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# A patient with conventional and follicular variant papillary thyroid microcarcinoma and Eagle's syndrome, Langlais type III, with hyoid bone variation

Demet Sengul\*, Ilker Sengul\*\*, Ozkan Ozen\*\*\*

\*Department of Pathology, Giresun University Faculty of Medicine, TR28100 Giresun, Turkey

\*\*Division of Endocrine Surgery, Department of General Surgery, Giresun University Faculty of Medicine, Giresun, Turkey

\*\*\*Department of Radiology, Giresun University Faculty of Medicine, TR28100 Giresun, Turkey

## A patient with conventional and follicular variant papillary thyroid microcarcinoma and Eagle's syndrome, Langlais type III, with hyoid bone variation

**AIM:** Presenting an extraordinary case of Eagle's syndrome (ES) with the classical and follicular variant of papillary thyroid microcarcinoma (PTmC) and an uncommon stylohyoid ligament calcification, aside from O'Carroll's System.

**CASE REPORT:** A 52-year-old Turkish man with an histopathology of the follicular variant of PTmC (FVPTmC) of his right lobe and conventional PTmC of his left lobe of the thyroid with an adjuvant 100 mCi radioactive iodine (RAI) ablation was admitted. On the routine follow-up of the present case, a linear structure at the 3rd compartment of neck in his ultrasonography, a styloid ligament calcification in his noncontrast computed tomography (CT), and a left styloid process of the temporal bone of 29 mm in size in his three-dimensional (3D) CT scanning had been revealed. Conclusively, an ES, Langlais Type III with The Calcification Pattern IV, was recognized.

**DISCUSSION:** A noninvasive peroral medical management was administered as a first-line treatment for the neuropathic sequelae of ES and it has provided relief of signs and symptoms without any recurrence to date, for three years and three months.

**CONCLUSION:** To our knowledge, it is an highly extreme and first case of ES in the English literature, recognized in the course of his programmed follow-up of his conventional PTmC and FVPTmC, simultaneously possessing an exceptional stylohyoid ligament calcification, apart from O'Carroll's System, even with an anatomic variation of the hyoid bone.

**KEY WORDS:** Eagle's Syndrome Follicular variant of papillary thyroid microcarcinoma, Hyoid bone variation, Langlais classification, O'Carroll's System, Radioactive iodine, Papillary thyroid microcarcinoma

## Introduction

The embryologic assessment of the styloid process, stylohyoid ligament, and hyoid bone is a bit controversial. However, Revilla and Stuyt propounded the stylohyoid ligament, styloid process, and also lesser cornu of the hyoid bone being derived from the endochondral ossifi-

cation of Reichert's cartilage from the cartilaginous component of the second branchial arch in the embryologic life. The styloid process is an elongated, fine slender pointed outgrowth rising from the inferior surface of base of the skull, temporal bone, moving downwards and anteriorly toward the maxillo-vertebropharyngeal recess, that contains the carotid arteries, internal jugular vein and VII<sup>th</sup>, IX<sup>th</sup>, X<sup>th</sup> and XII<sup>th</sup> cranial nerves <sup>1</sup>. There is no any concurrence for the prevalence of the Eagle's Syndrome (ES), nonetheless prevalence ranges between 0.04% and 40%. Additionally, the higher percentages of 20% are mostly based on the radiological data, regarding all types of imaging procedures used in discrete courses for achieving the distinct diagnosis. ES has a small propensity for much female prevalence and the styloid process which length >25-30 mm is concluded as unnatural, concerning the radiological research <sup>2</sup>.

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Correspondence to: Prof. Ilker Sengul MD, Professor of General Surgery, The Founder Chairman, Division of Endocrine Surgery, Department of General Surgery, Vice Chair, Department of Surgical Sciences, Giresun University Faculty of Medicine, Gazipasa Compound, Gazi Avenue, TR28100 Giresun, Turkey (E-mail: [ilker.sengul.52@gmail.com](mailto:ilker.sengul.52@gmail.com))

In the present study, it is purposed here in to present extreme case of ES with an uncommon stylohyoid ligament calcsification, aside from O'Carroll's System, depicted with his three-dimensional computed computerized tomography (3D CT) imagings, recognized during his programmed routine postoperative and postRAI follow-up for his disorder of classical and follicular variant of papillary thyroid microcarcinoma (PTmC).

## Case Report

A fifty-two-year-old Turkish man admitted to the Department of General Surgery on May 2016 with an histopathology of follicular variant PTmC (FVPTmC) of his right lobe with two foci, 3 and 6 mm, and conventional PTmC of his left lobe with one focus, 7 mm, who underwent to administration of an adjuvant radioactive iodine (RAI) treatment, a 100 mCi RAI ablation, after his total thyroidectomy<sup>3</sup>. On his neck ultrasonography, A linear calcific echogenity, 5.1 mm in a diameter, at his 3rd compartment of left lateral cervical area was revealed, July 2016. His noncontrast CT imagings of the neck, the images were examined and analysed by using 3D software program, his left styloid process of the temporal bone was measured as 29.0 mm (Fig. 1) and the calcsification in a dense manner of 1.8x4.2 mm were exhibited on his hyoid bone adjacent with right lesser corn (Fig. 2). Both of the lesser and greater corns of hyoid bone were symmetrical and had the same lenght, but the right greater corn had a higher volume regarding the left one. A medical noninvasive treatment had been choosed and administrated to the case and no any recurrence has been detected during his follow-up period of 3 years and 3 months.

## Discussion

An elongation of the styloid process, a symptomatic anomaly, or mineralization of the styloid complex indicates

ES<sup>4</sup>. Langlais et al.<sup>5</sup> had classified styloid process based on the type of elongation and calcification. The elongated styloid process complexes of Langlais classification includes: (1) Type I, elongated; (2) Type II, pseudoarticulated; and (3) Type III, segmented. These types Langlais classification<sup>5</sup> are further described by four pattern of calcification: (1) calcified outline, (2) partially calcified, (3) nodular, and (4) completely calcified<sup>6</sup>. As the morphological classification, the present case was compatible with the "segmented type" styloid process and the "completely calcified type" of classification pattern concerning Langlais et al<sup>5</sup>, 1986 (Langlais Type III with The Calcification Pattern IV). His stylohyoid complex classification was detected apart from all the 12 subtypes of O'Carroll's Classification System<sup>7</sup>, suprisingly. A medical and noninvasive management, peroral medical agents, consisting amitriptyline, gabapentin, valproate, carbamazepime, and image-guided corticosteroid administrations is first-line treatment for the neuropathic sequelae of ES. These non-invasive tratment can provide a temporary relief of signs and symptoms, such as dull and persistent pain in the oropharynx face, and tonsillar fossa that may refer to the ipsilateral ear and get exacerbated by swallowing, yawning and chewing. The surgical interventions such as an intraoral resection of the styloid process and an external resection for the styloid process are considered just after the administration of the noninvasive therapies have failed. The medical noninvasive treatment of choice had been administrated to the patient in the present study and no recurrence has been detected for the 39 months follow-up.

## Conclusion

It is an extraordinary and, to our knowledge, first case in the English literature, of ES revealed concurrent with his routine postoperatif and postRAI planned control of the PTmC with two variants that possessing an uncommon stylohyoid ligament calcsification, aside from O'Carroll's System<sup>7</sup>, which also showed an co-occurrence



Fig. 1: The left styloid process of the temporal bone was measured as 29.0 mm on his noncontrast CT imagings of the neck (*red thin oblique line*) (3D CT imagings by 3D software program).



Fig. 2: The calcsification in a dense manner of 1.8x4.2 mm on his hyoid bone adjacent with the right lesser corn (*white thick mild oblique line*) (3D CT imagings by 3D software program).

with an hyoid bone variation. A medical noninvasive treatment had been administered to the patient in the present study <sup>8</sup> and no evidence of recurrence has been observed during the follow-up of period of 39 months.

## Riassunto

**OBIETTIVO:** Presentare un caso straordinario di sindrome di Eagle (ES) con la variante convenzionale e follicolare del microcarcinoma papillare della tiroide (PTmC) e un raro calcificazione del legamento stiloideo, a parte il sistema di O'Carroll.

**CASO REPORT:** È stato ammesso un uomo Turco di 52 anni con un'istopatologia della variante follicolare di PTmC (FVPTmC) del lobo destro e PTmC convenzionale del lobo sinistro della tiroide con ablazione con iodio radioattivo (RAI) da 100 mCi adiuvante. Sul follow-up di routine del caso in esame, una struttura di lineer al terzo compartimento del collo nella sua ecografia, una calcificazione del legamento stiloideo nella sua tomografia computerizzata non controllata (CT) e un processo stiloideo sinistro dell'osso temporale di 29.0 mm in sono state rivelate le dimensioni nella sua scansione tridimensionale (3D). In conclusione, è stato rivelato un ES, Langlais Type III con The Calcification Pattern IV.

**DISCUSSIONE:** una gestione medica orale non invasiva è stata somministrata come trattamento di prima linea per le sequele neuropatiche di ES e ha fornito sollievo da segni e sintomi senza alcuna recidiva fino ad oggi, per tre anni.

**CONCLUSIONE:** A nostra conoscenza, si tratta di un caso estremamente estremo e primo di ES nella letteratura inglese, riconosciuto nel corso del suo follow-up programmato del suo PTMC e FVPTMC convenzionale, in

possesso simultaneamente di un'eccezionale calcificazione del legamento stiloideo, a parte O'Carroll's Sistema, anche con una variazione anatomica dell'osso ioide.

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