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Osteoma of the stylohyoid chain: A rare presentation in a CBCT study

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ABSTRACT

A 54-year-old male patient presented for a periodic check-up at the dental clinic. A panoramic radiograph showed bilateral ossification of the stylohyoid ligament with an oval radiopacity on the right side. Cone-beam computed tomography revealed a well-defined, homogenous hyperdense entity from the lower third of the ossified stylohyoid ligament on the right side. The differential diagnosis of osteoma on the stylohyoid chain includes Eagle syndrome and benign tumors of the stylohyoid chain and adjacent structures. Osteoma rarely manifests in the neck. Even more infrequent are tumors originating from the stylohyoid chain, with only a single documented case of osteoma reported in the literature in 1993. Due to the asymptomatic status, no surgical intervention was advised, and the case would be monitored periodically. This case report describes the details of an osteoma that emerged from the stylohyoid chain, marking it as the second recorded occurrence of this highly rare condition. ***(Imaging Sci Dent 2024; 54: 109-13)***

KEY WORDS: Osteoma, Cone-Beam Computed Tomography, Osteogenesis, Parapharyngeal Space

Osteoma is a primary benign bone tumor that originates in mature membranous bones. It typically exhibits slow progressive growth and is often asymptomatic, although it can cause swelling and pain later in life.[1,2] The tumor is characterized by well-circumscribed mature bone formation on other bones, consisting of compact and/or trabecular lamellar bone.[2] While the exact etiology of osteoma remains unclear, it is thought to be associated with inflammatory, congenital, trauma, or endocrine factors.[3] Osteoma is relatively rare, accounting for approximately 2.9% of all bone tumors and 12.1% of benign bone tumors, with a slightly higher incidence in males.[4] These tumors predominantly occur in the craniofacial skeleton, especially in the paranasal and frontal sinuses, as well as the orbit.[5] They are less commonly found in other craniofacial sites, such as the temporal bone, middle ear, and hyoid bone.[6]

The styloid process is a slender bony projection extending from the inferior surface of the temporal bone. Its length varies from 5 mm to 50 mm, with the average normal length being between 20 and 30 mm.[7,8] The stylohyoid ligament connects the styloid process to the lesser horn of the hyoid bone, albeit at a distance. Together, these structures comprise what is known as the stylohyoid chain, also referred to as the stylohyoid complex or apparatus.[9] The stylohyoid chain can undergo a discrete osteogenic transformation, which involves the elongation of the styloid process and/or the ossification of the stylohyoid ligament.[8,9]

Osteoma of the stylohyoid chain is exceedingly rare, and to our knowledge, only one case has been reported in the Engish-language literature.[10] This paper describes the second case of an osteoma that developed from the stylohyoid chain.

Case Report

A 54-year-old male patient presented for a periodic check-up. His initial visit in 2009 was for a periodontal evaluation. During his most recent exam in 2023, both intraoral and extraoral examinations revealed no clinically significant findings. Panoramic radiographs from the pa-

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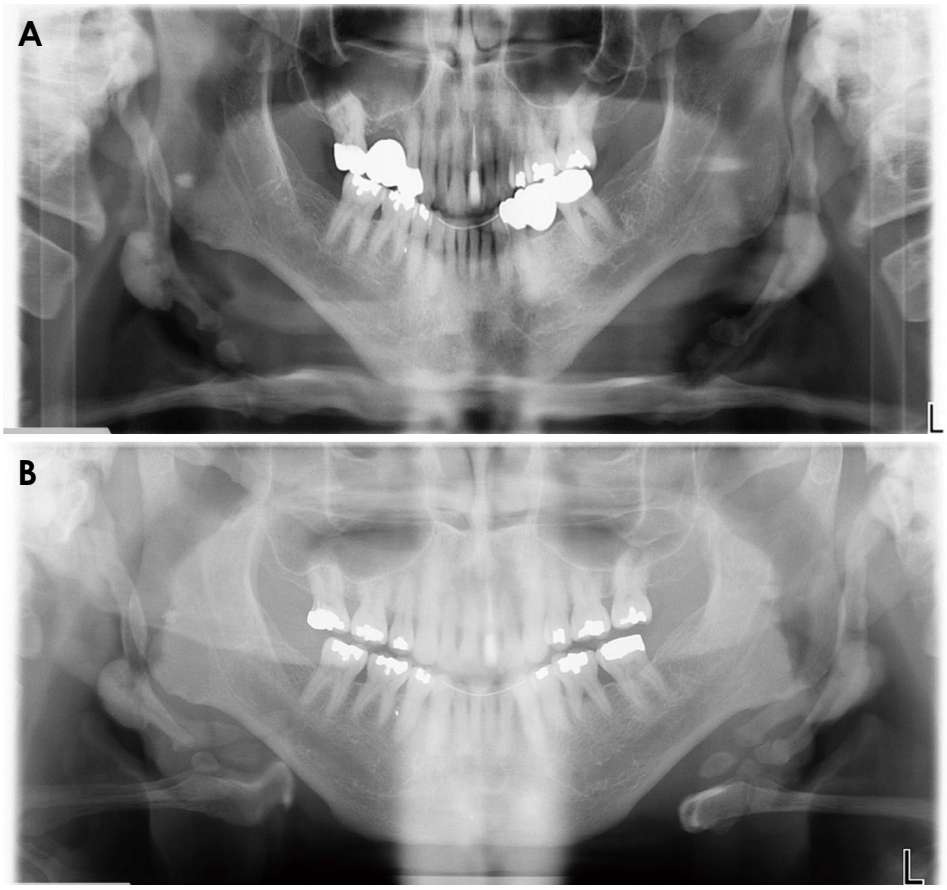

Fig. 1. A. The round radiopacity on the right stylohyoid chain remains unchanged, as shown in a panoramic image taken in 2022. A ghost image of the lesion is observed along the right stylohyoid chain reflected on the left side. B. A panoramic radiograph taken in 2009 shows bilateral ossification of the stylohyoid chain, extending past the angle of the mandible. A well-defined, round radiopacity is shown along the right stylohyoid chain.

tient's first visit in 2009 and the periodic exam in 2022 showed bilateral soft tissue ossification of the stylohyoid chains, extending beyond the angle of the mandible. A well-defined, round, dense radiopacity was observed attached to the right ossified stylohyoid chain at the level of the angle of the mandible, as depicted in the panoramic radiograph from 2022 (Fig. 1A). A comparison with a previous panoramic image from 2009 showed no significant changes in the size or extent of the mass (Fig. 1B). The patient did not report any dysphagia, pain, discomfort, or limited range of motion in the neck.

The patient was referred for a cone-beam computed tomography (CBCT) scan to further evaluate the lesion. The CBCT scan showed ossified stylohyoid chains on both the left and right sides, extending past the mandible toward the hyoid bone, and a well-demarcated, homogeneously ossified oval mass measuring $9.59 \times 11.37 \times 15.51$ mm at its widest points. In the sagittal view, the mass appeared to be broadly attached to the cortical layer of the parent ossified stylohyoid chain (Fig. 2A). The axial and coronal sections revealed a dense, round lesion clearly distinguishable from the radiolucent center of the ossified chain (Fig. 2B, 2C). The ossified mass originated from the lower third of the right ossified stylohyoid chain near the angle of the mandible at the C2 level and extended downward to the C3 level (Fig. 2D). There was no evidence of destructive change or clear invasion into adjacent structures. The CBCT scan also showed no adherence of the mass to the hyoid bone. The left stylohyoid chain exhibited a similar length of ossification, extending below the angle of the mandible, but without the distinct presence of an oval, homogeneous radiopacity along the chain (Fig. 3). Given the radiographic appearance, which was similar to dense cortical bone, and the lesion's indolent nature, a diagnosis of osteoma arising from the right stylohyoid chain was proposed.

No surgical intervention was recommended since the patient was asymptomatic. Consequently, histopathological analysis will be deferred until there is evidence of active growth or the onset of symptoms. The patient was discharged and reassured that monitoring would continue.

Discussion

Osteoma can be classified as compact (ivory), trabecular (spongy), or combined, depending on the bone density and microscopic appearance of the lesion.[1] They are further

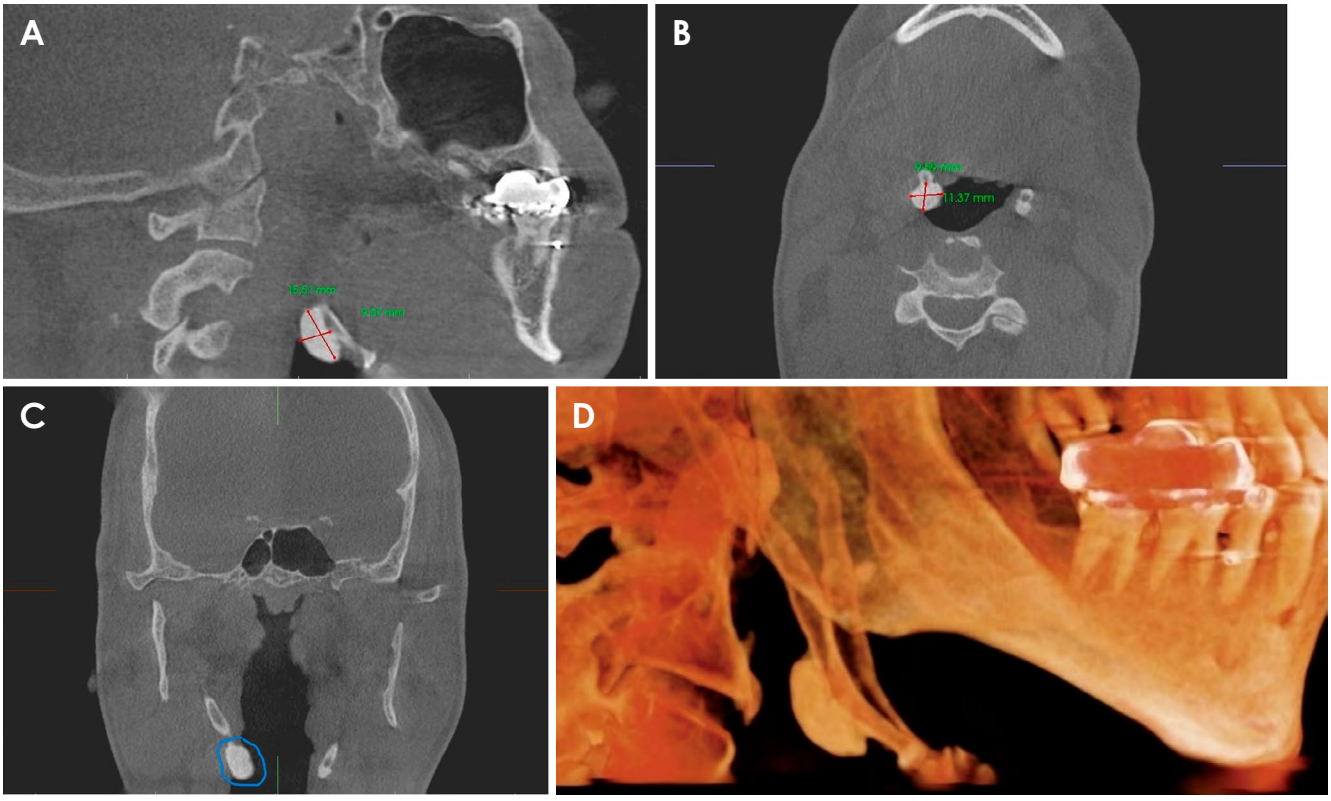

Fig. 2. A. Sagittal section reveals an oval-shaped, hyperdense lesion measuring 9.07 × 15.51 mm along the right stylohyoid chain. The mass is attached to the cortical layer of the ossified ligament. B. Axial cone-beam computed tomography (CBCT) scan shows a homogenously enhanced osseous mass measuring 9.59 × 11.37 mm in diameter. C. Coronal CBCT image displays the osseous mass originating from the right ossified stylohyoid ligament and the ossified left stylohyoid chain without the mass. A homogeneously radiopaque osteoma is clearly distinguished from the radiolucent center of the ossified ligament. D. Three-dimensional cone-beam computed tomographic reconstruction demonstrates bilateral ossification of the stylohyoid chains, with a homogeneously hyperdense mass attached to the right stylohyoid chain between the C2 and C3 levels.

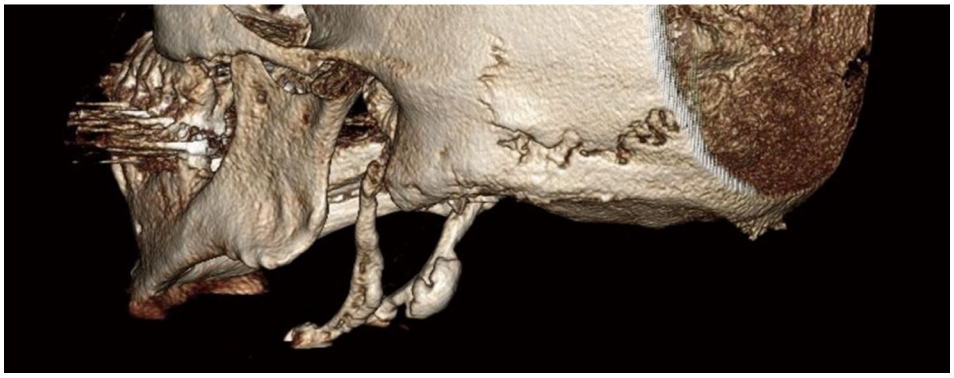

Fig. 3. Three-dimensional volume cone-beam computed tomography rendering image with bone window illustrates the left stylohyoid chain with similar ossification, but no distinct osseous mass arising from it.

categorized by their location of occurrence into three subtypes: central, peripheral, and extra-skeletal.[1] In the present case, the lesion exhibited a highly dense radiopacity without any trabeculation, making it clearly distinguishable from the parent ossified stylohyoid chain. A broad base connected the solitary mass to the outer layer of the ossified stylohyoid chain. CBCT is generally considered the imaging modality of choice for evaluating osseous changes, including osteomas. It provides highly detailed multiplanar images of hard tissues that are as clear as those from traditional CT scans, but with the advantages of lower radiation exposure and cost.[11]

Osteoma originating from the stylohyoid chain is very unusual, and before our report, only 1 case was previously

discussed in the medical literature in 1993.[10] The limited availability of case reports underscores the unique nature of this clinical entity. The first case in 1993 presented a 2-cm-wide osteoma on the middle third of the ossified left stylohyoid ligament.[10] The mass was palpable on examination, unlike our case with a 9.24 mm diameter. Both osteomas arose from the stylohyoid chain at the angle of the mandible; however, the previous case showed an extension of the ossification to the hyoid bone, making the ossified stylohyoid chain further elongated.[10] The ossification did not reach the junction with the hyoid bone in the present case, and no additional bone formation was noted on the hyoid bone. As the present case of osteoma was discovered from the ossified stylohyoid ligament, one might consider this case as a potential representation of Eagle syndrome. However, Eagle syndrome was ruled out because the patient did not display the triad of symptoms of Eagle syndrome: dysphagia, globus pharyngeus, and pain upon swallowing or opening the mouth.[12]

Anomalies in the stylohyoid chain are infrequent. A rare case of an epidermal inclusion cyst from the styloid process was reported in the medical literature in 2009.[13] Unlike osteoma, radiographic images showed a soft tissue mass expanding to nearby structures.[13] Osteochondroma in the stylohyoid chain is also rare and features unique aspects that can be differentiated from osteomas.[14,15] The typical imaging findings are cortical expansion continuous with the medullary cavity of the bone, a cartilage cap at the edge of the lesion, and flaky or linear calcifications inside the lesion.[16]

Benign tumors in the adjoining areas of the stylohyoid complex can provide additional information in the differential diagnosis. Benign tumors arising from the hyoid bone are uncommon, and there have been a few cases of chondroma, osteochondroma, giant cell tumor, and osteoblastoma derived from the hyoid bone.[17-21] A giant cell tumor presents an enlarged soft tissue mass with proliferative signs of bone destruction, often displacing the surrounding structures.[17,18] In chondroma, osteochondroma, and osteoblastoma, multiple spots of calcification in the tumor matrix are a distinct characteristic for identifying these conditions.[16,19-21] About 80% of parapharyngeal space tumors are usually benign, predominantly pleomorphic adenomas in the salivary gland and schwannomas.[22] Similar to the osteoma in our case, symptoms do not manifest until these benign tumors grow.[22]

Due to its indolent nature, surgical excision of an osteoma is generally not advised if the patient is asymptomatic.[2,5] If symptoms are reported but remain subtle and indefinite, non-steroidal anti-inflammatory drugs with neck exercises are advised, followed by a corticosteroid injection to the site of pain.[7,23] Surgical resection is recommended if the patient experiences swelling of the face or neck, facial/cervical neuralgia, or compression of other functional organs, muscles, or blood vessels.[3] A transoral approach usually results in fewer postoperative concerns of esthetics and nerve injury. As an alternative, a cervical approach provides easier access to the lesion and less risk of neurovascular infection.[1,3,7,23,24] The introduction of minimally invasive endoscopy improved treatment success and reduced the risk of postoperative complications for both surgical techniques.[24] Complete removal of the lesion from the cortex of the affected bone is the standard surgical management of osteoma.[1,4] Partial or complete resection of the ossified styloid chain may be performed, depending on the patient's symptoms and potential implications associated with the adjoining anatomical structures.[23,24]

This paper describes a rare case of osteoma originating from an ossified stylohyoid chain, marking only the second such case reported in the English literature. The lesion's virtually stagnant growth over several years suggests that ongoing observation should be the primary management strategy. The presence of asymptomatic bilateral ossification of the stylohyoid chain further supports a conservative approach to managing this condition. The analysis of this case may contribute new diagnostic considerations to the current differential diagnosis for solitary radiopaque lesions involving the stylohyoid complex.

Conflicts of Interest: None

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