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Varied presentations of a mineralized styloid process in oral submucous fibrosis patients visualized on digital panoramic radiographs

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ABSTRACT

Background: Oral Submucous Fibrosis (OSMF) is a widely prevalent, premalignant condition which adversely affects the quality of life of an individual and can warrant stringent surgical regimen for its treatment. An Elongated Styloid Process (ESP), if coexisting, can invariably impair the post-surgical mandatory physiotherapy necessary to maintain the mouth-opening achieved intraoperatively.

Aims: To evaluate digital Orthopantomograms (OPGs) of patients having OSMF and to observe presentations of various combinations of calcification types and patterns of ESP in them.

Settings and Design: Patients with clinically diagnosed OSMF (Grade III and IV according to Khanna and Andrade 1995) were advised a digital OPG to measure and observe the ESP using the C.S. Imaging Software 7.0.3.

Materials and Methods: The digitized OPGs were converted into KODAK large format prints and then ESP was measured starting from the point where it is visualized to leave the tympanic plate extending upto its tip and its myriad of combinations of calcification patterns were recorded by a single observer.

Results: The presence of unilateral and bilateral elongation along with the type and pattern of calcification was effectively noted using the C.S. Imaging Software 7.0.3. Visualization of various unique, varying patterns of ESP was noted. Sixteen such different radiological presentations were shown, captured through digital OPGs.

Conclusion: An undetected ESP can prove to be detrimental for a patient who is recently treated for OSMF as it hinders efficient postoperative mouth opening exercises. This can easily be prevented by a simple maneuver of identifying the ESP on a digital OPG using computer aided software. The presence of assorted radiographic patterns in patients having OSMF indicate that extensive research should be directed to identify and categorize such patterns which do not conform to the past methods of classification. This study hints towards the existence of a scope to study the factors responsible for the occurrence of the varied combinations of calcification and thickness of the ESP and their clinical relevance.

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1. Introduction

With approximately 600 million people around the world actively engaged in the daily consumption of betel nut in its various formations, it is not surprising that betel-nut is the

fourth most consumed addictive substance in the world.¹ Owing to the continuous contact, about 15 minutes to 1 hour, of the mixture with oral mucosa, the alkaloids and flavonoids become a source of constant irritation leading to chronic inflammation as the habit becomes persistent.² In conjunction to this, arecaidine and tannin present in

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the betel-nut mixture increase the cross-linking of the fibres and obstruct collagenase activity leading to fibrosis and accompanying trismus (37.2- 90.8%), predominantly seen across the South Asian population in form of Oral Submucous Fibrosis (OSMF).²⁻⁵

Alarming, these subjects are younger, have shorter history of usage of tobacco products and the disease process does not reverse on cessation of habit but becomes gradually progressive adversely affecting the Quality of Life of the individual.⁶ Chewing betel quid containing tobacco is a prime risk factor for occurrence of oral carcinoma, which is reflected by the fact that a patient having OSMF is 19.1 times more likely to be diagnosed with oral malignancy than the normal counterpart which warrants early and prompt diagnosis followed by medical or surgical therapy.^{1,7-9}

In addition to the active surgical management employed for OSMF, it has to be inevitably supplemented with rigorous physiotherapy which includes various muscle stretching exercises to augment mouth opening and failure to comply with this, often painful and rigid regimen, can unavoidably lead to recurrence of severe trismus.

Negligence towards physiotherapy may be precipitated by the presence of an Elongated Styloid Process (ESP) amongst other probable causes. The embryonic origin of the styloid process, which normally continues to lengthen till 30 years of life, can exhibit mineralization at a much later stage at its insertion to the lesser cornu of hyoid bone due to chronic inflammation from constant mucosal trauma. This may lead to vague facial pain on swallowing and turning the head, in turn ensuring total omission of postoperative physiotherapy in a patient already treated for intraoral fibrosis. The aim of this paper was to identify the presence and morphology of ESP specifically in patients having OSMF based on this hypothesis.^{10,11}

2. Materials and Methods

2.1. Procedure for procuring an orthopantomogram

The prospective study was carried out in the Department of Oral and Maxillofacial Surgery wherein prior written consent was taken from all patients in whom complete intraoral examination was not feasible due to trismus caused by OSMF. Patients were then advised a digital orthopantomogram (OPG) solely for dentoalveolar purpose as detailed clinical intraoral examination was not possible due to trismus. The field of accommodation was widened to include the styloid processes by shifting the bilateral focus lines of the digital OPG machine (Kodak 8000C Digital Panoramic System, Carestream Health, Inc., Rochester New York, United States). An ESP was detected by measuring its length from the point where it leaves the tympanic plate of the temporal bone to its mineralised tip using the C.S. Imaging Software 7.0.3. (Carestream Health, Inc, 2010, Rochester New York, United States

2.2. Inclusion criteria

1. Patients giving positive history of oral destructive habits using plethora of tobacco products.
2. Patients suffering from clinically proven OSMF.
3. Patients with Mouth Opening less than 10 mm due to OSMF. (Grade 4A and 4B according to Khanna and Andrade classification 1995).¹²

2.3. Exclusion criteria

1. Patients with trismus due to causes other than OSMF.
2. Evident ulceration and presence of significant findings on lymph node examination or any other sign of malignancy.

2.4. Visualization of styloid process length and ossification/calcification

The calcification and measurement of the elongation was carried out with help of standardized digital OPG at first presentation of the patient in the out-patient clinic. The OPG obtained were digitized as DICOM files to be later converted into KODAK large format files and stored in a computer.

2.5. Standard length measurement procedure

1. First open the C.S. Imaging Software 7.0.3 from the Desktop and choose a new patient card
2. Insert the details of the patient and click on Import image option
3. Choose the KODAK large format file and click on Open
4. Once the digital OPG is displayed, select the Measurements option from the side-toolbar
5. Choose the desired color from the options given and click on the point from where the styloid process is visualised to leave the tympanic plate of the temporal bone (emergence point)
6. Continue directing the arrow downwards along the path of the styloid process and click again at its mineralised tip
7. The calculated length is instantly displayed along its length and within the chosen color box.
8. Repeat the measurements in a similar manner for the contralateral side and note it.
9. The results of each patient were then compiled in a Microsoft Excel worksheet (Microsoft Excel 2013, Microsoft Corporation, USA) and relevant observations were deduced. All measurements were accomplished by a single, experienced observer and data collector to prevent bias. Fifty OPG were randomly selected and measurements were repeated twice with a month interval. Deviations of the mean length of styloid process between the two recordings were less than 2%.

3. Results

The application of C.S. Imaging Software 7.0.3 for the evaluation of the ossification and length of the styloid process on digital 274 OPG images was asserted to be suitable and satisfactory. The presence of unilateral as well as bilateral elongation was noted specifically in addition to the demographic data and distinct radiographic images of 16 patients. The radiographs were segregated into different groups by comparison of both styloid process on basis of length, calcification pattern, thickness and presence of curvatures. Figure 1 depicts the OPG of a patient having bilaterally symmetrical ESP. Figure 2 depicts OPG showing unilateral elongation on the right side. Figure 3 reviews the OPG of patients showing varied calcification types on either sides. Figure 4 A,B and C encompasses the radiographs showing disparity or similarity in the thickness of the bilateral styloid processes. Figure 5 and Figure 6 displays images where there exists different angulations noted at specific points throughout the styloid process.

4. Discussion

OPG has by and large dominated the field of maxillofacial radiography since its evolution in the mid-twentieth century. It offers an unobstructed view of an otherwise anatomically complex maxillofacial skeleton and is the most routinely used radiographic modality to accurately examine the upper and lower jaws in the clinical practice.¹³ The simplicity of technique, the broad area of visualisation, reduced superimposition, low radiation exposure on its application and convenient storage compel maxillofacial surgeons to include it as a mandatory preoperative examination tool.¹⁴ In spite of vast selections of radiographic methods used to observe the styloid process such as anteroposterior skull radiograph, Towne's view radiograph, Lateral skull radiograph, OPG was chosen as been majorly employed by various researchers due to its numerous advantages.^{10,15-17}

Over the years clinicians have noted that radiographs when used to calculate dimensions must be accurately reproducible. The advent of a digital image analysis system ushered an era of minimal inaccuracy with the two-dimensional radiographic technique such as OPG, as it provided results which were 20 times more precise than the human eye.¹⁸ Comparative studies have concluded that OPG can significantly refine the diagnostic quality in terms of radiographic density and contrast.¹⁹ Furthermore, the evident benefits of speedy communication of images in an interdisciplinary approach, small storage requirements, negligible harm to the environment and comparatively lower radiation exposure to the patient as well as the radiologists has cemented the usage of digital panoramic radiographs in maxillofacial practice.²⁰ The radiation dose exposure of 5-14 μSv is remarkably acceptable as compared to that of conventional radiography which is around 16-21 μSv .²¹

As a two-dimensional radiography such as an OPG conveys a virtual image of a three-dimensional entity, it is understandably flawed to a certain extent. Keur et al. asserted that the projection of the head of the patient will affect the linear distortion and while manually calculating the length of the styloid process, the estimated error of measurement was ± 2.5 mm. The amount of enlargement of the radiographic image in the vicinity of the styloid process was considered to be 11%. Hence it became imperative to utilise digital panoramic analysis using the C. S. Imaging Software to precisely monitor the exact length as well as the degree of calcification of the ESP in the present study. It goes without saying that in symptomatic patients the ideal imaging technique to be selected must be a 3-dimensional Computed Tomography which can definitively delineate the orientation of the medially directed styloid process along with its length.^{16,22} As the present study included preoperative patients who were advised OPG only as part of their dentolaveolar examination due to trismus, 3-dimensional radiography, to visualise the entire morphology of Styloid process, was reserved only for symptomatic patients having pain on swallowing, throat pain, etcetera.

Although a number of probable explanations such as result of reactive metaplasia or hyperplasia, anatomic anomaly or loss of elasticity post-trauma are put forward to explain the occurrence of ESP, none have been conclusively established.^{16,23} Styloid process elongation can be consequent to the lifestyle as well as dietary consumption of the population. specifically in relation to the craniomandibular complex for patients having oral destructive habits of gutka chewing or arecanut consumption, the strenuous exercises on the jaw musculature augments the masticatory load on the ligament thereby prompting its ossification.^{23,24} This phenomena explains the increased prevalence of bilateral elongation in few reports^{16,23} similar to the one observed in this study [Figure 1] whereas the presence of ESP on the right side in case of unilateral elongation in the present study [Figure 2] can be due to habit of unilateral chewing of oral mucosal irritant products similarly as observed by previous studies.^{23,24} Bilateral calcification may also be accompanied by hoarseness of voice, bilateral tinnitus or globus hystericus.^{25,26}

As far as the distribution of the type of elongation and pattern of calcification of the styloid process, as described by Langlais et al.,¹⁵ is concerned, various researches collectively portray Type I i.e. Elongated as the most common type of elongation whereas Pattern B i.e Partially calcified as the most prevalent pattern of calcification.^{16,23,27,28} However, there is utter lack of information regarding the presence of diverse patterns of calcifications present within a single individual or even multiple presentations of patterns of calcification in a single styloid process, which is noteworthy. Figure 3 A portrays an

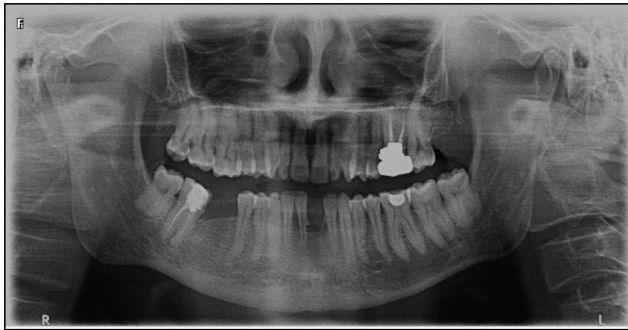


Fig. 1: Bilaterally symmetrically elongated styloid process of a 35 years old male patient



Fig. 2: Unilateral (Right) elongation of styloid Process a 32 years old male patient

OPG of a single individual showing a completely calcified styloid process on the left side whereas the right styloid process displays complete calcification at the base and calcified outline pattern at the tip. Similarly, Figure 3 B shows the OPG of a single individual with completely calcified left styloid process and a partially calcified styloid process on the right side. Similarly Figure 3 C and 3D display OPG of single individuals displaying different calcification patterns on either side. This observation begets discussion on factors governing the variation of patterns in a single individual as well as in a single styloid process.

Sufficient reports have been published to elaborate on the magnitude as well as the cause of styloid process elongation. However there seems to be a dearth of literature on the incidence of increased thickness of the styloid processes as well as its etiology and clinical implications. Regarding the calcification of the soft tissues, the ossification of the stylohyoid ligament results in the elongation of the styloid process due to its connection with the lesser horn of the hyoid bone. However, the calcification of the soft tissues surrounding the styloid process causes an increase in its thickness as seen in Figure 3A and Figure 4A.^{26,27} On the other hand, the formation of thin bilateral styloid processes (Figure 4 B, C) may be due to lesser advancement of stylohyoid ligament ossification,

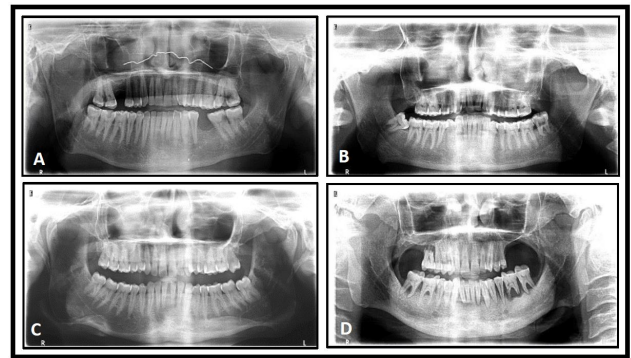


Fig. 3: **A:** Left styloid processes showing a completely calcified pattern and a complete calcification at the base and calcified outline pattern at the tip seen on Right side; **B:** Left styloid process with a completely calcified pattern and Right Styloid process showing a partially calcified pattern; **C:** Right styloid process shows partial calcification at the base with a calcified outline tip and Left styloid process shows complete calcification at the base and tip with a calcified outline pattern seen at the body; **D:** Right styloid process shows complete calcification and is angulated whereas the Left styloid process is straight with nodular pattern

generally in the upper part of the ligament, resulting in a single calcified line.²⁹ Figure 4C depicts a typical presentation of Sakhdari Type 4 Elongation pattern seen as a result of distant ossification.²⁷

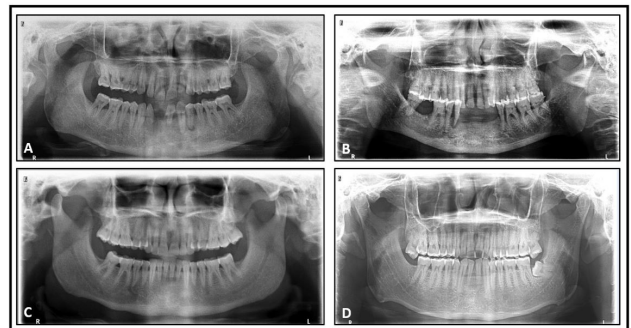


Fig. 4: **A:** Left styloid process with increased thickness at base with gradual thinning towards the tip in comparison to the right process which appears to be thin throughout; **B:** Bilaterally completely calcified thinned styloid processes; **C:** Bilaterally thinned elongated styloid processes due to distant ossification (Sakhdari Type 4 elongation pattern)

Irregular and abnormal ossification of the constituents of the stylohyoid chain may result in various peculiarities along the contour of the ESP.²⁶ The ESP may be straight or exhibit curvature at different locations. Patients may compose of similar variations on either side with a straight ESP on one side and a curved process on the other side. Furthermore, the location of this curve may vary with some showing single curve at the middle region of the

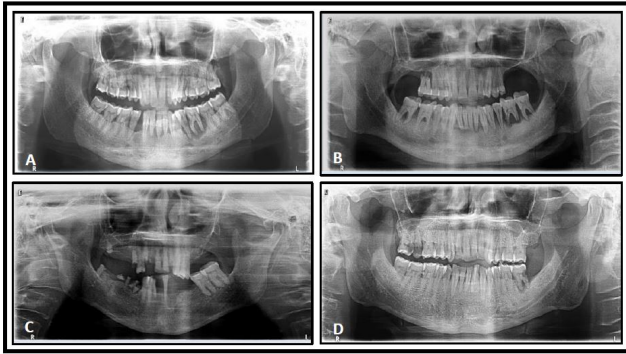


Fig. 5: **A:** Styloid process (Left) showing single curvature (arrow) at the middle region; **B:** Styloid process (Right) showing single curvature (arrow) nearing its tip; **C:** Styloid Process (Right) showing multiple curvatures (arrows showing dual curvatures); **D:** Styloid process (Right) showing multiple curvatures (arrows showing triple curvatures)

process [Figure 5 A] whereas other processes displaying single curvature nearing its tip [Figure 5 B]. The number of curvatures also show multiple presentations as few patients presented with multiple curvatures along the entire length of the ESP [Figures 1, 5 and 6]. Figure 6 shows single patient showing an angulated styloid process on the right side with no curvatures seen on the left side. Variations among the degree of curvature were also evident with few OPG revealing a sharp lateral or medial curvature on either side with a straight styloid process on the contralateral side. The limitation of this study is the incorporation of only OPG as the imaging modality, which cannot project the definite medial or anterior angulation of styloid process responsible for the multiple symptoms associated with Eagle's Syndrome.



Fig. 6: Styloid (Right) process showing single curvature (arrow) and straight styloid process on left side

This study attempted to highlight certain unique radiological presentations, such as presence of bilaterally symmetrical styloid process in an individual [Figure 1] and presence of variations in thickness and angulation of the ESP with the assumption that many such combinations of



Fig. 7: Styloid (Right) process showing sharp lateral curvature (arrow) and straight styloid process on left side



Fig. 8: Styloid (Right) process showing sharp medial curvature (arrow) and straight styloid process on left side

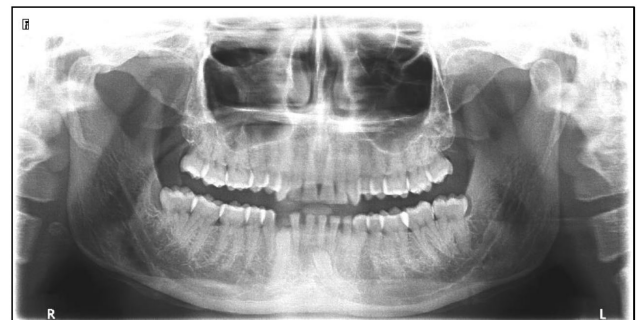


Fig. 9: Elongated styloid process (Right) showing sharp medial curvature

patterns, as per the present system of categorisation as given by Langlais, must be existing and thus are needed to be detected over a larger sample size so as to propose a broader classification including these categorizations. Such diverse combinations might indicate the need to re-evaluate the present criteria of segregating the ESP, with an important inclusion of the width/angulation of the ESP. Subsequent co-relation of the radiological images with the patient's clinical symptoms might be instrumental in establishing vital consensus regarding the etio-pathogenesis of this enigmatic condition.

The operative confirmation of the presence of ESP could not be achieved as this is a preoperative observational

study solely focused on establishing a plausible correlation between the concomitant occurrence of OSMF and ESP in the same individuals. Such an interconnection has been previously conclusively stated in a single research paper which highlighted a positive rate of 31.4% of patients with OSMF exhibiting an ESP as opposed to 2-4% of the general population.¹¹ This coupled with our observations signals an absolute need to stringently screen each and every patient of OSMF using a simple OPG so as to obviate any postoperative complication in the form of hindrance to physiotherapy due to overlapping pain of an ESP.

5. Conclusion

The versatility of a digital OPG can be effectively harnessed to provide a fairly accurate idea about the length as well as the calcification pattern of the styloid process instead of opting for more invasive and expensive modalities such as Computed Tomography and Magnetic Resonance Imaging. This preoperative protocol accepted by any oral radiologist or surgeon should be invariably followed in a patient having OSMF so as to recognize any probable postoperative impediment by the ESP and can be simultaneously used to treat both conditions in a single-stage procedure to prevent the recurrence of fibrosis.

6. Source of Funding

None.

7. Conflict of Interest

The authors declare that there is no conflict of interest.

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