Evaluation of Habit Patterns and Clinical Findings of Oral Submucous Fibrosis in South Indian Population

ABSTRACT

Objective To evaluate the habit patterns and clinical features of oral submucous fibrosis.

Materials and Methods A hospital-based cross-sectional study was conducted in a sample of 75 subjects with clinically diagnosed oral submucous fibrosis. Prior to the treatment, a detailed habit history was elicited. Clinical symptoms and signs were recorded. Mouth opening, cheek flexibility and tongue protrusion were also recorded.

Results 93.33% had the habit of using processed areca nut products like Gutkha (54.66%), Mawa (20%). The burning sensation was the most common symptom seen in 82% of cases, followed by restricted mouth opening. Blanching and palpable fibrous bands were present in all cases. The mean mouth opening of the sample was 32.73 (±9.10) mm, cheek flexibility was 6.48 (±3.06) mm, and tongue protrusion was 43.92 (±10.24) mm.

Conclusion It is important to be familiar with the habit patterns and clinical features of oral submucous fibrosis to facilitate early diagnosis and prompt management to reduce oral cancer-related morbidity and mortality.

KEYWORDS oral submucosa fibrosis, oral cavity, areca nuts

INTRODUCTION

Oral submucous fibrosis is a chronic, insidious scarring disease of the oral cavity, characterized by progressive inability to open the mouth due to loss of elasticity and development of vertical fibrous bands in labial and buccal mucosa. Oral submucous fibrosis is a well-recognized potentially malignant disorder of the oral cavity, characterized by fibrosis of the lining mucosa of the upper digestive tract involving the oral cavity, oropharynx and frequently the upper third of the oesophagus. The aetiology of oral submucous fibrosis is considered to be multifactorial: betel quid chewing, excessive use of chillies and spices, nutritional deficiency, infectious agents, genetic susceptibility and autoimmunity are some of the suggested contributors in the etiopathogenesis. But, many epidemiologic studies that included case-series reports, large cross-sectional surveys, case-control studies, cohort and intervention studies have identified areca nut as the major etiologic agent. Chewing of betel quid (areca catechu, lime and tobacco) as well as other areca nut containing products (e.g. pan masala and Gutkha) for mouth freshening and the mild euphoric effect is a fairly common practice in India, Pakistan and Sri Lanka. Among the chemical constituents of the areca nut, it is believed that alkaloids from areca nut are the most crucial in the development of the disease while tannin may have a synergistic role. These chemicals appear to interfere with the molecular processes of deposition and/or degradation of extracellular matrix molecules such as collagen. It is estimated that more than 2.5 million people worldwide have oral submucous fibrosis. The majority of whom are in the Indian subcontinent and have a history of betel nut consumption. Histologically, oral submucous fibrosis is characterized by the fibrosis of the lamina propria, which progresses to involve the submucosa and deeper tissues, including the muscles of the oral cavity, resulting in the loss of fibro elasticity. A cochrane database review on interventions for the management of oral submucous fibrosis analyzed the medical treatment options including iron and multivitamin supplements.
lycopene, an extract of tomato, and a range of medicines (e.g. intra lesional injection of steroids, hyaluronidase, human placenta extracts, chymotrypsin, pentoxifylline and collagenase) and surgical options including cutting of the fibrous bands, jaw muscles and joint. The authors concluded that there is a lack of reliable evidence for the effectiveness of any specific interventions for the management of oral submucous fibrosis as illustrated by the paucity and poor methodological quality of trials retrieved for the review 7.

Since oral submucous fibrosis is potentially malignant, early diagnosis is absolutely essential to reduce morbidity and mortality. With the increasing use of Gutkha among the younger population, the prevalence of oral submucous fibrosis can only be expected to increase in the near future with a consequent increase in oral cancer-related morbidity and mortality. This study was conducted to assess the chewing habits of the patients with regard to the agent used, frequency and duration of the habit as well as the clinical signs and symptoms in oral submucous fibrosis.

MATERIALS AND METHODS
A hospital-based cross sectional study was conducted among the patients attending the outpatient department of a teaching hospital. The study was approved by the institutional ethics committee. A written informed consent was obtained from all the subjects participating in the study. The study sample comprised of 75 subjects with clinically diagnosed oral submucous fibrosis. A total of 75 consecutive subjects with a positive history of habitual use of areca nut products and a clinical diagnosis of oral submucous fibrosis were included in the study. The criterion used for the clinical diagnosis of oral submucous fibrosis was the presence of blanching with fibrous bands in at least one part of the buccal mucosa. The exclusion criteria were the history of skin disease and radiotherapy to head and neck region as they could also produce fibrous bands in the buccal mucosa similar to oral submucous fibrosis. The clinical examination was performed by three independent observers. The observers were calibrated prior to the start of the study and were found to have good agreement between them with the kappa value ranging between 0.9 and 0.94 (P < 0.01), when tested with a test sample of 10 cases each. The majority opinion among these three observers was taken as the recorded finding for all categorical variables. The mean of the measurements given by the three observers was recorded finding for all categorical variables. The mean was then asked to blow his cheeks fully and the distance measured between the two points are recorded as the cheek flexibility value. CF = V2–V1.

Measureable clinical parameters
The other clinical parameters recorded were mouth opening and cheek flexibility, as per the technique described by Reddy et al 11.

i. Mouth opening (in mm): Mouth opening (MO) was measured as the distance in mm between the mesioincisal angle of the upper central incisor to the mesioincisal angle of the lower central incisor on the same side.

ii. Cheek flexibility (in mm): Cheek flexibility (CF) is given as the distance between two points on the cheek; two points are marked, one on either side of the cheek corresponding to one third the distance from the angle of the mouth on a line joining the tragus of the ear and the angle of the mouth. The distance between them is measured at rest and denoted as V1. The subject is then asked to blow his cheeks fully and the distance measured between the two points are measured again and given as V2. The difference between the two measurements is given as the cheek flexibility value. CF = V2–V1.

iii. Tongue protrusion (in mm): Tongue protrusion (TP) is measured as the distance between the mesioincisal angle of upper central incisor to the tip of the tongue when maximally extended with mouth wide open.
RESULTS

The mean age of the sample was 31.46 and the standard deviation was 8.16. The minimum age was 20 and the maximum age was 50. The median value was 29 and the mode was 22. 65.3% of the patients were under the age of 35 and 50.66% were in their twenties.

Habit patterns of the sample

Out of the 75 patients, only 5 patients (6.66%) had the habit of chewing conventional betel quid. All the other 70 patients (93.33%) had the habit of using commercially available preparations of processed areca nuts like Gutkha (54.66%), Mawa (20%) and Hans (5.33%). 10 patients (13.33%) had the habit of chewing more than one areca nut product, of which 2 patients (2.66%) chewed Gutkha and Hans, and 7 patients (9.33%) chewed Gutkha and Mawa while one patient (1.33%) chewed Mawa and Hans.

On an average, the subjects in the study sample chewed 5.63 ± 5.068 packets per day. However, the range of chewing frequency varied from 1 packet per day to about 30 packets per day. 51 subjects (68%) chewed 5 packets per day or less. 14 subjects (18.67%) chewed 6–10 packets a day and 10 subjects (13.33%) chewed more than 10 packets a day. On an average, the subjects in the study group had been chewing some areca product for 6.61 ± 5.63 years. However, the duration of the habit ranged from 1 year to 30 years. A total of 46 subjects (61.33%) had the habit of for 5 years or less, 21 subjects (28%) had the habit for 5 to 10 years, 4 subjects (5.33%) had the habit for 11 to 15 years while another 4 subjects (5.33%) had the habit for more than 15 years.

The average length of time that the areca product remains in contact with the oral mucosa in the study sample is 16.58 ± 13.03 minutes. The range of contact time per chew was from 2 minutes to 60 minutes. 11 subjects (14.67%) had the product stacked in the mouth for less than 5 minutes, 27 subjects (36%) had the product stacked in the mouth for 6 to 10 minutes, 19 subjects (25.33%) had the product stacked in the mouth for 11 to 15 minutes, 18 subjects (24%) had the product stacked in the mouth for more than 15 minutes.

In addition to the chewing habits, 20 subjects (26.67%) had the history of smoking as well. The average frequency of smoking in the study sample was 3.95 packets per day and the average duration of smoking habit was 4.15 years. In addition to chewing habits, 21 subjects (28%) have a history of regular alcohol consumption. 10 subjects (13.33%) have a positive history of all the 3 habits (chewing, smoking, alcohol consumption).

Clinical symptoms

Burning sensation of the oral mucosa was by far the most common symptom seen in 82% of cases, followed by restricted mouth opening and dryness of the mouth (29%). Other symptoms included restricted mouth opening (65%), restricted tongue protrusion (18%), altered taste (10%), difficulty in swallowing (6.6%) and difficulty in hearing (4%) (Chart 1).

Clinical signs

Blanching of the oral mucosa and palpable fibrous bands were present in all cases (100%) (Figs. 1, 2). Hyperpigmentation was seen in 44% of cases. Clinical evidence of inflammation was seen in 38% of cases. This inflammation presented as erythematous areas (28%), ulcerated areas (6.6%) or a combination of erythematous and ulcerated areas (4%). Clinically appreciable oral dryness, as evidenced by sticking of the diagnostic instruments to the oral mucosa and the absence of salivary pooling on the floor of the mouth was seen in 18% of cases.

Fibrous bands were seen in all the cases. The distribution of fibrous bands was as follows: Fibrous bands...
were most common in the posterior buccal mucosa in 55 cases (73.3%), followed by the anterior buccal mucosa in 14 cases (18.6%), soft palate in 13 cases (17.3%) (Fig. 4), tongue in 9 cases (12%), labial mucosa in 8 cases (10.6%), floor of the mouth in 8 cases (10.6%), (Fig. 3) and uvula in 6 cases (8%). 41 cases (54.6%) presented with fibrous bands in the posterior buccal mucosa alone. 14 cases (18.6%) presented with fibrous bands in anterior and posterior buccal mucosa. 20 cases (26.6%) showed the presence of fibrous bands in at least one extra-buccal site (palate, labial mucosa, tongue, floor of the mouth) in addition to the buccal mucosa (Charts 2 and 3).

**Measurable clinical parameters**

The mean mouth opening of the sample was 32.73 (±9.10) mm. The mean cheek flexibility was 6.48 (±3.06) mm and the mean tongue protrusion was 43.92 (±10.24) mm. Grade I mouth opening (≥36 mm) was seen in 38.6% of patients; Grade II mouth opening (21–35 mm) was seen in 56% of patients and Grade III mouth opening (≤20 mm) was seen in 5.3% of cases. Grade I cheek flexibility (≥9 mm) was seen in 21.3% of patients; Grade II cheek flexibility (5–8 mm) was seen in 56% of patients and Grade III cheek flexibility (≤4 mm) was seen in 22.6% of cases. Grade I tongue pro
trusion (≥50 mm) was seen in 25.3% of patients; Grade II tongue protrusion (21–49 mm) was seen in 72% of patients and Grade III tongue protrusion (≤20 mm) was seen in 2.6% of cases. A very good inter-observer agreement was seen between the two observers. The kappa value ranged between 0.92 and 0.94 (P < 0.01) for the various clinical parameters (Table 1).

**DISCUSSION**

The mean age in our sample was 31.46 and the mode was 22. 65.3% of the patients were under the age of 35 and 50.66% were in their twenties which clearly indicates that oral submucous fibrosis has increasingly become a disease of the youth affecting them in their most productive years of life. These findings are similar to those reported by Gupta et al. who stated that the disease seemed to be concentrated in lower age groups and that 84.1% of the cases occurred in patients less than 35-years old13 and to those reported by Kumar et al. who found that half of the cases occurred in 20–29 year age group13. This goes on to show the dramatic increase in the number of young patients afflicted with the disease in recent years. Gupta et al. observed that remarkably, the highest period of risk for engaging in areca nut alone is between the ages of 5 and 12 and that oral submucous fibrosis associated with areca nut in children is a great concern for the Society and the Government. Factors associated with the consumption of areca nut are the levels of awareness, household environment, peer pressure, low cost, easy availability etc14.

In our study, we found that Gutka was the most favoured product (54.66%), followed by Mawa (20%) and Hans (5.33%). 10 patients (13.33%) had the habit of chewing more than one areca nut product, of which Gutka was invariably the first component. Various authors feel that promoted by a slick, high-profile advertising campaign and aggressive marketing, pan masala and Gutka have become very popular with all sections of the Indian society, including school children15. This increasing use of refined areca nut products are a cause for concern. It has been reported that an evolving epidemic of oral submucous fibrosis attributed to Gutka use has been documented among youth, with a resultant increase in oral cancer in lower age groups16. Our findings are similar to those reported by Ahmad et al.17 who found that 152 out of 157 oral submucous fibrosis cases in their study used Gutka and other areca nut products and that the relative number of cases who used only Gutka were more17. It has also been reported that habitual chewing of pan masala/Gutka is associated with the earlier presentation of oral submucous fibrosis than betel quid use. Factors which may be responsible for these differences are the tobacco content, the absence of the betel leaf and its carotenes and the much higher dry weight of pan masala/Gutka.18 The relative risk of development of oral submucous fibrosis in habitual areca chewers was proposed by Sinor et al. who found that among the oral submucous fibrosis cases, 98% chewed areca nut regularly in one form or the other whereas among controls 35% chewed areca nut, giving an overall relative risk of 109.6 and that areca nut chewing was practiced most commonly in the form of mawa: a mixture containing mainly areca nut (over 90% by weight), some tobacco, and a few drops of lime. Mawa chewers and those who chewed mawa along with other chewing habits showed very high relative risks. The relative risks increased with an increase in the frequency as well as the duration of chewing habits6. In our study, we found that 13.33% of the sample chewed more than 5 packets per day; 5.33% had the habit of chewing areca products for more than 15 years; 24% had the product stacked in the mouth for more than 15 minutes. These groups of patients can be expected to have a more severe form of the disease since the frequency of chewing, the duration of the habit and the length of time that the product is in contact with the mucosa have all been found to have a positive correlation with the severity of the disease11.

However, there are other studies which suggest that the frequency of chewing rather than the total duration of the habit was directly correlated with development of oral submucous fibrosis and that smoking tobacco does not increase the risk of developing oral submucous fibrosis19. In our study, 26.67% of the subjects had a history of smoking and 13.33% of the subjects had the

**Table 1.** Distribution of the measurable clinical parameters among the different grades in oral submucous fibrosis.

<table>
<thead>
<tr>
<th>Clinical parameters</th>
<th>Mean (mm)</th>
<th>Range max to min (mm)</th>
<th>Grade I</th>
<th>Grade II</th>
<th>Grade III</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mouth opening</td>
<td>32.73 ± 9.10</td>
<td>18–52</td>
<td>29 (38.6)</td>
<td>42 (66)</td>
<td>4 (5.3)</td>
</tr>
<tr>
<td>Cheek flexibility</td>
<td>6.48 ± 3.06</td>
<td>3–18</td>
<td>16 (21.3)</td>
<td>42 (66)</td>
<td>17 (22.6)</td>
</tr>
<tr>
<td>Tongue protrusion</td>
<td>43.92 ± 10.24</td>
<td>12–65</td>
<td>19 (25.3)</td>
<td>54 (72)</td>
<td>2 (2.6)</td>
</tr>
</tbody>
</table>

**Reference values for grading**

<table>
<thead>
<tr>
<th>S. no</th>
<th>Parameters</th>
<th>Grade I (mm)</th>
<th>Grade II (mm)</th>
<th>Grade III (mm)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Mouth opening</td>
<td>≥36</td>
<td>21–35</td>
<td>≤20</td>
</tr>
<tr>
<td>2</td>
<td>Cheek flexibility</td>
<td>≥9</td>
<td>5–8</td>
<td>≤4</td>
</tr>
<tr>
<td>3</td>
<td>Tongue protrusion</td>
<td>≥50</td>
<td>21–49</td>
<td>≤20</td>
</tr>
</tbody>
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history of smoking and alcohol consumption in addition to areca chewing habits. Some studies indicate that cigarette smoking is an independent risk factor for the development of oral leukoplakia and not oral submucous fibrosis, but it has been noticed that the risk of oral leukoplakia and oral submucous fibrosis is greatly increased in the presence of both betel quid chewing and smoking while alcohol consumption was not related to the development of oral submucous fibrosis. Other studies categorically state that Betel chewing was the only significantly associated factor in the aetiology of oral submucous fibrosis and that there were no interaction effects of chewing, smoking and alcohol consumption in the causation of oral submucous fibrosis.

In our study, we found that burning sensation of the oral mucosa was by far the most common symptom seen in 82% of cases, followed by restricted mouth opening and dryness of the mouth. The burning sensation could be caused by epithelial atrophy and could initially manifest as intolerance to spicy foods, which later becomes persistent. In fact, relief from burning sensation is one of the prime goals of any treatment regimen and is the most frequently used parameter to assess the therapeutic efficacy of any new management option for oral submucous fibrosis. Restriction of mouth opening is slow and progressive and most patients seek treatment only when it becomes severely disabling. In our study, difficulty in swallowing was seen in 6.6%, possibly due to oropharyngeal involvement and difficulty in hearing in 4%, presumably due to Eustachian tube dysfunction, leading to conduction deafness. Gupta et al. observed that degenerative changes in palatal/paratubal muscles were found in the form of loss of cross striations, oedematous muscle fibres and atrophy and concluded that there was a definite involvement of palatal and paratubal muscles in oral submucous fibrosis. Similarly, blanching and palpable fibrous bands were seen in all cases, followed by hyperpigmentation (44%) and clinical evidence of inflammation that included erythema, ulceration or vesiculation (28%). Kerr et al. in their cochrane systematic review have noted that the clinical manifestations include blanching and stiffening of the oral mucosa leading to limitation in oral opening and that the presence of fibrous bands in lips, cheeks and soft palate is a hallmark of the disease.

Oral submucous fibrosis needs to be clinically differentiated from Scleroderma. In scleroderma, the tongue, soft palate and larynx are the intraoral structures usually involved. The tongue becomes stiff and board-like and may result in dysphagia and the gingival tissues are pale and unusually firm. However, patients with scleroderma also exhibit microstomia, purse-string mouth due to radiating furrows around the oral cavity and generalized widening of the periodontal ligament space, in addition to other systemic manifestations. Oral submucous fibrosis differs from the other examples of pathological fibrosis (e.g. juvenile aggressive fibromatoses, abdominal desmoids) in that it harbors with it a definite tendency to induce the overlying epithelium to undergo neoplastic transformation, at least in a small proportion of cases.

Blanching of the oral mucosa has been recognized as an early sign of oral submucous fibrosis. Blanching is defined as persistent white marble-like appearance of the oral mucosa. Unfortunately, however, blanching is not specific to oral submucous fibrosis. The pallor of anemia or the fibrosis in scleroderma could also mimic the blanching seen in oral submucous fibrosis. Pindborg et al. observed pigment changes—either as loss of pigment or as hyperpigmentation in most cases of oral submucous fibrosis. In our case, hyperpigmentation was seen in 44% of cases. But diffuse pigmentation of the oral mucosa is not rare or pathognomonic of oral submucous fibrosis. Hyperpigmentation is seen as part of racial or physiological pigmentation as well as some genetic syndromes, endocrine diseases and drugs. Other inflammatory changes like petechiae, vesicle formation, erythematous areas and ulceration are not only inconsistent, but may also be transient, given the fact that they represent the early “stomatis” phase of the disease, which is later replaced by the “fibrosis” phase. Both hypersalivation and oral dryness have been reported in oral submucous fibrosis by various investigators. Hypersalivation, excessive dryness of the mouth and defective gustatory sensation could all result from the irritant effect of the areca nut products. In our study, 29% of patients complained of dry mouth while only 18% showed objective evidence of dry mouth that included the absence of salivary pooling in the floor of the mouth or the mouth mirror sticking on to the oral mucosa.

It is the consistent presence of fibrous bands that characterizes the disease. The presence of fibrous bands in lips, cheek and hard palate is considered as the hallmark of the disease. In our study, we found that buccal mucosa was the most common site of involvement. Fibrous bands were most common in the posterior buccal mucosa (73.3%), followed by the anterior buccal mucosa (18.6%), soft palate (17.3%), Tongue (12%), labial mucosa (10.6%), floor of the mouth (10.6%), and uvula (8%). In an earlier study conducted by Wahi et al. palate was reported as the most common site of involvement (53%), followed by buccal mucosa in 46% and tongue in 3%. This clinical profile has probably changed in the recent years due to alterations in the patterns of areca nut use, prolonging stacking of commercially prepared freeze dried sweetened areca nut products in the buccal mucosa and probably a greater familiarity among the medical practitioners about the presentation of the condition, leading to higher and early detection rates. Recent evidence strongly suggests that buccal mucosa is the most commonly involved site, similar to our own observation.

Mouth opening, cheek flexibility and tongue protrusion have been used by various investigators as objective measurable parameters of disease severity in oral submucous fibrosis. Some investigators have found that the
ability of each subject to position 3 or 4 fingers, vertically aligned, between the upper and lower central incisors up to the first distal interphalangeal folds, showed a strong positive correlation with the maximal mouth opening. They suggested that the ability to position 3 fingers in the mouth during the dental examination is a convenient index for assessing normal maximal mouth opening. This is a convenient method for application in large community surveys, the more accurate and scientific method for measurement of mouth opening would be measuring the distance between the incisors during maximal mouth opening, as done in our study.

The cut-off values for mouth opening for various grades of oral submucous fibrosis are different as reported by different investigators. Kumar et al. classified oral submucous fibrosis into three grades based on mouth opening values as >45, 20–44 and <20 mm. In a more recent classification, More et al. classified oral submucous fibrosis into four grades based on mouth opening values as >35, 25–35, 15–25 and <15 mm. Normal mouth opening, as reported by several investigators, is in 5.03 cm in males and 4.5 cm in females. It is usually measured as the vertical distance between the mesioincisal angles of the upper and lower incisors. Cheek flexibility and tongue protrusion have also been realized as a measurable parameter in oral submucous fibrosis. We believe that cheek flexibility could be a more reliable indicator of disease severity in oral submucous fibrosis than mouth opening as it is more likely to reflect the extent of fibrosis in the buccal mucosa and the underlying buccinator muscle than mouth opening which is likely to be influenced by the anatomic site of fibrosis. But since protrusion is affected in only the advanced stages of the disease, it may not be rational to use tongue protrusion as an indicator of disease severity in all cases of oral submucous fibrosis.

CONCLUSION

This hospital based cross sectional study has shown that the prevalence of oral submucous fibrosis is increasing among the youth of south India. Gutkha is the most common chewing agent associated with oral submucous fibrosis. Burning sensation and tongue protrusion have also been measured as the vertical distance between the incisors during the dental examination is a convenient index for assessing normal maximal mouth opening. They suggested that the ability to position 3 fingers in the mouth during the dental examination is a convenient index for assessing normal maximal mouth opening. This is a convenient method for application in large community surveys, the more accurate and scientific method for measurement of mouth opening would be measuring the distance between the incisors during maximal mouth opening, as done in our study.

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REFERENCES