posterior and medial–lateral angulations of the styloid process

Side	Length	Anterior–posterior angle	Medial–lateral angle
Left	27 mm	60°	61°
Right	24 mm	64°	60°

Discussion

Regarding the angulations of the styloid process, there are two angles of significance: anterior–posterior angle (sagittal angle) and the medial–lateral angle (transverse angle). The anterior–posterior angle (APA) is the angle of intersection between a line tangential to the tip of the mastoid process and the axis of the styloid process on the lateral view of 3D-CT images, and the medial–lateral angle (MLA) is defined as the angle between the line connecting the base and long axis of the styloid process on the anteroposterior view of 3D-CT [9]. This angle is more relevant compared to the APA because the more acutely deviated styloid process will lie directly in relation to the tonsillar bed and may irritate the glossopharyngeal sensory nerve endings. This may cause a sharp, shooting pain as was seen in this patient. Regarding the angulation of the styloid, Buyuk et al. [10] classified the transverse angles into three groups: < 65° were determined as narrow, 65–75° as normal, and more than 75° as wide angles. Similarly, sagittal angles were divided

Fig. 4 3D reconstruction of the left styloid process, showing its increased deviation medially and surrounding anatomy

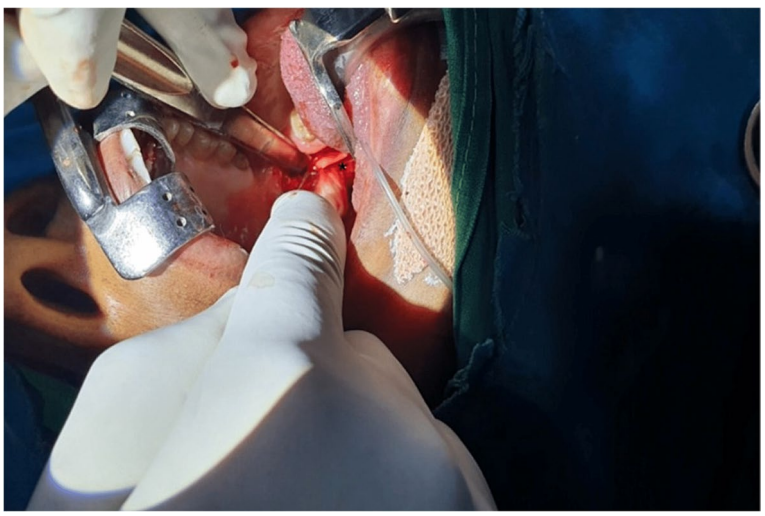

Fig. 5 Intraoperative image, pointer showing the dissected styloid process

into three groups: < 60° were defined as narrow, 60–70° as normal, and more than 75° as wide angles. According to a study by Basekim (2005) [9] and others [10, 11] which considered both length and angulation of the styloid process, SP were classified based on length, angulations, and morphology (LAM classification). The length (L) was classified as short (< 2.00 cm), long (2.00–4.00 cm), and elongated (> 4.00 cm). The angulations (A) were also classified into three categories: narrow (< 65.0°) which was seen in our patient, normal (65.0–75.0°), and wide (> 75.0°). The morphology (M) was categorized as absent SP, normal SP, and other morphological findings which

Fig. 6 Resected left-sided, distal, pseudo-articulated styloid process, which measured 20 mm

were deemed significant; such as the absence of proximal SP, duplication of proximal SP, bent SP, and pseudo-articulated SP, which was observed in our patient.

According to this classification, the patient was L2A1M2 on the right and L2A1M3 on the left, thus explaining the presence of symptoms despite the length being less than 30 mm. It is further validated by the fact that the patient was relieved of her symptoms postoperatively with complete reduction of preoperative pain. Thus, angulations of the styloid process, rather than its length, play an important role in the symptomatology. It is imperative to measure the angulations, more importantly medial–lateral angulation (MLA) on CT scan rather than the length alone to select patients for styloidectomy. Using a comprehensive classification like the LAM classification in the evaluation of patients for Eagle syndrome improves the quality of diagnosis and reduces the incidence of misdiagnosed cases.

Conclusion

Eagle syndrome is suspected when there are complaints of non-resolving neck pain and elongated or palpable styloid process. However, these not only are the criteria to be considered other characteristics of the styloid process, such as angulations and morphology, but also are also important features and will aid in accurate diagnosis. Measuring the sagittal and transverse angles of the styloid process, along with length, correlates well with the symptomatology and helps in accurate diagnosis and selection of patients for surgery, thus improving the quality of care.

Abbreviations

SP	Styloid process
CT	Computerized tomography
OSHC	Ossified stylohyoid complex
APA	Anterior-posterior angle
MLA	Medial-lateral angle
3D-CT	3-Dimensional computerized tomography
LAM	Length, angulation, and morphology

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Authors' contributions

NM examined and diagnosed the patient, performed relevant investigations and interpreted the patient data regarding the syndrome, and operated on the patient. YBS revised the manuscript and examined the data. AP drafted the manuscript and examined the patient. TJ assisted in surgery and gathered data. SD examined the patient and was a major contributor in writing the manuscript. All authors read and approved the final manuscript.

Availability of data and materials

The dataset used and analyzed during the current study is available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Institutional ethics review board approval was taken for this study (S. S. Institute of Medical Sciences & Research Centre, Institutional Ethics Review Board, Reference no: IERB/O No. 611/2022 dated 31 October 2022)

Consent for publication

Informed written consent for publication has been taken from the patient.

Competing interests

The authors declare that they have no competing interests.

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