Evaluation of Elongated Styloid Process in Patients with Oral Submucous Fibrosis Using Panoramic Radiographs

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Abstract

Introduction Oral submucous fibrosis (OSF) is an insidious disease affecting the oral cavity, pharynx, and upper digestive tract. It is characterized by a juxtaepithelial inflammatory reaction followed by fibroelastic change in the lamina propria and associated epithelial atrophy. Higher levels of TGF- β present in patients with OSF could be responsible for impetus to remnants of Reichert's cartilage present in styloid complex leading to partial or complete ossification of associated ligaments. So, a study was conducted to evaluate the elongation of the styloid process in patients with OSF by using panoramic radiographs.

Materials and Methods Panoramic radiographs of patients with OSF were studied from 2007–2011. The apparent lengths of styloid process were measured with the help of divider and steel metric ruler. The length of the styloid process and/or ossification of stylomandibular ligaments which were longer than 30 mm were considered.

Results Out of 47 patients, 35 patients (34 males & 1 female) met the inclusion criteria. Eleven patients (31.4%) were found to have elongated styloid processes which included 10 male patients and 1 female patient.

Conclusion It had been estimated that between 2 % and 4% of the general population presents radiographic evidence of an ossified portion of the styloid complex. The high incidence of elongation of styloid process (31.4 %) in patients with oral submucous fibrosis highlights that

progressive OSF might have some influence on elongation of styloid process.

Keywords Oral submucous fibrosis · Elongated styloid process · Ossification of stylomandibular ligament

Introduction

Oral submucous fibrosis (OSF) is an insidious disease affecting the oral cavity, pharynx, and upper digestive tract. Its etiology is directly linked to betel nut usage, common to the Indian subcontinent, Far East, and Pacific Rim [1]. It is characterized by a juxtaepithelial inflammatory reaction followed by fibroelastic change in the lamina propria and associated epithelial atrophy. This leads to a restricted mouth opening, resulting as trismus leading to restriction of food consumption, difficulty in maintaining oral health, as well as impairs the ability to speak [2].

The styloid process arises from the temporal bone immediately in front of the stylomastoid foramen. The normal length of this process is 20–25 mm in adults [3]; the mean radiographic length of the styloid process has been reported to be between 20 and 30 mm [4]. It had been estimated that between 2 and 4 % of the general population presents radiographic evidence of an ossified portion of the stylohyoid chain [5].

An 18 years old male patient presented to us in 2006 with inability to open the mouth wide which was diagnosed as OSF clinically and histopathologically. Regular panoramic radiograph of the patient was obtained and filed (Fig. 1). He underwent surgical treatment (resection of fibrous bands and reconstruction with buccal fat pad flaps) for the same after which he did well. In 2011, he reported back with the same complaint and it was considered to be

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Fig. 1 Panoramic radiograph of the patient (Case no. 9) when reported in 2006

recurrence of OSF. Again a regular panoramic radiograph was obtained (Fig. 2) but this time no significant changes could be seen, except for the changes in dentition, and the appearance of elongated styloid process and/or ossification of the stylomandibular ligament on the right side. This led to a suspicion that there might be some changes in styloid complex with progressive OSF, so we decided to formulate a study to evaluate these accompanying changes in operated OSF patients. The purpose of this study was to evaluate the elongation of the styloid process only in patients with oral submucous fibrosis (OSF) by using panoramic radiographs.

Materials and Methods

Preoperative panoramic radiographs of patients who reported to us for the management of histologically proven oral submucous fibrosis (OSF) were studied from 2007–2011. Inclusion criteria were availability of full details and panoramic radiograph of the patient obtained by a standard method at 70 kV, 10 mA in 12 s. Exclusion criteria were patients having panoramic radiograph not done in our radiology department so as to take care of the magnification and different parameters used by variable systems. A uniform magnification of 1:1.2 was taken into consideration.

On the panoramic radiograph, the apparent lengths of styloid process were measured with the help of divider and steel metric ruler, from the point of emergence of the process to their tips, regardless of whether or not the styloid process was segmented. The length of the styloid process and/or ossification of stylomandibular ligament which were longer than 30 mm were considered significant.

All these patients underwent surgical treatment by resection of fibrous bands and reconstruction with bilateral nasolabial flaps or buccal pad of fat flaps. A control group was not kept because incidence of elongated styloid process is well studied in general population while this study evaluated the incidence only in patients with OSF.

Fig. 2 Cropped panoramic radiograph of the same patient showing calcified stylomandibular ligament on right side when reported in 2011

Approval from ethical committee was not sought as the study was a retrospective study.

Results

Out of 47 patients, 35 patients (34 males and 1 female) met the inclusion criteria. All of these patients had mouth opening of ≤ 20 mm and surgical treatment was done. On panoramic radiographs, 11 patients (31.4 %) were found to have elongated styloid processes and/or ossification of the stylomandibular ligaments which included 10 male patients and 1 female patient. Seven of these patients had bilateral elongation of styloid processes and/or ossification of the stylomandibular ligaments while rest of the patients had unilateral enlargement. The radiographic length of these styloid processes varied from 31 to 52 mm.

Table 1 depicts details of the patients—age, gender, OSF grade according to Khanna and Andrade [6], length of styloid process and involvement i.e. unilateral or bilateral.

Discussion

Styloid complex is composed of the styloid process, stylohyoid ligament and the stylomandibular ligament [7]. The styloid process and ligament are derived from the first and second branchial arches, in addition to Reichert's cartilage. It has been demonstrated that during fetal development Reichert's cartilage links the styloid bone to the hyoid bone. The potential for calcification of this complex of structures and the resulting trauma is from this origin, namely, Reichert's cartilage during embryogenesis. In the adult the stylomandibular ligament, which is normally composed of dense fibrous connective tissue, may

Table 1 Depicting details of patients-Age, Gender, OSF grade, length of styloid process and involvement

Serial number	Age (in years)	Gender	OSF Group	Side involved	Length/Langlais type	
					Right	Left
1.	35	Female	III	Unilateral	35 mm/I	–
2.	48	Male	IVA	Bilateral	38 mm/I	36 mm/I
3.	24	Male	III	Unilateral	–	37 mm/I
4.	48	Male	IVA	Bilateral	45 mm/II	43 mm/I
5.	26	Male	IVA	Bilateral	33 mm/I	31 mm/I
6.	35	Male	IVA	Unilateral	32 mm/I	–
7.	28	Male	III	Bilateral	36 mm/II	38 mm/I
8.	25	Male	III	Bilateral	52 mm/II	37 mm/I
9.	23	Male	IVA	Unilateral	45 mm/I	–
10.	53	Male	III	Bilateral	33 mm/I	34 mm/I
11.	17	Male	IVA	Bilateral	38 mm/I	36 mm/I

retain some of its embryonic cartilage and thus have the potential to become partially or completely ossified. If these structures solidify, they can cause the pain and suffering in symptomatic cases [4].

The shape of styloid process is cylindrical, tapering gradually toward the apex like a pinnacle. The apex of the styloid process is clinically important, because it is located between internal and external carotid arteries, just lateral to the tonsillar fossa within the lateral pharyngeal wall. The tip of the styloid process is continuous with the stylohyoid ligament, which extends to the lesser cornu of the hyoid bone. Muscular and ligamentous structures are attached at various locations of the process. The attached muscles are the stylopharyngeus (arising from the base), the stylohyoid (attached to the middle portion) and the styloglossus muscle (originating from the extremity of the process). The innervations of these three muscles are the glossopharyngeal nerve, the facial nerve, and the hypoglossal nerve, respectively [8].

Eagle's syndrome can be defined as the symptomatic elongation of the styloid process or mineralization (ossification or calcification) of the stylohyoid ligament complex. This syndrome was first documented by Watt W. Eagle an otorhinolaryngologist [3]. Over a twenty-year period, Eagle reported over 200 cases and explained that the normal styloid process is ~2.5–3.0 cm in length. He observed that slight medial deviation of the styloid process could result in severe symptoms of atypical facial pain. From Eagle's early descriptions, patients were categorized into two groups: those who had classical symptoms of a "foreign body" lodged in the throat with a palpable mass in the tonsillar region following tonsillectomy; and those with pain in the neck following the carotid artery distribution (carotid artery syndrome) [9]. Although these two types have a common etiology, their

symptomatology differ. None of our patients had a recent history of tonsillectomy and there was absence of symptoms described above.

Langlais proposed three radiographic appearances: Type I, elongated; Type II, pseudo-articulated; and Type III, segmented. The Type I pattern represents an uninterrupted, elongated styloid process. Type II is characterized by the styloid process apparently being joined to the stylohyoid ligament by a single pseudo-articulation. This gives the appearance of an articulated elongated styloid process. Type III consists of interrupted segments of the mineralized ligament, creating the appearance of multiple pseudo-articulations within the ligament [7]. Langlais type distribution in our patients is described in Table 1.

Langlais further described these types depending upon patterns of calcification: Calcified outline—Thin radiopaque cortex and a central lucency that constitutes most of the process, Partially calcified—Thicker radiopaque outline with almost complete opacification as well as small and occasionally discontinuous radiolucent core, Nodular complex—Knobby or scalloped outline which may be partially calcified with varying degree of central radiolucency, Completely calcified—Totally radiopaque with no evidence of a radiolucent interior [7].

It had been estimated that between 2–4 % of the general population presents radiographic evidence of an ossified portion of the styloid complex. The majority of these 2–4 % of patients are asymptomatic [5]. Despite these figures, only 1–5 % of patients are symptomatic [7]. When symptoms do exist, there is little correlation between the extent of the symptoms and the severity of the ossification. Those few patients who are symptomatic are usually over 40 years of age. The majority of symptomatic patients have had no recent history of tonsillectomy or other cervicopharyngeal trauma [5, 10].

However Correll et al. [11] in a review of 1,771 panoramic radiographs found incidence of mineralization of the styloid complex to be 18.2 % while Keur et al. [8] found it to be 30 %.

Oral submucous fibrosis (OSF) is a collagen disorder associated with chewing of arecanut. Arecoline an alkaloid component of arecanut, stimulates fibroblastic proliferation and collagen synthesis. The flavanoids—catechin and tannins—are also components of the areca nut and stabilize the collagen fibrils, rendering them resistant to degradation by collagenase [12]. The attendant trismus is a result of juxtaepithelial hyalinization and secondary muscle involvement (i.e., muscular degeneration and fibrosis) [6].

In pathogenesis of OSF, it has been well established that chronic inflammation of the oral mucosa leads to activation of T-cells and macrophages at the site with resulting increase in production of cytokines (Interlukin-6, Interferon-alpha) and transforming growth factor beta (TGF-beta) which through various pathways increases the deposition of insoluble collagen and decreases its degradation at the same time [13]. TGF-beta also has well-documented roles in chondrocyte maturation and endochondral ossification [14]. Autocrine and paracrine stimulation by TGF- β is important in the maintenance and expansion of the mesenchymal stem/progenitor cells, the progenitors of osteoblasts. Bone and cartilage contain large amounts of TGF- β and target cells for TGF- β activity [15]. It could be responsible for impetus to remnants of Reichert's cartilage present in styloid complex leading to partial or complete ossification of associated ligaments. This can be one of the reasons for increased incidence of elongated styloid process and/or ossification of associated ligaments.

In conclusion, we have evaluated the elongation of styloid process only in patients with histologically proven oral submucous fibrosis and results were significant (31.4 %) as compared to 2–4 % incidence in general adult population. This highlights that progressive OSF might have some influence on elongation of styloid process. One of the shortcomings of this study was a small sample size. Many authors have studied incidence of elongated styloid process or Eagle's syndrome/Stylohyoid syndrome in general adult populations. This is the first study of its kind to evaluate the incidence in OSF patients. Therefore

establishing the incidence of elongation of styloid process primarily in cases of oral submucous fibrosis (OSF) may need further studies.

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