Clinical Features: Oral Submucous Fibrosis

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Contents

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3.1 Introduction

Oral submucous fibrosis (OSF) develops insidiously, and as the early signs and symptoms are rather non-specific, the clinical presentation can be easily overlooked. As the diseases progresses, OSF may present with a wide range of signs and symptoms affecting several subsites of the oral cavity, the pharynx and the upper part of oesophagus. The sites mostly affected are the lips, buccal mucosa, tongue, and palate. Fibrosis of oral tissues leading to limitation of mouth opening remains the hallmark of this disease. This chapter presents a comprehensive description of clinical manifestations to enable a health professional to make a clinical diagnosis during an initial consultation.

Learning Goals

The primary goal of this chapter is to educate dentists and physicians in the detection of the early stages of oral submucous fibrosis by learning the signs and symptoms of the disease.

The chapter also lists the essential clinical criteria for patient selection when planning field surveys on oral submucous fibrosis.

3.2 Brief Review of the Literature

This disorder was first reported by Schwartz in 1952 [1], in five Indian women living in Kenya but as stated in Chap. 1 this description is no longer accessible in the published literature. Subsequently, the clinical features of OSF were reported, by three Indian clinicians, Joshi [2], Lal [3] and Desa [4], and three cases of OSF were described by Su from Taiwan [5]. A comprehensive clinical and pathological description of OSF was first published by Pindborg during his travels to India [6–9]. The establishment of a large-scale epidemiological investigation by Fali Mehta's group based at the Basic Dental Research Unit of the Tata Institute of Fundamental Research that investigated and reported on 50,915 villagers from five different regions in India [10, 11] contributed largely to enhance the knowledge of the international research community on this disorder. Around the same period, a further impetus to the study of OSF came from investigations on Indian residents in South Africa in Durban by Shear's group [12, 13] and by van Wyk's group [14, 15].

Staging of OSF based on clinical signs and symptoms, and taking into account degrees of mouth opening, functional aspects and presence or absence of a malignancy, has been presented by various authors [16–21], and some modifications to the Kerr classification were done by More [22, 23], and the latest by Arakeri et al. [24]. See details of OSF staging in this volume, ▶ Chap. 6.

Clinical aspects of OSF have been a subject discussed at several international conferences. In 1992, the first Asia-Pacific Workshop on Oral Mucosal Disorders was held in Nagoya, Japan [25]. In 1997, two expert symposia on OSF were held, one in London [26] and the other in Kuala Lumpur [27]. Criteria for diagnosis of OSF were published following the Kuala Lumpur Symposium [27, 28]. The Fifth World Workshop on Oral Medicine included OSF as a topic to develop a systematic review on the disease [21] and the WHO Collaborating Centre's Working Group on Oral Potentially Malignant Disorders recently enumerated the criteria for diagnosis of OSF [29].

Definition

A definition of clinical features should include the presence of fibrous bands in lips, buccal mucosa, soft palate and fauces as the hallmark of the disease that leads to marked limitation of mouth opening.

3.3 General Aspects: Age and Sex

OSF is a disease found in population groups from South and Southeast Asia and South Pacific islands or among the migrants from these countries to East and South Africa, Europe and North America. The initial literature referred to a female preponderance, but later studies reported that males are more affected by the disease see ▶ Chap. 2. The proportion of males affected in China and in Taiwan appears to be significantly higher than that in India [30, 31]. The common age range at presentation is between 20 and 40 years. Oral submucous fibrosis in a 4-year-old girl whose parents had moved from India to Canada was reported by Hayes [32]. The disease has been described in all ages ranging from 4 to 89 years.

3.4 Signs and Symptoms

The signs and symptoms of OSF could be broadly divided to early, intermediate and advanced stages of the disease and are listed in ◘ Table 3.1. The anatomical sites affected by OSF are listed in ◘ Table 3.2.

The most common early symptom is a burning sensation of the mouth, often noted at mealtimes while consuming spicy food. Appearance of transient vesicles, diffuse blanching of the mucosa and loss of tongue papillae have been found in population surveys as early forms of submucous fibrosis [33].

Table 3.1 Clinical presentation of oral submucous fibrosis by stages of development

Early stage	Intermediate stage (additional features)	Advanced stage (additional features)
Burning sensation	Sore mouth Sensitivity to hot and cold foods	Stomatitis
Blanching of oral mucosa	Depigmentation	Marble-like appearance of oral mucosa
Leathery mucosa	Palpable fibrous bands in lips, buccal mucosa, retromolar trigone and soft palate	Thick and broad fibrous bands of buccal mucosa
No limitation in mouth opening	Limited mouth opening (20–40 mm)	Limited mouth opening (<20 mm)
Depapillation of tongue	Limited movement of tongue	Fixed tongue
		Sunken cheeks
		Dysphagia, rhinolalia, loss of hearing
		Weight loss

Table 3.2 Morphological features noted in different anatomical sites

Sites	Presenting Features
Lips	Thinning and distortion of lips Depigmentation
Cheeks	Sunken appearance, Vertical skin folds
Buccal mucosa	Vesicles, blanching, marble-like appearance, fibrous bands, patchy hyperpigmentation
Tongue	Depapillation of dorsum, later fixation
Soft palate	Petechia, fibrous banding, distorted uvula
Fauces	Fibrous banding

As the disease progresses, the oral mucosa becomes more blanched, and whitish areas appear in discrete locations giving the appearance of depigmentation of the mucosa. The mucosa develops a leathery feeling, and fibrous bands first appear around the faucial pillars.

The pathognomonic features of OSF are the presence of palpable fibrous bands, tongue becoming progressively immobile and fibrous bands in the buccal mucosa and retromolar regions leading to limited mouth opening.

As the disease extends inwards from the oral cavity to the pharynx and the upper third of the oesophagus, additional symptoms may be noted during advanced stages. These include dysphagia, deafness, rhinolalia and loss of weight. Characteristic facial features include sunken cheeks and presence of multiple vertical perioral skin folds.

OSF could coexist with several associated oral mucosal lesions [34]. These presentations are discussed further in ▶ Chap. 4 (see ▶ Sect. 4.3).

Important

Key features of the disease include

1. Palpable fibrous bands in the buccal mucosa and retromolar regions.
2. Tongue becoming progressively immobile.
3. Limited mouth opening.

3.5 Burning Sensation

Burning sensation is a universal feature of OSF and can be present spontaneously but is more commonly reported during consumption of spicy foods. This important early symptom is often disguised as a feature of anaemia, and the disease is overlooked by general practitioners during routine consultations. Many patients with OSF tend to reduce the consumption of chillies and spicy food over the evolution of the disease. An OSF patient could suffer from burning sensation for the rest of the life, and this does affect the quality of life. The cause is not well understood but is attributed to the atrophy and flattening of rete ridges of the oral epithelium that could be present from the very early stages of the disease.

3.6 Blanching of the Mucosa

Blanching of the oral mucosa was first described as an early feature of OSF by Pindborg et al. [11] based on a field study undertaken in Ernakulam district in India in their baseline survey of 10,169 subjects in 1967. Blanching is characterised by loss of the pink colour and mucosa appearing whitish in discrete areas of the oral mucosa. This feature appears even before the onset of fibrosis and is often under-recognised. Figure 3.1 illustrates a blanched mucosa in a 30-year-old man—an areca nut chewer—who later developed fibrous bands on follow-up. This could often be mistaken for pallor often observed in deficiency states such as anaemias.

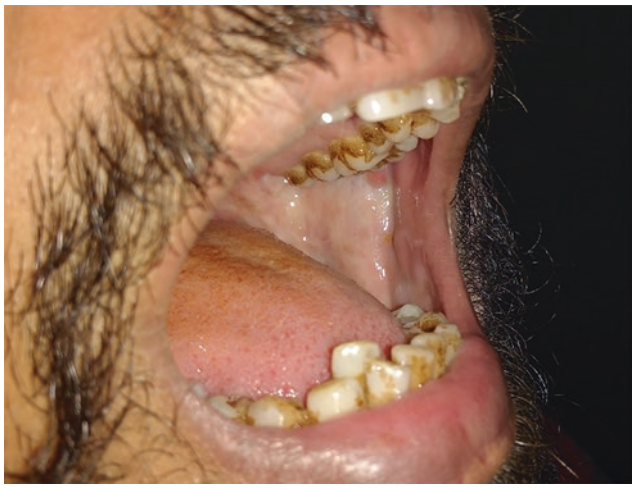

Fig. 3.1 Blanching of buccal mucosa in an areca nut chewer—an early feature of OSF

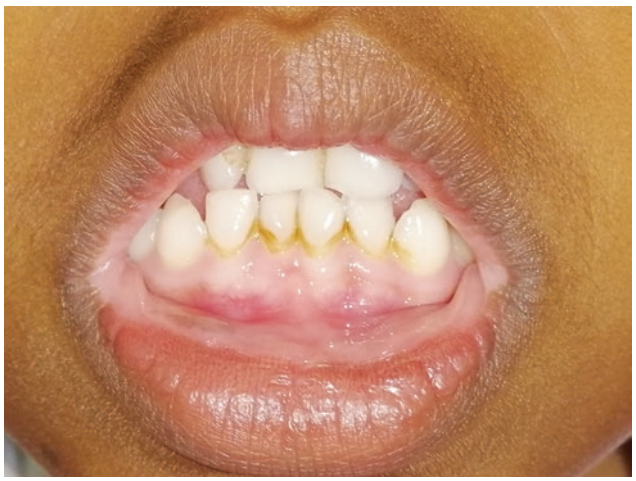

Fig. 3.2 Depigmentation of labial mucosa and the commissure in a child

3.7 Depigmentation

Loss of pigmentation of the oral mucosa (Fig. 3.2) has been reported in several studies. Depigmentation near the vermilion border of the lip in five young children aged 2–3 years as the sole clinical feature and as the earliest sign of OSF was reported by Sitheeque et al. [35].

The authors claim that depigmentation is different from blanching referred to above. Depigmented sites retain a glossy appearance, while blanched mucosa exhibits a matt appearance. In cases with loss of pigmentation, several conditions should be considered in the differential diagnosis before confirming OSF: These include focal type of vitiligo, localised scleroderma and lichen sclerosus particularly when affecting the lip.

In advanced cases along with loss of pigmentation, there may be patchy hyperpigmented areas (Fig. 3.3) of the oral mucosa [33].

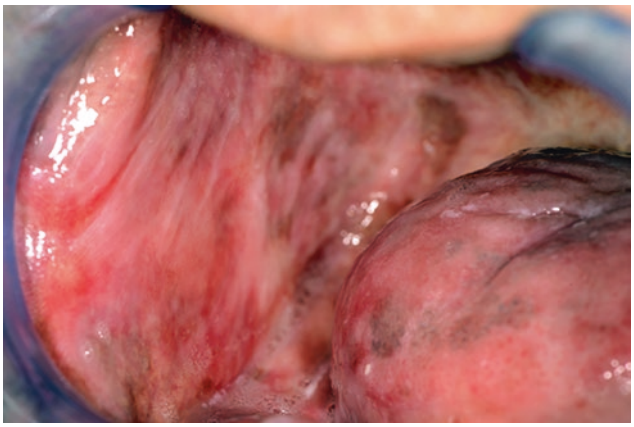

Fig. 3.3 Patchy hyperpigmentation of buccal mucosa and dorsal tongue in an adult patient

3.8 Leathery Mucosa

Another early feature of the disease is a leathery consistency that is observed during palpation of lips and buccal mucosa. This feature often precedes the development of fibrous bands and is an indication of the early loss of fibroelasticity of the mucosa. The leathery feeling is fairly well generalised over the lining surfaces of the mucosa. The process is somewhat similar to a tanned skin changing to leather and gives a tough feel to the oral mucosa.

3.9 Marble-Like Appearance

Due to increased blanching, pallor and loss of pigmentation, the oral mucosa takes up a characteristic generalised marble-like appearance (Fig. 3.4), particularly affecting the buccal mucosa bilaterally.

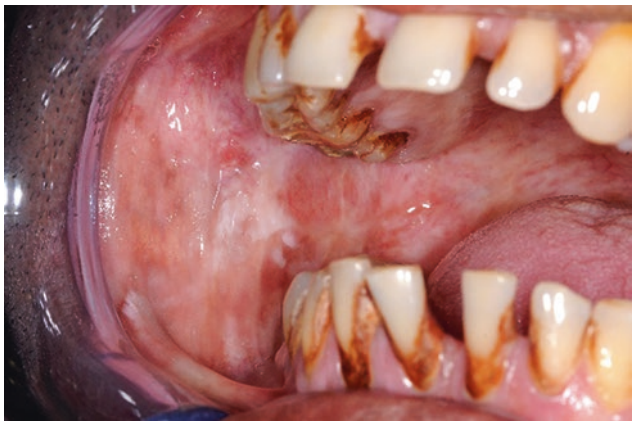

Fig. 3.4 Marble-like appearance of buccal mucosa in moderately advanced OSF

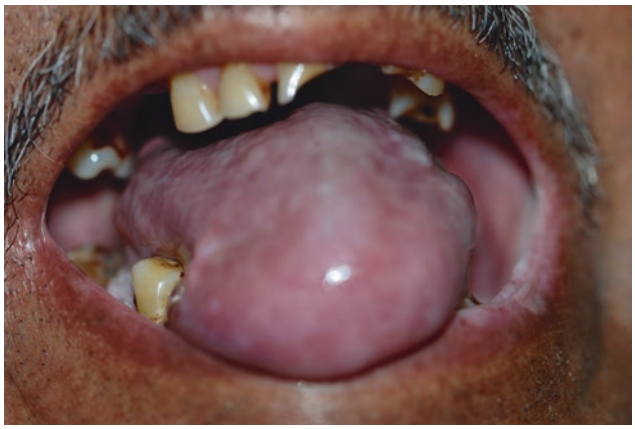

Fig. 3.5 Totally depapillated tongue giving a glossy appearance

3.10 Depapillation of Tongue

Loss of filiform papillae from the dorsum of tongue (Fig. 3.5) is another common feature found in OSF patients. Initially, the feature is noted as a partial loss, and as the disease evolves, there is almost complete loss of tongue papillae. This results in a glazed tongue with or without any erythema. Figure 3.5 illustrates the feature of a glazed tongue in OSF due to loss of papillae. This appearance could masquerade glossitis. The differential diagnosis should include erythema migrans and glossitis found in iron-deficiency anaemia and Plummer-Vinson syndrome.

3.11 Vesicles

Vesicles that may last up to few days may appear during any stage of OSF but are more common in the early stages of the disease. In OSF, vesicles are usually located on the soft palate and the anterior pillars of fauces but may also be found on other areas of lining mucosa. Vesicles are subepithelial and retain the normal surface colour of the oral mucosa. They usually rupture within 24 to 72 hours discharging an aseptic fluid [36] leaving small ulcers.

3.12 Petechia

Tiny, circular red or blue spots on the oral mucosa, caused by minor vascular dilatations or a minor bleed from broken capillary blood vessels, were reported by Bhonsle et al. [37]. The authors found blue spots in 22% of 40 patients with OSF in India.

3.13 Ulceration and Stomatitis

Mouths of OSF patients may demonstrate superficial ulceration especially in the area of retromolar trigone. A sore mouth and stomatitis are common findings.

3.14 Fibrous Bands

The most outstanding feature and the important reliable sign of OSF is the presence of palpable bands in the oral mucosa. The soft palate and fauces are first affected by fibrosis (Fig. 3.6), and as the disease advances, fibrosis gradually involves the retromolar region, buccal mucosa (Fig. 3.7) and lips. Haider et al. [19] based their clinical staging on the location of fibrous bands in the mouth and grouped clinical stages by (1) faucial bands only, (2) faucial and buccal bands and (3) faucial, buccal and labial bands. They concluded that bands are common at the back of the mouth in mild cases of OSF and, as the disease increases in severity, are more likely

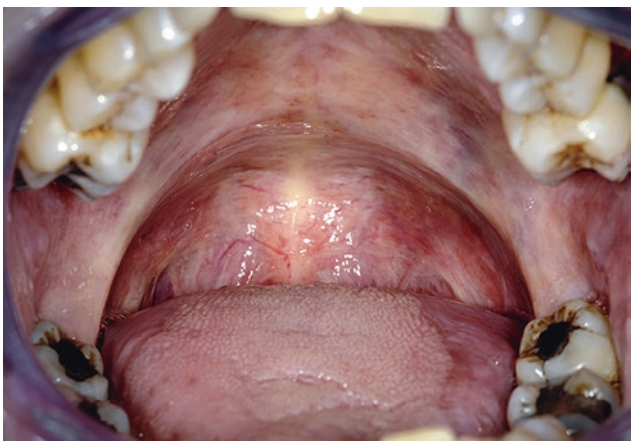

Fig. 3.6 Horizontal fibrous band across the soft palate

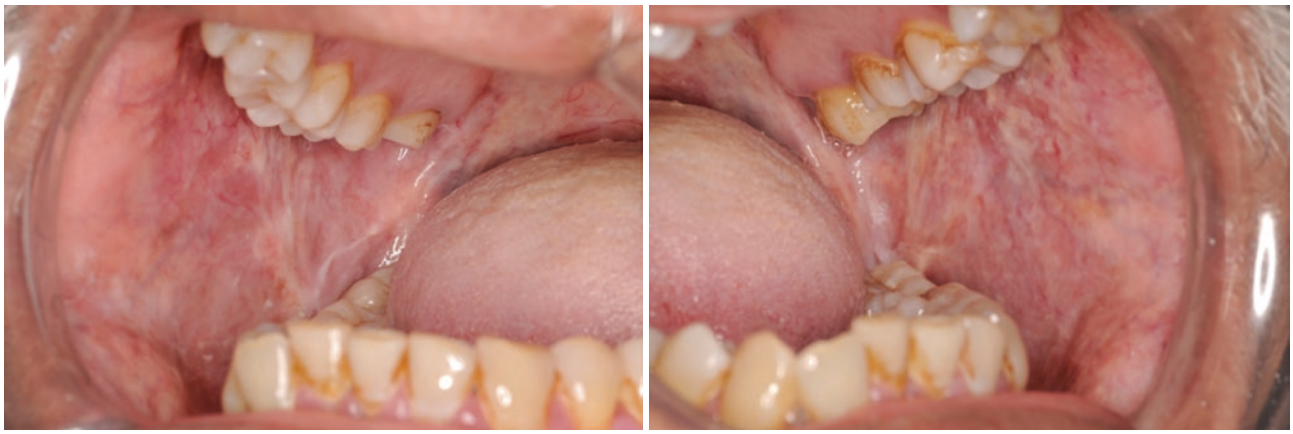

Fig. 3.7 Vertical fibrous bands bilaterally on the buccal mucosa

to be found anteriorly. However, a study from Taiwan reported that only a quarter of their OSF patients presented with fibrosis of palate [38]. More frequent affliction of posterior parts of the mouth has been attributed to the habit of pure areca nut consumption (without betel quid) as noted in Maharashtra state in India [39]. The degree of fibrosis can be semi-quantitatively assessed in the buccal mucosa by the presence of a single band, multiple bands or a broad band when it extends over two cm in width [16]. The extent and severity of fibrous banding found in the retromolar region (around the pterygomandibular raphe) or the buccal mucosa have a significant correlation with the function of mouth opening. Though fibrous bands may be visible on opening the mouth during a systematic oral examination, it is important to palpate both sides of the buccal mucosa to find any palpable fibrous bands. In the field surveys conducted by Fali Mehta's group, the demonstration of fibrous bands was mandatory for the diagnosis of OSF [33, 40, 41]. The expert group that met in Kuala Lumpur also included this as an essential inclusion criterion for diagnosis of OSF [28].

3.15 Distorted Uvula

As a component of fibrosis of the palatal arch, the shape and size of the uvula could be affected and could appear shrunken or bud-like. The uvula may also show deviations [42]. The uvula may point anteriorly instead of downward pointing and present as inverted or in the shape of a hockey stick (Fig. 3.8).

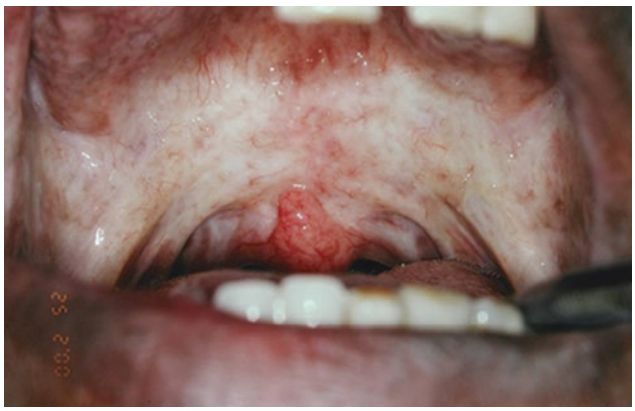

Fig. 3.8 Anteriorly pointing uvula due to palatal fibrosis

Tip

Observing a deformed uvula should raise the suspicion of patient presenting with OSF.

3.16 Limited Mobility of Tongue

Fibrosis generally affects the tongue during intermediate and advanced stages (Table 3.1). This leads to limited mobility of the tongue. On protrusion, a normal tongue could reach beyond the mucocutaneous junction of the lower lip. However, when affected by fibrosis, mobility may be restricted and the tongue cannot be protruded beyond the incisal edges (Fig. 3.9). Lateral movement will also be restricted. In advanced cases, the

tongue remains in a fixed position and limits the rolling movement. Once the fibrosis involves the striated muscle, the tongue structure will demonstrate hardness on squeezing the tongue during palpation.

3.17 Limited Mouth Opening

As fibrosis advances, one of the main consequences of OSF relates to the restriction of mouth opening (Fig. 3.10). In an Indian survey, 90.8% of the survey subjects reported inability to open the mouth wide as their chief complaint [39]. Degree of mouth opening measured as interincisal distance has been used as an objective criterion to stage the disease [16] and in many subsequent systems (e.g. [23, 29, 43]). As a reference guide, the average size of the mouth opening of South Indian males was reported as 47.5 mm, and 44.6 mm in females [44]. The majority of OSF patients present with a mouth opening <40 mm and > 20 mm [45]. An opening of less than 20 mm is considered as a presentation of advanced disease. A device useful to measure the interincisal distance is illustrated in Fig. 3.11.

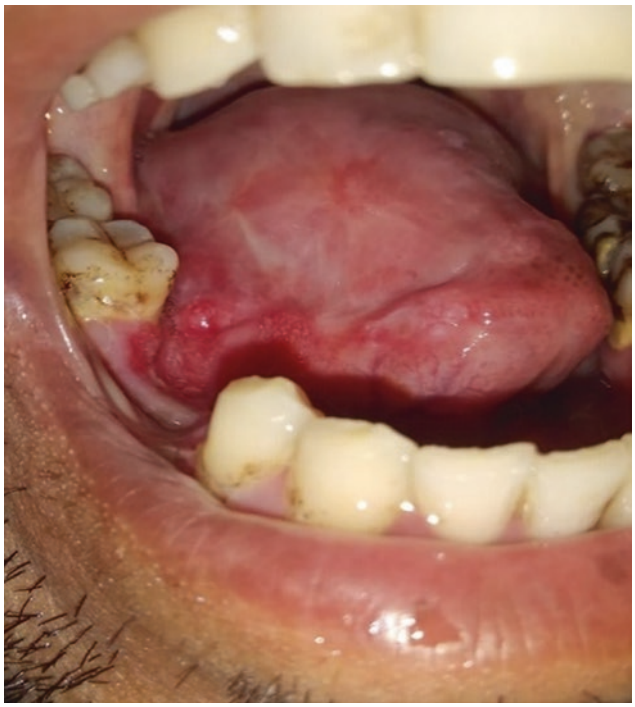

Fig. 3.9 Limited mobility of tongue. On attempting to protrude, the tongue does not reach lower incisors

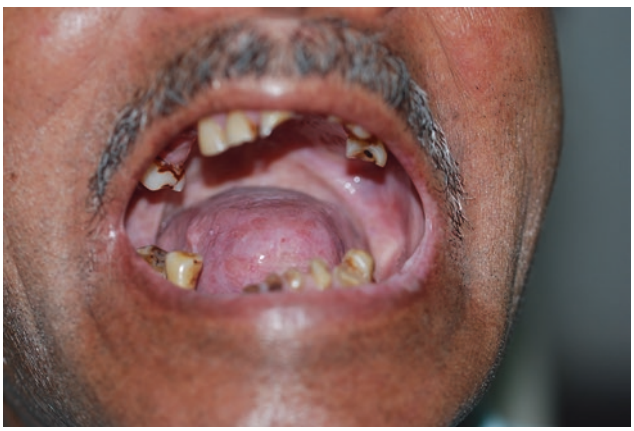

Fig. 3.10 Limited mouth opening in a moderately advanced OSF patient. Vertical skin fold on cheeks is also seen

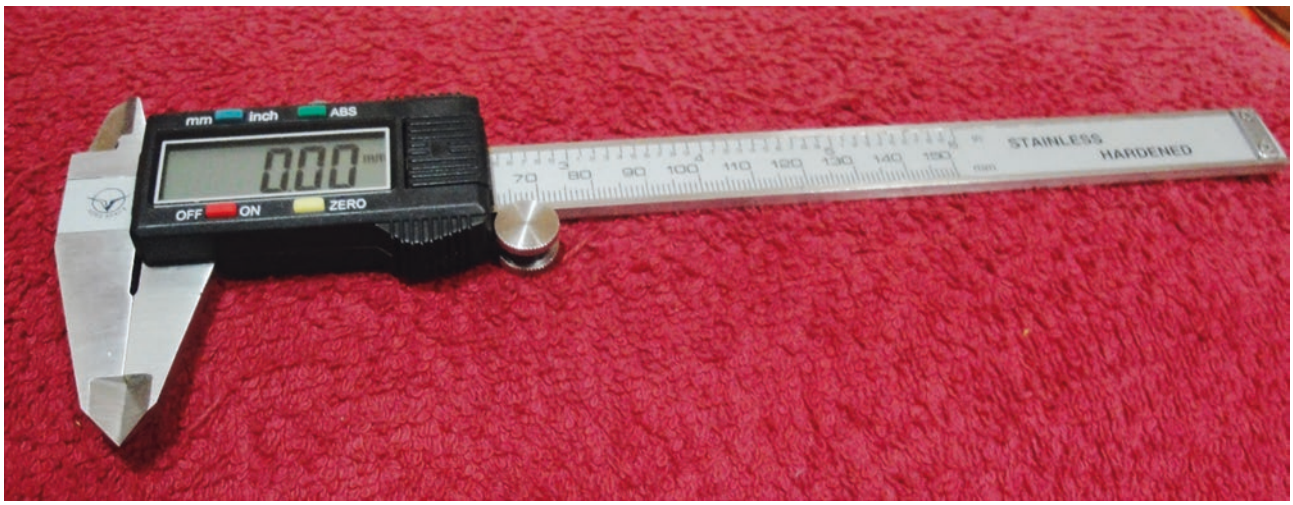

Fig. 3.11 Callipers to measure mouth opening (Courtesy Prof. Vinay Hazarey)

3.18 Other Associated Clinical Conditions

OSF could coexist with several associated oral mucosal lesions, some that are also potentially malignant [29, 46], e.g. oral leukoplakia, erythroplakia, erythroleukoplakia, exophytic verrucous hyperplasia [47] betel quid-associated oral lichenoid lesions [48, 49], Oral squamous cell carcinoma arising from OSF may be found in patients with poor access to care [50]. These presentations are discussed further in ▶ Chap. 4.

Oesophageal subepithelial fibrosis as an extension of OSF leading to thickening and narrowing of the oesophagus was first reported by Maher et al. [51] and by taking endoscopic biopsies by Misra et al. [52].

3.19 Mastication and Deglutition

The ability to masticate food is affected due to reduced oral opening and generalised stomatitis. Moreover, the mucosa becomes extremely sensitive to hot, cold and spicy food that makes it difficult to tolerate any form of food in the mouth. In advanced stages, the patient may have to be tube fed. Loss of suppleness of tongue severely affects the formation of a bolus of food, and due to fibrosis and narrowing of the upper digestive tract, deglutition may be affected (Maher et al. [51]). Dysphagia could be a presenting symptom particularly among rural populations who have not had access to early diagnosis. Defective gustatory sensation was reported by Seedat and van Wyk [15].

3.20 Extraoral

Thinning or distortion of lips is visible extraorally. Chaturvedi referred to sunken cheeks in people affected by OSF as a part of what he coined as Gutka syndrome [53]. Ranganathan et al. [44] referred to reduced cheek flexibility. Other aspects include the presence of multiple peri-oral skin folds instead of a single deep nasolabial fold. Due to stiffening of cheeks, inability to whistle or to blow out a candle is reported [6]. Referred pain in the ears and deafness due to occlusion of Eustachian tubes were reported in Indian studies. Speech may be affected due to difficulty in the pronunciation of words as a result of fixation of the tongue muscles.

Summary

Oral submucous fibrosis is a generalised condition that presents with a plethora of signs and symptoms involving the whole oral cavity and the upper digestive track. An understanding of the initial signs and symptoms could help a healthcare provider to make an early diagnosis and to institute interventions, primarily advice on the cessation of areca nut that could halt the progression of the disease. Presence of fibrous bands in lips, buccal mucosa, soft palate and fauces is the hallmark of the disease that leads to marked limitation of mouth opening. In advanced stages, the oral health and quality of life are severely compromised leading to loss of weight and a cachexic state.

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