

## Anatomical Evidence of Styloid Chain Ossification: Review of Literature and Case Report

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**Abstract:** An interesting case of exceptionally long right styloid process of 8.5 cm length with intermittent nodular tuberosities was found on a dry skull of a male. The possible cause of such an anatomical variant and its clinical importance is discussed with relative literature in this paper.

**Keywords:** Elongated styloid process, Stylohyoid chain, Eagle's syndrome, Temporal bone.

### INTRODUCTION

The styloid process (SP) is a slender spike attached to the base of the skull, projecting from the tympanic plate of the temporal bone anteromedial to the stylomastoid foramen. It descends with its tip reaching to a point medial to the posterior border of mandibular ramus. The process is related to significant anatomical structures including; internal jugular vein, glossopharyngeal nerve, vagus nerve, accessory nerve, facial nerve and carotid arteries [1]. The apex of the SP is connected to the lesser cornuae of the hyoid bone by stylohyoid ligament forming styloid- stylohyoid ligament complex or stylohyoid chain [2]. The stylohyoid chain develops from Reichert's cartilage derived from second branchial arch. During third month, the arch divides into 5 segments; the proximal segments, termed tympanohyal and stylohyal give rise to SP. The remaining distal segments; ceratohyal, hypohyal and basihyal form the stylohyoid ligament and lesser cornuae of hyoid bone [3].

The SP is variably developed, ranging in length from few millimeters to few centimeters, but the average length is 2.5cm [4]. It is often straight but occasionally curved anteromedially. Abnormal elongation of SP or ossification of stylohyoid ligament may cause compression of adjacent vital anatomical structures producing multiple symptoms including chronic pharyngeal pain, radiating otalgia, foreign body sensation, dysphagia, pain on head rotation, dizziness & headaches described as Eagle's syndrome [5].

The present case describes an unusually long SP in an adult human skull on right side that may have been overlooked during clinical examination. This case is unique as the elongated process showed intermittently nodular tuberosities at four sites, which correlates with its embryological development.

### CASE REPORT

During a study on dry human skulls, we incidentally encountered a skull with distinctly long right SP. The length of elongated right SP, measuring from the caudal margin of the tympanic plate to the tip was 8.5cm. The elongated right SP also showed increased acuity in deviation from norma, both anteriorly and medially. Its medial deviation & anterior angulation were 45° & 60° respectively. The tip of the SP was reaching anteriorly upto the level of maxillary tuberosity and was 2.5cm away from mid-sagittal plane. Interestingly, the elongated styloid process (ESP) showed four nodular tuberosities of varying thickness along its length. The first proximal tuberosity was 1.0cm in diameter and was located 0.5cm distal to tympanic plate. The second tuberosity also had a diameter of 1.0cm, but was placed 1.7cm distal to tympanic plate. The third tuberosity was 0.7cm in diameter and was situated 4.0cm distal to tympanic plate. The fourth, distal most tuberosity, had a diameter of 0.4cm and was sited 7.8cm distal to tympanic plate. On the other hand, the left SP was normal measuring 2.5cm in length. All the measurements were taken with the help of standard ruler and angles were measured with protractor.



**Fig. 1: Showing elongated styloid process in right lateral view (A) and posterior view (B). Both the views are demonstrating elongated styloid process extending from the tympanic plate with 4 intermittent nodular tuberosities (\*)**

## DISCUSSION

The length of SP ranges from 0.5 cm - 5.0 cm, but is usually considered normal upto 3.0 cm [2]. The incidence of ESP has been reported in 4% of population with frequent bilateral occurrence and female dominance. However there are reports contradicting these above claims [6, 7]. Most patients with ESP were more than 50 years and symptoms ranged from mild to severe [8].

An abnormally long SP is a rarely identified anatomical and clinical entity, and was first described by Manchetti of Padua in 1652 [2]. An ESP is not symptomatic but the symptoms range from mild to severe. A constellation of associated symptoms characterized by throat pain, limitation of neck movements, hoarseness, dysphagia and foreign body sensation is termed as Eagle's syndrome [9]. The syndrome derived its name from Watt Weems Eagle, an American Otolaryngologist who first described in detail the clinical sequelae associated with ESP in a series of 200 patients [10]. He described two patterns of clinical presentations, one characterized by dysphagia, dull nagging pharyngeal pain radiating to ear, especially on neck rotation, attributed to impingement of glossopharyngeal nerve and adjacent cranial nerves. The second presentation showed carotid artery syndrome, due to irritation of sympathetic nerves accompanying the internal and external carotid arteries characterized by sharp pain referred along the distribution of vessels involved. In the involvement of external carotid artery, the pain is experienced in neck radiating to eye, ear, mandible, soft palate and nose. If internal carotid artery is affected, symptoms are parietal headaches and pain along the ophthalmic artery branches [11]. Apart from these symptoms, there are unusual reports of clicking sound on jaw movement, cases of fractures of ESP and even incidences of suicide

of patients with severe pain and psychotraumatic stress [12, 13].

Early reports suggested that the SP was considered elongated only if its length exceeds 3.0-3.3 cm [8]. Later, several authors claimed that, a threshold length of 4.5 cm or more should be accepted as abnormal [14]. In the present case, a length of 8.5 cm has been reported, which is probably the maximum length of SP ever reported. Further, it has been emphasized that the length in isolation is not the risk factor, but its combination with increased acuity in deviation, both anteriorly and medially make the ESP symptomatic [15]. In 267 patients, Montalbetti reported a medial angulation range of 10° -27° and anterior angulation range of 30° - 50° [3]. In the present report, we documented a medial deviation of 45° and anterior angulation of 60°. The thickness of normal SP varies between 0.2 - 0.5 cm but in our case, thickness varied from 1.0 - 0.4 cm proximo-distally [3].

The styloid-stylohyoid ligament complex (stylohyoid chain) develops from 2<sup>nd</sup> branchial arch, initially consisting of a cartilage; the Reichert's cartilage. The cartilage forms the template for the stylohyoid chain [16]. During the 3<sup>rd</sup> month the arch divides into 5 segments, the proximal 2 segments, known respectively from above as tympanohyal, stylohyal give rise to SP. The remaining segments; ceratohyal, hypohyal and basihyal form the stylohyoid ligament and lesser cornuae of hyoid bone. In humans, the ceratohyal element of 2<sup>nd</sup> branchial arch may retain some of its embryonic cartilage and gets partially or completely ossified resulting in an ESP [3]. In lower species, the ceratohyal segment ossifies to form a distinct bone termed epihyal. A chain of 4 bones connected by fibrous tissue performs the function of stylohyoid ligament here [16]. Similar morphology of

stylohyoid chain with intermittent joints has been reported in few incidences in man [11, 17].

Steinman proposed 3 theories to explain the ossifications. Firstly, the theory of Reactive Hyperplasia, which states that ossification begins at the terminal end of SP at the expense of stylohyoid ligament. Secondly, the theory of Reactive Metaplasia states that the separate ossification centres within the 5 segments initiates ossification and become intermittently ossified. Thirdly, the theory of Anatomic variance proposed that early elongation of SP or ossification of stylohyoid ligament are anatomical variations that occurs even in children without recognizable pharyngeal trauma [16]. Later, Camarda proposed a fourth theory of ageing developmental anomaly to explain "Pseudo-stylohyoid syndrome", where the symptoms occur in the absence of radiologic evidence of ossification. This theory states that with ageing, the soft tissues loose elasticity, the stylohyoid ligament stiffens leading to inflammatory reaction mimicking the Eagle's syndrome [18].

The present case of ESP showed 4 intermittent nodular tuberosities along the length, supporting the theory of Reactive metaplasia suggesting ossification at multiple segments which later fused to form one ESP. The nodular tuberosities represent the sites of fusion between the 5 embryonic segments of 2<sup>nd</sup> arch, which were destined to form SP, stylohyoid ligament and lesser horn of hyoid bone. Though stylohyoid apparatus variations are frequent, completely ossified unilateral stylohyoid chain with intermittent tuberosities is rare. This makes the present case unique. Moreover, there is paucity of literature correlating the SP appearance with theories prevailing.

## CONCLUSION

The present report of an extremely elongated, undiagnosed case of styloid process highlights the need of including the possibility of ESP as a routine part of differential diagnosis especially in cases of unexplained throat and ear pains.

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