**Keywords:** Autopsy; Death, Sudden; Forensic Pathology; Renal Dialysis; Stylohyoid Ligament Ossification.

## CASE BACKGROUND

A 76-year-old man had visited to his relative's grave and cleaned it up in August 2019. According to his sister, he had been a history of arteriosclerosis and chronic renal failure, and had been receiving hemodialysis three times a week. The next morning, he was found dead around the grave in a supine position. No signs of struggle were found around the body. To determine the cause and manner of death, a forensic autopsy was performed 2 days after the body was found.

His height was 158.0 cm and weight was 46.8 kg. A nephrostomy tube had been placed in his left lumbar region, and there were no other marked external injuries. Internally, there was advanced atherosclerosis with calcification and ulceration in the aorta. In the left kidney (weight 126.1 g), there was advanced calcification, renal calculus, and accumulation of pus, with a nephrostomy tube connected to the outside. The right kidney showed marked atrophy (weight 28.1 g). Histopathologically, progressive glomerulus sclerosis, arterial intimal thickening, and atrophic changes of the tubules with casts were shown. In the neck region, bilateral bony hard structures extended from the lesser cornu of the hyoid bone and continued in a posterior and cranial direction to fuse with the styloid process (SP) at the base of the skull on both sides (Fig. 1); parts of these structures had a small degree of flexibility, histologically consisting of cartilage, bone, bone marrow structures, and bony trabeculae (Fig. 2). Although the bilateral

carotid arteries showed calcification and arteriosclerosis, these structures were not causing stenosis or compression. Because there was extended fibrosis of the myocardium with coronary atherosclerosis in the heart, the cause of death was diagnosed as ischemic heart disease.

## DISCUSSION

The SP, stylohyoid ligament (SL), and lesser cornu of the hyoid bone develop from endochondral ossification of Reichert's cartilage, which is the cartilaginous component of the second branchial arch[1]. The SL is a connective tissue band that originates from the apex of the SP and attaches to the lesser cornu of the hyoid bone. Elongation of the SP or partial ossification of the SL have often been reported; however, complete ossification of the tissue that connects the SP to the lesser cornu of the hyoid bone is rare[2-9]. An elongated SP or ossified SL may contact the adjacent structures, such as the carotid artery, internal jugular vein, facial nerve, glossopharyngeal nerve, vagus nerve, and hypoglossal nerve. Contact with these structures may potentially cause a variety of symptoms including recurrent throat pain, foreign body sensation, dysphagia, and facial pain. This condition is called Eagle’s syndrome, and occurs in 4% of individuals with an elongated SP or ossified SL. Especially, if the ossified SLs compress the carotid sinus complex, decrease of blood pressure, momentary unconsciousness or vagus nerve medicated cardiac inhibition may be occurred[10]. Therefore, when the ossified SLs are found or Eagle’s syndrome is observed before death of the victim, forensic pathologists have to suspect the relationship between these findings and the cause of death.

The three proposed mechanisms for ossification of the SL are: 1) metastatic calcification due to disorders causing abnormal serum Ca and P levels; 2) dystrophic calcification due to mineral deposition into metabolically impaired or dead tissue despite normal serum levels of Ca and P; 3) ectopic ossification with end-stage renal disease (ESRD). Patients with ESRD generally have several disorders that lead to abnormal calcium metabolism, such as renal failure, dialysis, and secondary hyperparathyroidism, which puts them at risk of ectopic calcification or ossification[11-14]. Ectopic calcification or metastatic calcification in nonosseous soft tissue is very common in patients with ESRD. In addition, the SP is significantly longer in patients undergoing kidney transplantation who have been treated with hemodialysis compared with control subjects[15].

In the present case, the SLs were completely ossified and connected to the lesser cornu of the hyoid bone, which is the first such case in a patient undergoing hemodialysis. The patient had chronic renal failure and had been undergoing hemodialysis three times a week for 2 years, but had no other symptoms of Eagle's syndrome. In the present case, it was felt than the ossifications were merely an incidental finding and not related to the cause of death. Moreover, as the presence of ossification is considered to be related to abnormal renal metabolism, the progression of ossification might reflect the degree of abnormality of renal metabolism. In forensic autopsies, if the cadaver is not identified and information regarding previous histories is not obtained, progression of ossification might also aid to predict the health status of the victim before death and to determine the cause of death.

Although SP elongation has been reported in patients undergoing hemodialysis[15-18], the authors

are not aware of a reported case in which the SLs were completely ossified. The present autopsy case may aid in the understanding of the relationship between ossified SLs and elongated SPs and renal metabolic function. In future clinical and autopsy cases, patients with ESRD should be examined for ossification of the SLs, which may provide useful information regarding renal metabolic function and the degree of SPs' elongation.

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**Conflict of Interest:** The authors declare that they have no conflict of interest.

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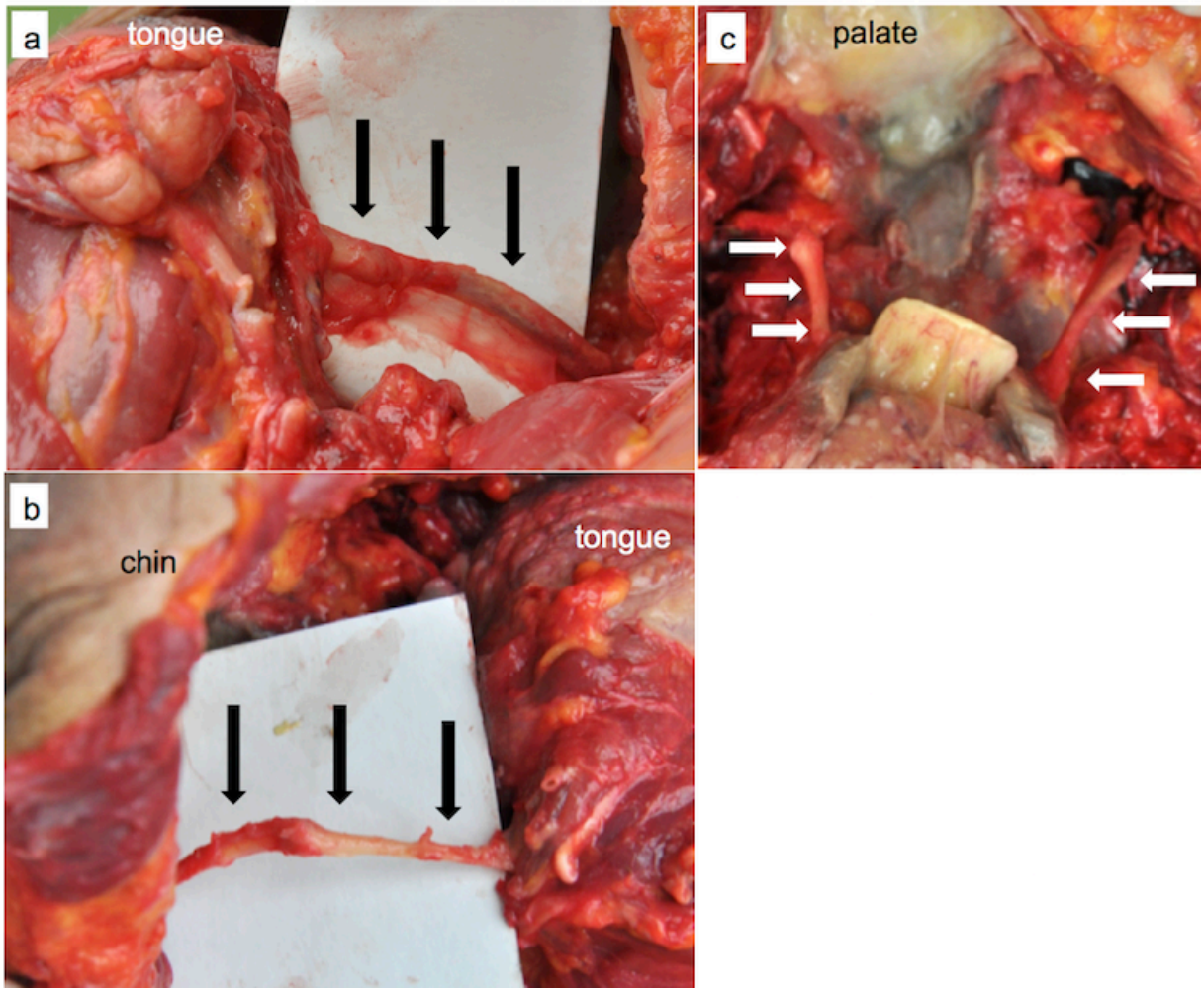
**Figure legends**

Fig. 1: Completely ossified stylohyoid ligaments. a) Left side view, b) right side view, c) frontal view.

Fig. 2: Microscopic findings of the ossified stylohyoid ligaments. The stylohyoid ligaments are mainly composed of (a) bone and (b) bone marrow structures, and covered with periosteum and connective tissue-related film (c) (x40 magnification).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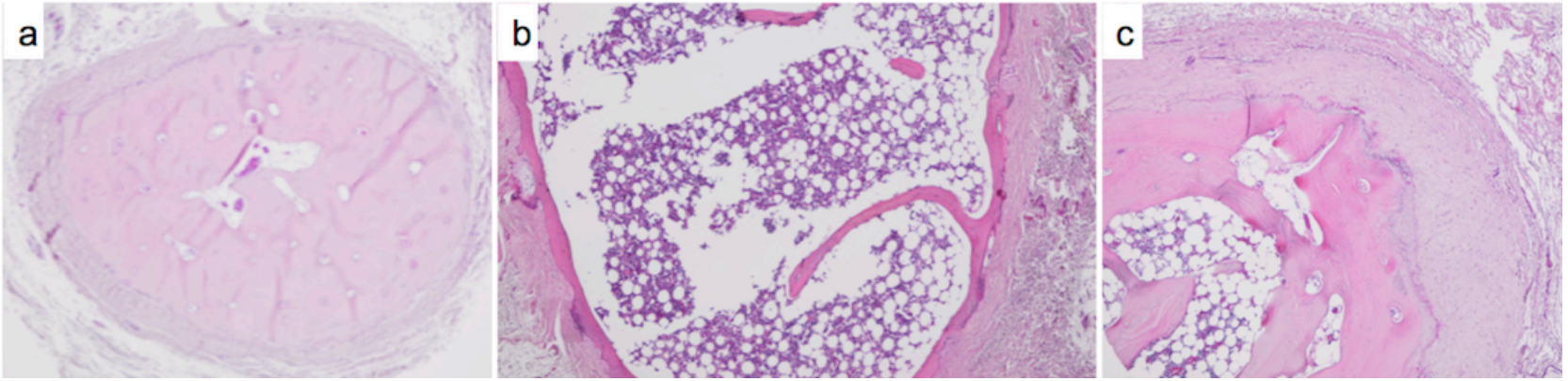
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