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Study of Anatomical Variants of the Temporal Bone Pen Process during Eagle's Syndrome

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Abstract

Anatomical variations of the pen process (PS) and ossification of the pen-hyoid ligament constitute Eagle syndrome. This is a rare syndrome, often bilateral, that summarizes the clinical manifestations due to an excessive length of the pen process that is greater than 25mm. Other important morphometric and topographical variations are associated with this anomaly.

Taking these anatomical data into account is essential for the etiopathogenic analysis and surgical treatment of Eagle syndrome. The purpose of this work was to investigate anatomical variations of the pen process during Eagle syndrome.

Materiel and method: This was a retrospective study over a period of 07 months from December 2020 to June 2021 inclusive. It concerned 32 patients, received in the radiology and medical imaging department of the National Pikine Hospital for suspicion of Eagle syndrome confirmed by scanner without and with injection. Patients who had a normal scanner were not included in the study. We used SPSS 24.0 and Excel 2016 to analyze the data.

Resultats: 24 women (75%) and 8 men (25%), a sex ratio of 0.33. The average age of patients was 39 years, a standard deviation of 12 years and extremes of 21 to 71 years. The age range of 30-40 was 34% and 41-50 was 33%. Examination found pharyngeal foreign body sensation in 65% of cases, dysphagia in 67% of cases, headache in 71% of cases and cervical neuralgia. Brachial in 95% of cases. At CT, the average length of all pen processes was 39.2 mm (extremes: 21-76 mm). The elongation of the pen process was bilateral in 68%, unilateral right in 7% of cases, unilateral left in 25% of cases. Morphologically, the PS consisted of a single long fragment in 78% of cases (Langlais types 1), or two fragments articulated either by fibrous tissue or by a pseudoarticulation with an angular shape in 19% of cases (Langlais types 2), or several fragments (3% Langlais types 3).

Conclusion: Eagle syndrome is a radioanatomical entity with several variants whose knowledge is essential to its management.

Keywords: Styloid process; Eagle; morphometry; CT scan.

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INTRODUCTION

Anatomical variations in the styloid process of the temporal bone or ossification of the stylohyoid ligament (LSH) or both, define Eagle Syndrome (SE) according to Dr. Watt Weems Eagle since 1937.[1,2]

It is characterized by an abnormality in the length of the pen process, the measurement of which is greater than 25mm, associated with very heterogeneous clinical signs.[3-7]

According to the literature, this is a rare, clinically poorly known syndrome that can present a multitude of symptoms affecting the cerebral and cervical regions[7,8], which can occur at any age.[9,10] Its symptomatology is bilateral in most cases.[10,11]

Surgery has been shown to be effective in its management but the results are influenced by several factors including clinical signs, type and projection area of the pen process. This underlines the importance of the morphology of the pen process and its anatomical relationships in the selection of surgical candidates.[12]

It is in this context that we undertook this work in order to describe the anatomical variants of the pen process that can enter Eagle syndrome and to understand their locoregional complications.

MATERIAL AND METHOD

We selected patients referred for clinical suspicion of Eagle syndrome confirmed by the cervical scanner whose images were analyzed through a Syngo Acquisition Workspace post-processing console in bone window (FO) and small parts (PM) with MPR, MIP, and Volume Rendering (VRT) reconstructions.

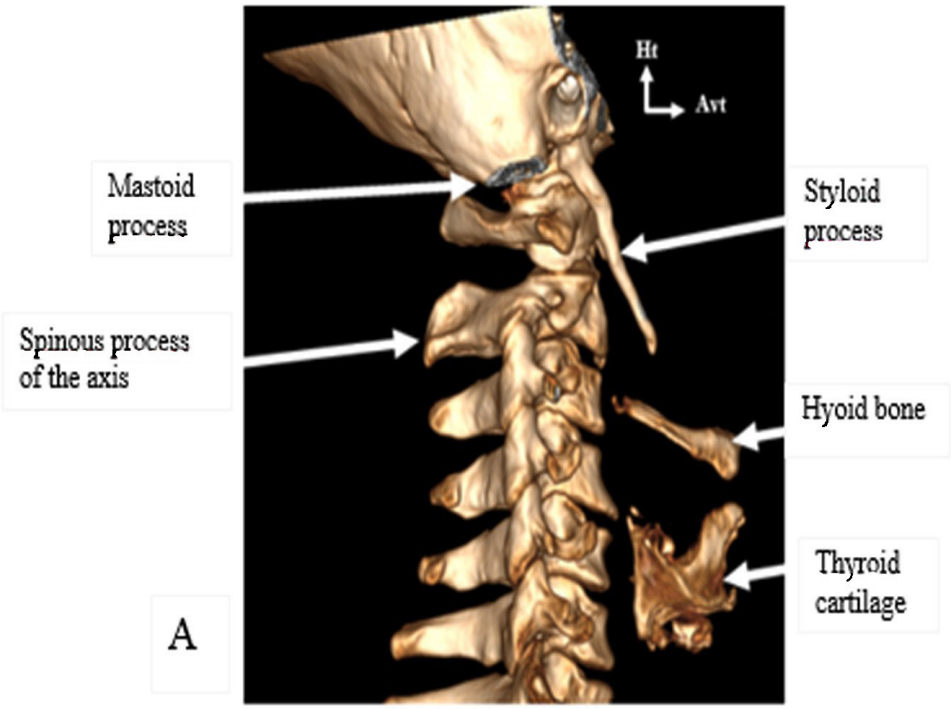
The measurement of the process was carried out from its implantation base at the level of the temporal bone to its tip which constitutes its termination and includes the ossified part of the pen-hyoid ligament. For each patient, age, sex, clinical signs and the cardiovascular complication of the sample were recorded.

RESULTS

These were 32 patients of which 24 women (75%) and 8 men (25%) hence a sex ratio of 0.33.

The average age of patients was 39 years with a median of 37 years, a standard deviation of 12 years and extremes of 21 to 71 years. The age group of (21-29) represented 18% of the population, that of (30-39) was 34%, followed by the group of (40-49) whose percentage was 33 and the age group over 51 years old which were 15%.

The average size of the pen processes was 39.2 mm with extremes of 26 and 76.6 mm example (Fig. 1)



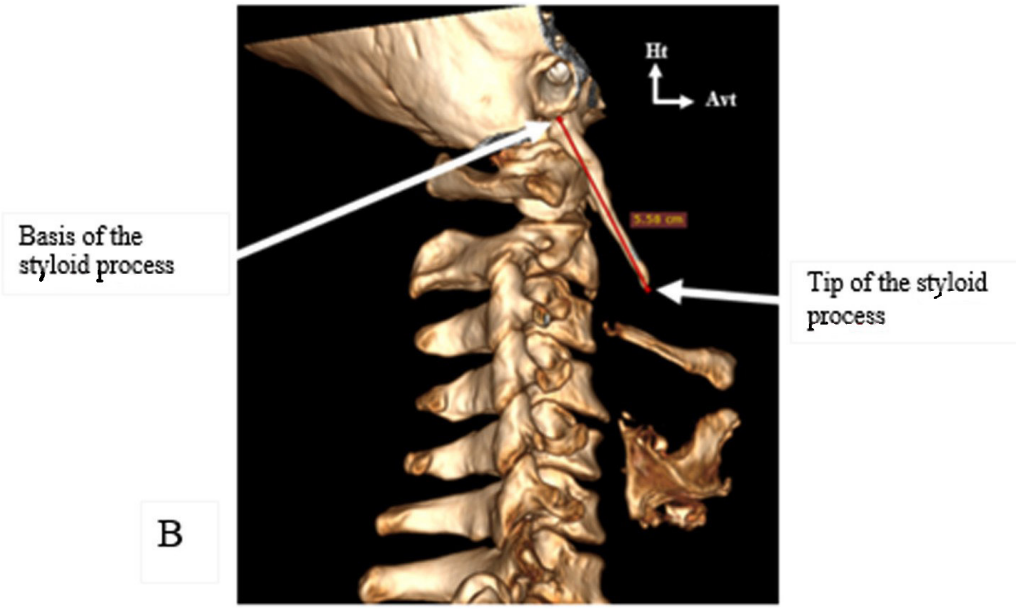


Fig. 1: A; Highlighting of styloid process and surrounding structures, B ; Styloid process measurement method from volume rendering

On the right, the average size was 36 mm with 26 and 71 mm (Fig. 2) extremes and on the left, it was 38 with 26 and 76.6 mm extremes.

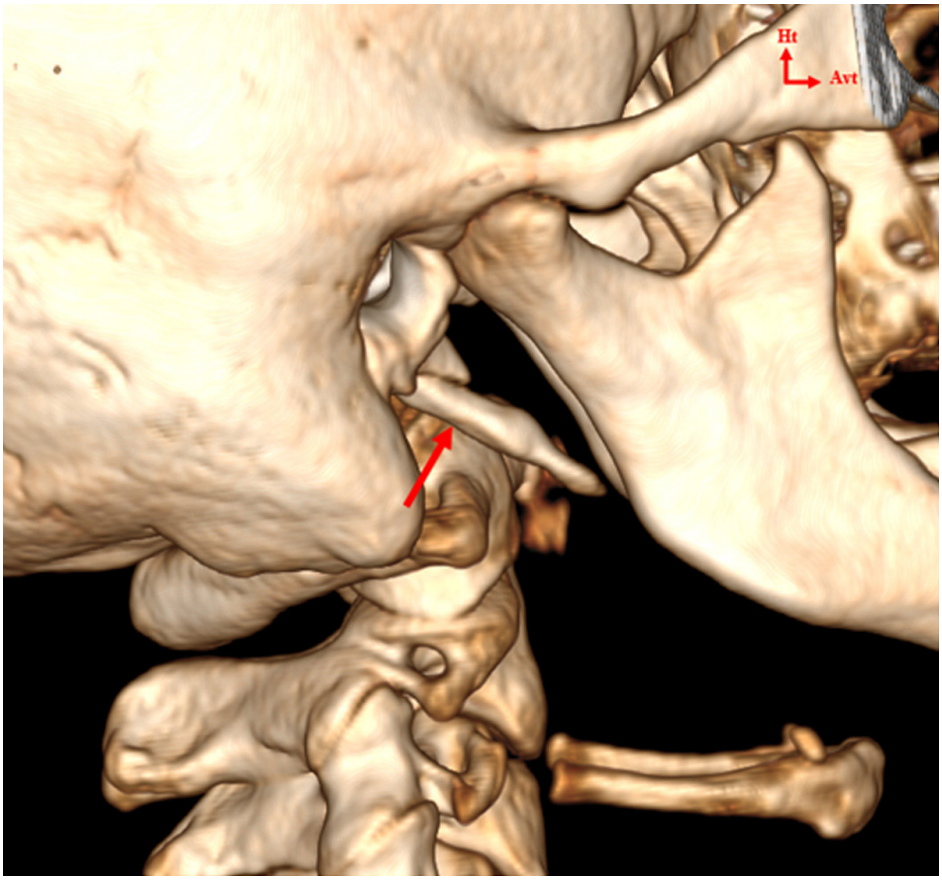


Fig. 2: VRT mode reconstruction of the abnormally elongated right styloid process (red arrow).

Pen processes between 26 and 35 mm in size accounted for 68%, and 32% represented styloid processes strictly greater than 35 mm.

For one of the patients who received surgery following repetitive TIA-type complications secondary to compression of the left internal carotid artery, we took the image of the pen process (Fig. 3).

We found in our population the three types

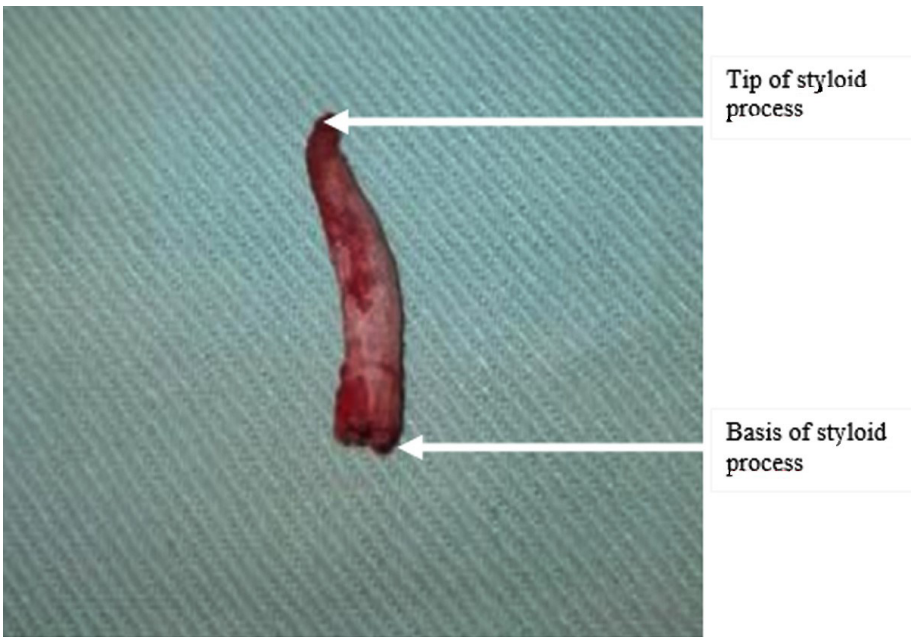


Fig. 3: Photo of a resected styloid process

described by Langlais. Morphologically, the PS consisted of a single long fragment in 78% of cases (Langlais types 1), or two fragments articulated either by fibrous tissue or by a pseudoarticulation with an angular shape in 19% of cases (Langlais types 2; cf Fig. 4), or several fragments (3% Langlais Types 3).

Table I below summarizes the distribution of these types by topography with a predominance of bilaterality which was 68% (Fig. 5).

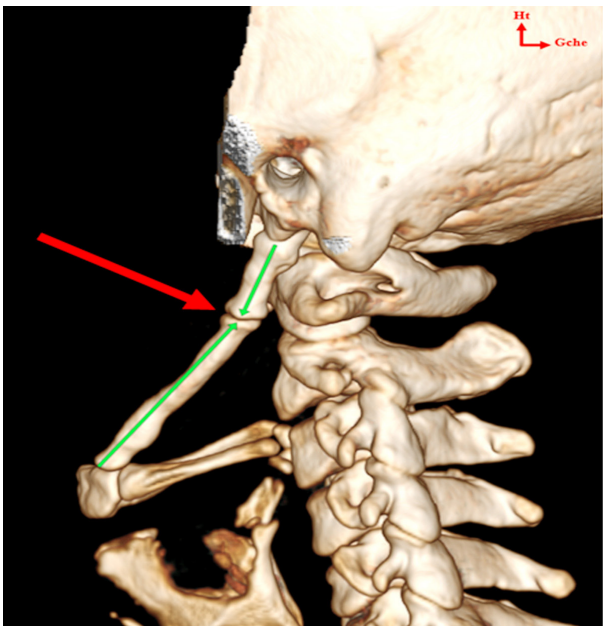


Fig. 4: Reconstruction in VRT mode showing type 2 elongation of the left styloid process (arrow) according to the Langlais classification with the pseudo-articulation (arrow).

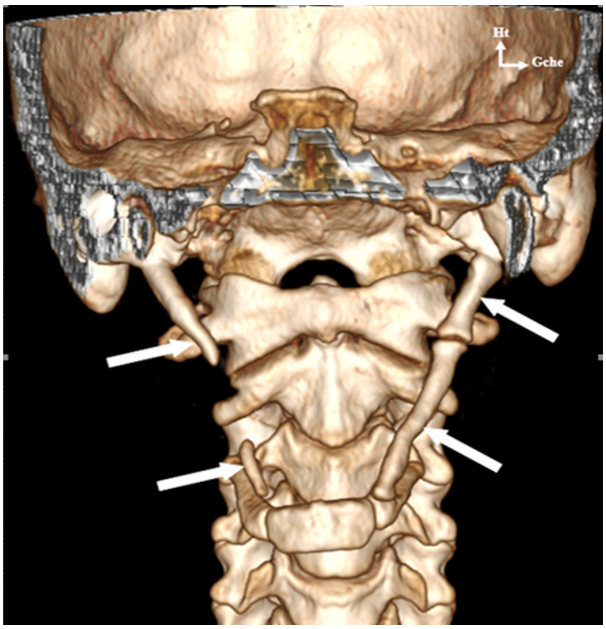


Fig. 5: Reconstruction in VRT mode showing bilateral lengthening of the styloid process (arrows).

Table 1: Distribution of types by topography.

Topography	Types			Total	Percentage (%)
	T1	T2	T3		
Left	4	4	–	8	25
Right	2	–	1	3	7
Bilateral	20	1	–	19	68
Total	26	5	1	32	100

Examination revealed a pharyngeal foreign body sensation in 65% of cases, dysphagia in 67% of cases, headache in 71% of cases and cervico-brachial neuralgia in 95% of cases.

DISCUSSION

In our study, the predominance was female with 75% or a sex ratio of 0.33. Our results are superimposed on those found by El Hassanni, Piagkou et al.[3,13] The female sex seems to be a predisposing factor in the occurrence of the abnormally elongated pen process (SE). There is a relationship between the presence of systemic pathology and (ES).[10] Other authors blame hormonal disorders due to menopause.[14]

This female predominance was not shared by Keur JJ et al.[15] for whom there would be no female or male difference on the occurrence of elongation of the pen process. This may be due to population size. Consideration should also be given to the fact that osteoarthritis occurs at a relatively late age regardless of gender.[16]

In our sample, the average age was 39, so it is close to what is described by certain heights.[3,17,18] Camarda and Al are a little bit ahead of us. They are at 40[17] while Fini et al.[10] had found 37. This observation of the average around the wall age could be explained by the fact that the ossification of the pen process is a continuous phenomenon throughout life and starts late. During youth, pen process is absent or is very short.[19] The study of Al-Khateeb et al.[13] was in phase with ours. This was the case with the results of Camarda et al.[21]

Our work contradicted those of Eagle[20] that criminalize surgery, or trauma to the cervical region, in the occurrence of elongation of the pen process. This is confirmed by the work of Fini et al. who concluded that there is a strong correlation between a history of tonsillectomy and/or trauma with the occurrence of Eagle syndrome.[18] However, no association could be made between the clinic and the degree of elongation of the pen process.[21]

The 3D reconstruction scanner (3 Dimensions) allowed the exact measurement of the pen process and the knowledge of the surrounding structures.[22] It remains a good anatomical study tool in vivo.

In our study the bilaterality of the abnormally elongated pen process was found in 68% of cases. This is superimposed on the results of the literature[10,11] which showed a bilaterality around 85% of cases. Harma et al.[23] found 48%.

When the elongation of the pen process is unilateral (32% of cases), we find a 25% left predominance in contrast to Jung T and all.[24] who found no difference in distribution between right and left. This could be explained by the size of their sample which was 1000 patients.

In our study a pen process was considered pathological, that is to say abnormally elongated when it was greater than 25 mm like some authors.[1,4]

Ceylan[25] agrees with us that a normal pen process should be less than 25 mm. Other authors set a threshold greater than 30 mm.[11,15,21,26,27]

According to Monsour and Young 28, SP is considered an elongated variant when its length exceeds 40 mm, as well as for Savranlar et al.[29]

It should be noted that the majority of elongated pen processes (ESP) are between 30 and 40 mm (54%) on both the right and left sides. The ossification of the pen process is a continuous phenomenon throughout life, and especially very variable: the length of the pen process varies considerably from one individual to another. The underlying phenomenon is unclear but would influence length variability.[12,31]

In our study, type 1 predominated with 78% followed by type 2 with 19% and finally type 3 with 4%. Our results are superimposed on those of EL Hassani and Gossman et al.[3,32] who found a majority of pen processes that were elongated without interruption of their integrity (type 1) in 78% of cases. But unlike them, Type 3 was the least represented with 4% versus 27% among them. Type 2 is not found in their study but only in our case, it was not negligible (19% of cases). In addition, we

noted that there was a type match in our series when a stretching of the pen process is bilateral (78%). It is indeed quite rare to find two different types in the same patient. We did not find studies on the correspondence of types in the literature.

Many vascular complications such as ACI dissection have been described in the literature.[4, 33-37] The same was true of one of our cases with repeated transient ischemic stroke on an anatomical variant of the pen process. This justifies the interest to study the relations of the pen process with the particularly vascular neighbourhood structures.

CONCLUSION

Eagle syndrome is a radioanatomical entity with several variants whose knowledge is essential to its management and understanding of its complications, especially since it was classified in 1988 as "not sufficiently demonstrated", by the International Headache Society.

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