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CASE REPORT

Incidental finding: Ossification of the anterior longitudinal ligament of the spine depicted on a panoramic radiograph – A case report with narrative review of the literature

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Abstract

A 55-year-old male had a panoramic radiograph taken at the University of Western Australia Dental School. Radiographic assessment depicted periapical lesions, caries, fractured teeth, horizontal periodontal bone loss, and a distinct ossification anterior to the cervical spine stretching from C2 to C4: (partial) Ossification of the anterior longitudinal ligament (OALL). Differential diagnosis: Osteophytes and segmental OALL. Relevant patient's history: Motor vehicle accident with facial and dentoalveolar trauma. Subsequently, a narrative literature review was conducted to verify the prevalence and etiology of OALL. A PubMed® search was conducted for ossified anterior longitudinal ligament, Forestier disease, and diffuse idiopathic skeletal hyperostosis (DISH). Radiology handbooks were also consulted. It was found that this ossification, first identified as senile ankylosing hyperostosis (Forestier disease), was renamed later: DISH. Its prevalence varies between 2.9 and 25%. The spinal form of DISH is characterized by OALL with involvement of the cervical spine in 76% of patients (typical C4-C5 level). Cervical spine OALL can be associated with dysphagia, obstructive sleep apnea, and pain. Segmental OALL and anterior vertebral osteophytes can be confused with DISH. Its etiology is unknown, but associations have been suggested with advanced age, obesity, and type 2 diabetes mellitus. This case emphasizes the importance to include the assessment of the cervical spine, if visible, on dental and maxillofacial radiographs, as it may prompt more targeted questions toward patients, referral to medical specialists and help diagnose conditions that have been unnoticed or misdiagnosed.

Keywords: Hyperostosis, radiography, spine

Introduction

Part of the cervical spine can sometimes be visible in panoramic radiographs (PR). Although not the primary area of interest in the dental setting, the cervical spine should still be assessed. In the current dental practice, the cervical spine is often overlooked most likely due to dental professionals lacking training to assess it or not examining structures beyond the jaws. PR can project and visualize the cervical spine on the right and left hand side of the radiographic image. Depending on the patient's positioning, the cervical spine is either not visible at all, partially visible or visible from the atlas to third, fourth, or sometimes fifth cervical vertebra. Often only the anterior aspect of the bodies of the vertebrae is visible and therefore the intervertebral spaces can be assessed. Some examples of features that can be distinguished on the cervical spine include heterotopic ossifications of the cervical vertebrae, osteophytes, Schmorl's nodes, cervical Pott's disease, ossification of the posterior longitudinal ligament, ossification of the anterior longitudinal ligament (OALL), ossification of the ligamentum flavum, and alar ligament calcifications.[1-8] The diagnostic yield from a PR with regard to cervical vertebrae conditions is relatively low, compared to cone-beam computed tomography, multislice computed tomography, or magnetic resonance imaging. Nevertheless, if the cervical spine is visible in a PR, assessment of it can assist in incidental findings which may lead to deeper diagnostic investigations that may benefit the patient's health. Obviously, this falls beyond the scope and expertise of a dentist and/or a dental and maxillofacial radiologist and referral to a physician in the first place and subsequently to a medical radiologist if deemed necessary should follow.

Case Report

The current case report is an example of an incidental finding on a PR, at the Oral Health Centre of Western Australia and University of Western Australia Dental School, of a 55-year-old Caucasian male whose medical history was unremarkable [Figure 1]. A motor vehicle accident 6 months before the taking of the PR is evident from the patient's history. The PR was part of a new patient examination to establish a dental treatment plan. A distinct radiopaque entity was appreciated anterior to the second to fourth cervical vertebrae, which did not appear to be attached directly to the vertebral bodies. A deviation of the anterior soft-tissue lining of the spine was observed due to the ossified extension anterior to the spine. The radiographic diagnosis was (partial) OALL of the spine. This incidental finding was asymptomatic and the patient did not report any discomfort besides some occasional stiffness in the neck which he attributed to previous physically demanding labor at his job within the mining industry.

Ligaments of the spine

The cervical spine is attached with the skull by anterior and posterior atlanto-occipital membranes, anterior and posterior longitudinal ligaments, and the ligamentum nuchae. The anterior atlanto-occipital membrane extends from the anterior margin of the foramen magnum toward the anterior arch of the atlas, whereas the posterior atlanto-occipital membrane extends from the posterior margin of the foramen magnum to the posterior arch of the atlas. The anterior longitudinal ligament is attached to the anterior surfaces of the vertebral bodies and stretches from the axis to the sacrum. Superior to the axis, the ligament is continuous with the anterior atlanto-axial ligament. The posterior longitudinal ligament, however, is attached to the posterior aspects of the bodies of the vertebrae (within the vertebral foramen) and also spans from the axis to the sacrum. Superior to the axis, it is continuous with the tectorial membrane, which, respectively, extends from the basilar portion of the occipital bone to the posterior portion of the body of the axis. The ligamentum nuchae, on the other hand, reaches from the external occipital protuberance and the median nuchal line toward the seventh cervical vertebra. It connects the posterior aspects of the seven cervical vertebrae, namely, the posterior tubercle of the atlas, and the spinous processes of the other six cervical vertebrae, respectively. Last but not least, the ligamenta flava connects the axis to first sacral vertebra with each other within the vertebral foramina.[8-11]

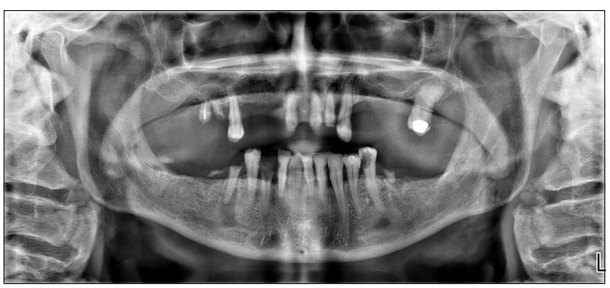

Figure 1: The panoramic radiograph of the 55-year-old male as he presented himself on the initial appointment with the Dental School of the University of Western Australia. Note the presence of an ossified anterior longitudinal ligament of the spine at levels C2 to C4. Besides this incidental finding, it is obvious there are several periapical inflammatory lesions in both jaws due to unfavorable periodontal and endodontic conditions

With exception to the atlas, the vertebrae have a body, a bilateral transverse process and a spinous process. The atlas has the shape of an oval ring (largest in its transverse direction), whereas its body is reduced to an anterior tubercle and the spinous process to a posterior tubercle. The axis is the only vertebra with a vertical extension, the dens axis, or the odontoid process, which articulates in the atlas and between which the following additional ligaments run: The alar ligaments (sometimes called check ligaments as they limit skull rotation) which attach to the dens axis and the medial aspects of the occipital condyles; the apical ligament to the dens which runs from the dens to the anterior margin of the foramen magnum; the cruciate ligaments, consisting of a superior longitudinal band, transverse ligaments of the atlas which pass posterior to the dens axis, and an inferior longitudinal band, which run from the body of the axis to the atlas and the occipital bone; and lateral to the latter runs the deep part of the tectorial membrane obliquely from the body of the axis to the medial surfaces of the transverse processes of the atlas.[8-11] Partial, segmental, or complete ossifications of these ligaments can be visible on panoramic, cephalometric, and cone-beam computed tomography radiographs.

Diffuse idiopathic skeletal hyperostosis (DISH) and OALL of the spine

Ossification of spinal ligaments can occur and hence show up on dental and maxillofacial radiographs. Often, these are incidental findings in otherwise asymptomatic patients. However, they can also be incidental findings in patients with vague complaints or undiagnosed or never taken serious complaints by their physicians.[7,12-14] DISH was first described by Meyer and Forester in 1938 and defined as a hyperostosis and calcification along the right side of the thoracic spine, which they named "moniliform hyperostosis."[15] In 1942, Oppenheimer described ossification of the vertebral ligaments and labeled it "spondylitis ossificans ligamentosa." He emphasized that predominantly the ossification involved the anterior spinal ligaments of the thoracic spine and therefore postulated that the ossification of undifferentiated connective tissue developed secondary to immobilization or decreased mobility of the spine.[15] In 1950, Forestier and Rotés-Querol systematically studied the abnormalities, which they referred to as "senile ankylosing hyperostosis."[15]

In a number of papers published between 1975 and 1978, Resnick *et al.* reviewed the literature and pointed out that

the radiological findings of DISH were always consistent, although the name given to this entity was not, and moreover that the old names predominantly described changes within the spine.[15-17] They described a large number of extraspinal manifestations which occurred in these patients, often without spinal involvement, which subsequently led to the introduction of the more appropriate term of "DISH," emphasizing the generalized nature of this disorder.[6,15,18] As mentioned above, Jacques Forestier and Jaume Rotes-Querol differentiated senile ankylosing vertebral hyperostosis from cervical spondylosis and ankylosing spondylitis and described it as a flowing calcification along the sides of the contiguous vertebrae of the spine, characterized by slowly progressive, ectopic, calcifications that can limit motion, cause stiffness, dysphagia, and dull intermittent pain.[6,19] These ossifications are assumed not to be inflammatory in nature and to be rather a form of degenerative arthritis or osteoarthritis. Nevertheless, the phenomenon has been associated with inflammation and calcification of tendons and ligaments, at their attachment points to the spinal vertebrae bodies (enthuses).[19,20]

Prominent DISH can cause compression of the esophagus or the trachea and cause symptoms such as dysphagia, airways obstruction, and stridor.[21] In most cases, OALL will be asymptomatic, but if not, the following symptoms have been recorded: Pain, neck stiffness, myelopathy, dysphagia, globus, dysphonia, dyspnea, and cough. Hypothesized mechanisms for OALL of the spine (OALL) include mechanical irritation, diet, metabolic conditions (e.g., abnormal bone cell growth activity), and environmental factors. Several associations with DISH have been reported in a myriad of publications: Male, obesity, diabetes mellitus, advanced age, coronary heart disease (incl. atherosclerotic disease), comorbid systemic diseases (e.g., rheumatoid arthritis, osteoarthritis, chondrocalcinosis, and calcium pyrophosphate deposition disease), dyslipidemia, Dupuytren's contracture, hyperuricemia, gout, hypertension, hyperinsulinemia with or without diabetes mellitus, gallstones, obesity, and prolonged use of isoretinol and Vitamin A derivatives.[12,15,18,19,22] Routine chest radiographs taken in a population of 436 patients with severe cardiovascular disease (coronary bypass surgery, valve replacement, or congestive heart failure) disclosed DISH (Resnick's definition) in 30.3% of cases.[23] It is also shown that OALL can in some patients compromise and jeopardize placement of an endotracheal tube for general anesthesia.[24]

A combination of DISH and cervical OALL is one of the most common causes of dysphagia requiring surgical management and DISH-related dysphagia is estimated to occur in 0.6–28% of DISH cases, according to different authors.[7,15,20] The spinal form of DISH is characterized by OALL with involvement of the cervical spine in 76% of the patients.[6] One can find OALL in the cervical level of the spine most often at levels C4 through C6 and at thoracic levels T8-T9.[6] In 2017, two independent studies stated that spinal ankylosis with DISH increases the risk of spinal fractures 4 times and that patients treated for discopathy show OALL in 1.5–2.5% of cases.[20,25] DISH thus increases an individual's susceptibility to spinal injuries. The complication and mortality rates for spinal fractures are significantly higher in patients with DISH than in patients with a normal spine.[13,25,26] A Japanese study on 285 patients with a mean age of 75 years showed that the diagnosis of a spinal fracture due to a fall in these patients was delayed in 40.4%. However, they did not find any correlation with OALL, but there was one with ossification of the posterior longitudinal ligament of the spine.[26] DISH, generally known as Forestier's disease and Forestier-Rotes-Querol disease, is mentioned in the literature also under different synonyms: Generalized juxta-articular ossification of vertebral ligaments, physiologic vertebral ligamentous calcification, senile ankylosing hyperostosis of the spine, spondylosis deformans, spondylosis hyperostotica, spondylitis ossificans ligamentosa, and vertebral osteophytosis.[18,19] Extraspinal or peripheral manifestations of DISH include the following sites: Elbow, foot, heel, wrist, lower extremity, patella, pelvis, shoulders, skull, and tendon appearance.[18]

The prevalence of DISH is thought to be 2.9–42% of the population, although age, ethnic, and racial differences exist. The prevalence of DISH in males over 50 years of age was reported to be 25% whereas for females, it was only 15%. The prevalence increased even to 28% in males over 80 years of age and to over 35% in males over 70 years of age according to a 1997 study.[15] DISH is less common in African, Native American, and Asian populations according to the same authors. Interestingly, they stated that the prevalence of DISH is far lower in patients with osteoporosis. The latter study was done in Minneapolis, USA, on 2364 subjects aged 50 or older. A Korean study on 3595 subjects over 50 years old reported a prevalence of 2.9–4.1%, depending on the selection criteria for DISH. They also mentioned an increase of the prevalence of DISH with age and a 7 times higher chance for males to contract the disorder. The lower incidence in Koreans compared to Caucasians confirms other reports. The prevalence can be summed up in descending order as: Caucasians, Japanese, Pima Indians, African Blacks, Jews in Jerusalem, and American Blacks.[6,13,15,16,25,27] Approximately 10% of the population of over 50 years of age has DISH characteristics, and a 1996 publication stated that between 2.4 and 5.4% of the population over 40 years of age has Forestier's disease.[22,28,29] A 1978 report on Pima Indians found that vertebral ankylosing hyperostosis or Forestier's disease was observed in approximately 50% of the population over 55 years of age.[29]

Differential diagnoses

Over the years, several authors have tried to fine-tune the selection criteria for DISH. According to Forestier, at least two and according to Resnick at least three contiguous intervertebral bridges should be involved for the diagnosis of DISH. A diagnosis of hyperostosis may be considered in patients with peripheral enthesophytes, even in the absence of vertebral bridges. Given that ossification of the longitudinal ligaments may produce changes similar to those seen in spondyloarthritis, the diagnosis of DISH also requires the absence of sacroiliac

sclerosis or fusion and facet apophyseal joint fusion or ankylosis. The enthesophytes seen in DISH must be differentiated from osteophytes due to osteoarthritis and from syndesmophytes due to spondyloarthritis.[16] Osteoarthritis also occurs on weight-bearing joints whereas DISH is not, however, DISH may contribute to the development of osteoarthritis or aggravate primary osteoarthritis.[18] The absence of a consensus about the definition of the disease is a major obstacle to determine its prevalence in epidemiological or paleopathological studies.[16] The radiographic appearance of segmental OALL can mimic that of anterior osteophyte formation which is associated with degenerative joint disease.[6,29] Anterior cervical osteophytes are a common finding in the elderly and when present are most common at levels C4 through C7, while above C4 level they are not. One also has to distinguish anterior osteophytes from DISH, ankylosing spondylitis, degenerative joint disease, and trauma.[3] Identification of calcifications or ossifications along the anterior to anterolateral aspect of four contiguous vertebral bodies with preservation of the intervertebral distance makes it distinct from degenerative discogenic disease. OALL does not show decrease in distance between the respective affected vertebrae.[6,15,16]

Conclusion

Proper assessment of the cervical spine on dental PR, if visible of course, should be part of the radiographic assessment. Observations about the spine should be recorded and in case of doubt patients should be referred to a physician and/or a medical radiologist for further investigation and follow-up. OALL of the spine can be part of DISH and it may or may not be related to physical complaints. Dentists and dental and maxillofacial radiologists should be aware of these conditions and should be able to refer patients to the appropriate medical specialist.

Statement of Clinical Relevance

Dental professionals should be able to identify all landmarks and structures on their radiographs. Identification of OALL of the spine is one of them, which may prompt specific targeted questions regarding swallowing, and neck pain for instance.

Declaration of Patient Consent

The patients who attend our dental school sign a waiver that allows us to use their data for scientific and publication purposes, if they are deidentified. This paper does not hold any identifiable data.

Human Rights Statement

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1964 and later versions. Informed consent was obtained from all patients for being included in the study.

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