



## An Unusual Pediatric Oral Submucous Fibrosis Case Report with Combinative Treatment Plan

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### Abstract

Oral Submucous Fibrosis (SMF) is considered as an irreversible, chronic and premalignant condition with the clinical manifestation of progressive fibrotic scar in the oral soft tissue. This phenomenon would lead to complications such as burning mouth sensation and xerostomia. Moreover, in more severe cases, this disease can lead to trismus, limitation of mouth opening, microstomia, speech disorders, as well as difficulty in the tongue movement and swallowing. Although SMF etiology is multi factorial; but the main cause of this lesion in children would remain unclear and ambiguous. This disease is a rare condition in children and the post-operative relapse is common.

In this paper, the authors presented an unusual case of 10-year-old boy with a rare oral submucous fibrosis, with unknown etiology, who was misdiagnosed as a temporomandibular joint ankylosis. The patient was managed successfully with a particular combined treatment, including pharmaceutical, surgical, physiotherapeutic, and prosthetic approaches, and followed for 6 months. Maxillofacial surgeons are recommended to be familiar with signs and symptoms of SMF as one of trismus etiological probabilities. The correct diagnosis and treatment planning in these patients are considered to be as essential factors to successfully achieve the desirable outcomes. Also, it reduces the morbidity and mortality associated with misdiagnosis or mismanagement of this condition.

**Keywords:** Oral submucous fibrosis; Trismus; Ankylosis

### Introduction

Oral Submucous Fibrosis (SMF) is considered as a chronic, progressive, premalignant and irreversible lesion that it was first described by Sirsat and Pindborg three decades ago [1-3]. The clinical manifestation of this lesion is presented as a fibrotic progressive scar of mucosa that covering upper areas of the gastrointestinal tract especially oral cavity, oropharyngeal region and upper third of esophagus [4-6]. The mucosa would become pale, tender and rigid in this condition which may cause some complications such as burning mouth sensation, spicy food intolerance and dry mouth (Xerostomia). As a matter of fact, SMF signs and symptoms were reported bilaterally in previous studies [1,2,7,8]. Furthermore this disease can lead to trismus, limitation of mouth opening, microstomia, speech disorders, as well as difficulty in the tongue movement and swallowing, in more severe cases [8]. SMF noticeably has higher prevalence in particular geographic areas such as Southeast Asia, Sri Lanka, Bangladesh and the Indian Subcontinent [9-12].

Several predisposing factors have been reported in relation to etiology of SMF such as local factors (including spicy food consumption, Areca-nut, Betel quid), systemic factors (including anemia, iron deficiency, vitamin B complex deficiency, connective tissue-collagen diseases and malnutrition), as well as genetic and immunological factors [13]. While the main reason of SMF remains obscure, but some studies have suggested the association of this lesion with nutritional habits and addiction [4]. Oral SMF typically occurs in adults aging between 20 to 40 years old and it tends to have a slightly higher incidence in women [8].

As a matter of fact, oral SMF occurrence in children would be considered as a rare condition and only limited studies have been reported this lesion in children (under 15 years old) since 1985 up to now [6,9,14-23]. It is important to note that the etiology of the pediatric SMF in these reports were related to spicy foods consumption habits or even the drug abusing by the children [17,22]. The

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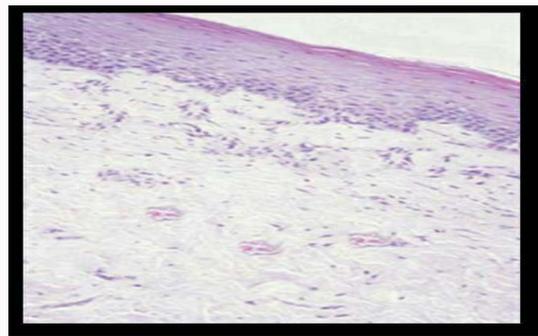
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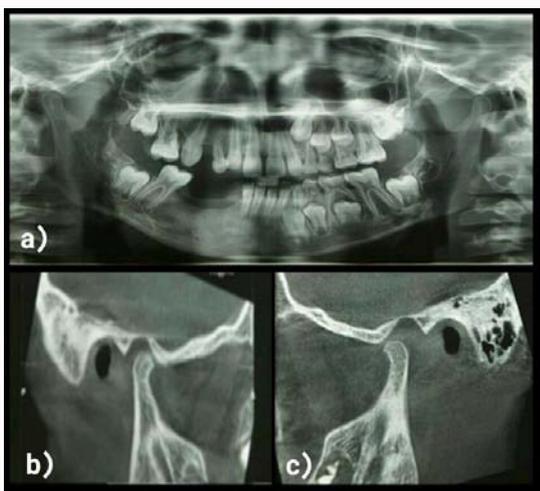
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**Figure 1:** Clinical examination of Maximum Mouth Opening (MMO) was measured 10 mm.



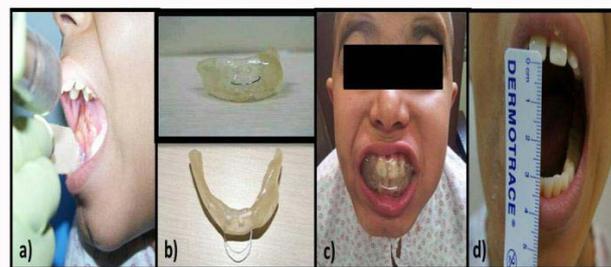
**Figure 3:** Formation of fibrous tissue with inflammatory cells was observed at submucosa layers.



**Figure 2:** Lateral or protrusive jaw movement radiographic examination (panoramic and CBCT).



**Figure 4:** Mouth opening up to 40 mm.



**Figure 5:** Intra lesion Triamcinolone injection was performed.

post-operative relapse of this disease is common, therefore various treatment protocols were presented in recent studies [24,25].

Herein, the authors reported an unusual case of 10-year-old boy with a rare unilateral oral submucous fibrosis, with unknown etiology, who was misdiagnosed as a TMJ (Temporomandibular Joint) ankylosis. The patient was managed successfully with a particular combined treatment, including pharmaceutical, surgical, physiotherapeutic, and prosthetic approaches. Also the review of literatures about this rare condition in children was presented here.

### Case Presentation

A 10-year-old boy was referred to the Oral and maxillofacial surgery department of Kerman Bahonar hospital in Iran with chief complaint of restricted mouth opening (10 mm). According to the patient’s companion statements, this complication has been progressively developed since 6 months ago. At first the patient was visited by a plastic surgeon then referred to maxillofacial surgery service with the misdiagnosis of TMJ ankylosis for further investigation and also bilateral condylectomy treatment plan.

The patient did not have any certain systemic problem in his past medical history, but he had the history of right buccal and submandibular abscess in about two years ago; which leads to 4-days hospitalization for antibiotic therapy and also both permanent right mandibular canines, first and second primary molars extraction. The Maximum Mouth Opening (MMO) was measured 10 mm in clinical

examination (Figure 1). The patient had no problems in lateral or protrusive jaw movement radiographic examination (panoramic and CBCT) ruled out the possibility of TMJ ankylosis as a main etiology for this trismus (Figure 2).

The patient did not have any problem in extra-oral examination, also facial motor and sensory nerves were normal too. Regarding the intraoral examination, a fibrous strip band measuring 1 cm<sup>2</sup> × 3 cm<sup>2</sup> was observed in right oral vestibule depth which was assumed to be related to previous infection and abscess history. These bands had a firm consistency and a pale reddish and pink color without any bleeding and tenderness in manipulation.

The results of laboratory tests were normal and it did not show any signs of iron deficiency anemia, however a slight increasing in ESR was just observed. The socioeconomic status of patient’s family was poor; furthermore none of our patient’s parents had the history of addiction or even spicy food consumption. This vestibular fibrous band was considered as the only probable cause of mouth opening limitation and trismus by ruling out the other differential diagnoses such as ankylosis.

**Table 1:** The review of all reported cases of pediatric (below 15 years old) SMF and their treatment plans.

Number	Author (Year)	Patient's age (Gender)	Patient's nationality	No of reported cases	Predisposing factors	Treatment plan	Follow up duration
1	Hayes [17]	04 (F)	Indian	1	Spicy food consumption, Areca-nut products chewing	Systemic and local corticosteroid therapy, Fibrotic tissue surgery, Graft	Not Reported
2	Anil & Beena [14]	12 (F)	Indian	1	Spicy food consumption, Areca-nut products chewing	Placentrex Injection	Not Reported
3	Mundra et al. [19]	08 (F)	Indian	1	Areca-nut products chewing	Microwave diathermy, Local corticosteroids injection, Excision of fibrotic bands, Supportive treatment (vitamins and minerals)	Not Reported
4	Shah et al. [20]	11 (F)	Bangladeshi	1	Areca-nut products chewing	Supportive treatment, Vitamin B12 and Iron injection	Not Reported
5	Yusuf & Yong [23]	12 (F)	Bangladeshi	1	Areca-nut products chewing	Oral physiotherapy	Not Reported
6	Setia et al. [26]	09 (M)	Indian	1	Tobacco and Areca-nut products chewing	Zinc acetate syrup, Ferrous fumarate tablet, Oral physiotherapy, Vitamin A tablet, Vitamin E capsule	15 days
7	Sitheequet et al. [22]	02 (F)	Sri Lankan	5	Areca-nut products and Betel chewing	Not Reported	Not Reported
8	Shirzaii [22]	15 (M)	Iranian	1	Areca-nut products chewing	Intra-lesion corticosteroid injection	1 year
9	Agrawal et al. [9]	09 (F)	Indian	1	Areca-nut products chewing	Placentrex, Betamethasone and Hyaluronidase Injection	Not Reported
10	Chakraborty et al. [2011]	10 (M), 12 (F)	Indian	2	Areca-nut and Gutka chewing	Not Reported	Not Reported
11	Dhariwal et al. [16]	10 (M), 12 (F)	Indian	2	Spicy food consumption, Gutka chewing/Spicy food consumption, Pan masala chewing	Zinc acetate syrup, Vitamin A tablet/Zinc acetate syrup, Vitamin A tablet, Vitamin E capsule	1 month
12	Deshpande et al. [15]	14 (F)	Indian	1	Areca-nut products and Scented tobacco chewing	Medical therapy (vitamin B complex capsule-antioxidant capsule-iron tablet), Oral physiotherapy	3 months
13	Gupta et al. [6]	10 (M), 11 (F)	Indian	2	Spicy food consumption and Supari consumption	Oral physiotherapy	1 month
14	Kumar et al. [18]	11 (M)	Indian	1	Areca-nut products chewing	Oral physiotherapy, Oral multivitamin, Antioxidant, Triamcinolone pomade, Pentoxifylline	Not Reported
15	Duggirala [8]	09 (F), 13 (M)	Indian	3	Spicy food consumption, Areca-nut and Gutka chewing	Zinc acetate syrup, Ferrous fumarate tablet, Vitamin A tablet, Vitamin E capsule/Vitamin A tablet, Vitamin E capsule, Zinc acetate syrup, Corticosteroids injection/Vitamin A tablet, Vitamin E capsule, Zinc acetate syrup, Ferrous fumarate tablet Corticosteroids injection, Rexitidine M forte gel	Not Reported, 15 days, Not reported
16	Kamat et al. [2016]	12 (M)	Indian	1	Raw and Roasted Tamarind seeds chewing	Not Reported	Not Reported
17	Khandelwal et al. [10]	14 (M)	Indian	1	Areca-nut products chewing	Iron tablet, Vitamin A tablet, Zinc acetate syrup, Placentrex injection, oral physiotherapy	2 months
18	Samieirad et al [2017]	10 (M)	Iranian	1	Unknown	Fibrotic tissue surgery, Buccal fat pad graft, Custom made prosthesis, Corticosteroids therapy, Local Triamcinolone, Oral physiotherapy, Supportive treatment, Vitamin B tablet	6 months

The operation was performed under general anesthesia, and the fibrous band excision with passive physiotherapy was done. At the end of this procedure, the mouth opening was significantly increased to 40 mm. The excised area was sutured primarily and the sample sent for histopathologic examination, and also the patient was discharged with home physiotherapy recommendation finally. The patient revisited one month after surgery, surprisingly the relapsed may be taken placed and the MMO was returned to 12 mm. Unfortunately the authors observed wider and greater hypertrophic bands in intraoral examination, it was developed after the surgery.

Focal submucosal fibrosis was diagnosed in histopathologic evaluation report. The section showed mucosal tissue with granulation tissue hemorrhage and focally chronic inflammatory cells infiltration. Diffuse formation of fibrous tissue with inflammatory cells was observed at submucosa layers (Figure 3).

Considering the progressive nature of this lesion, we decided to perform more aggressive surgery for second time. At this stage, all hypertrophic bands were completely removed up to the buccinators muscle layer under general anesthesia. The patient's mouth opening

was achieved up to 40 mm again. After that, the defect area was covered with buccal fat pad (Figure 4). In the aim of relapse prevention and also avoiding the recurrence of the lesion and fibrous bands, a unilateral custom made prosthesis was fabricated (Figure 5). This device was a modification of microstomia protective prosthesis which effectively reported to use in pediatric lip burning cases in order to prevent scars.

The patient was instructed to use this prosthesis part-time for 2 months; however both active and passive physiotherapy was prescribed for him. Also weekly intra lesion Triamcinolone injections were performed for 4 to 6 weeks (Figure 5). As a complementary treatment vitamin B-complex tablets (one tab per day) was recommended for one month.

In aim of ruling out the chewing drug abuse probability, the patient remained under the alert nurse supervision in hospital for 10 days. The regular follow ups were done weekly up to 6 months; fortunately there was no evidence of relapse and mouth opening limitation or even observation of fibrosis band reformation during this period (Figure 5). It is a pity that the patient did not refer for

more follow ups after that because of his parent's socio-economic problems.

## Discussion

It is interesting to note that by searching the Submucosal Fibrosis keyword in the PubMed database, only 17 studies have been found since 1985 up to now, which reported Submucosal Fibrosis (SMF) in children below 15 years old; while 355 articles have been reported this lesion in adults [6,9,14-23].

Table 1 shows the review of all reported cases of pediatric SMF and their treatment plans. These reports indicated a very low incidence of SMF in children below 15 years old which had less than 2.5% incidence rate [6], and also more prevalent in Indian nationality [11,12,18,19]. In addition, the Betel and Areca-nut abusing were reported as the main etiologic factor in most of these case specially in Southeast Asia, India, Sri Lanka, Bangladesh [9,16,26]. According to Arakeri et al. [1] update study in 2017 the surgical treatment is a definitive treatment for pediatric SMF, however it is noticeable that some studies have used some adjunctive therapies such as topical corticosteroid injection in conjunction in children, as well [9,19].

Areca nut is a fruit of Indian Areca (palm tree) found in South and Southwest of Asia which is abused as a chewing material. It can stimulate the gastrointestinal and central nervous system. As a matter of fact, there is a hypothesis that indicates SMF is a multi factorial condition and would be in relationship with dietary habits such as spicy foods as well as drug abuse [27]. In addition to mentioned topical factors, other systemic factors such as anemia, vitamin B deficiency, malnutrition, genetic and immunologic potential are introduced as probable etiologies in recent studies [7,28].

Rajendran et al. [7] presented a case of SMF in a family who did not have chewing habit of Areca nut and this article could support hypothesis of the effect of genetic and immune factors on SMF etiology. In present case report, the patient did not have a history of drug abuse (areca nut) and spicy foods consumption; so low socioeconomic status of his family and malnutrition or even immunological factors could be considered as the main etiology of this lesion.

Generally, SMF is considered as a progressive, pre-malignant and irreversible chronic condition [29]. The incidence of malignancy in patients with this lesion varies from 3% to 7.6% due to recent studies [30-32]. Buccal mucosa is commonly involved in SMF, however the involvement of other oral cavity areas even pharyngeal region has been reported up to now [8,24]. This lesion leads to thick and wide vertical fibrous bands formation in sub-epithelial layer of the buccal mucosa especially in lip region; then microstomia would happen as a consequence, similar to what occurs after children's lips burns.

In the study of Maher et al. [33] there was no relationship between age and incidence of SMF. In general, this lesion is more common in people between 20 to 40 years; though it may also be observed in more broad age ranges such as 2 to 98 years [1,6,8,34]. It is interesting to note that SMF incidence in youth and adolescent groups have been increased recently for unknown genetic and immunological reasons [2,4,5,8]. Despite the fact that this lesion tends to be more dominant in female adults [8]; but the complications in pediatric group would be more prevalent in boys (64.2%) comparing to girls (35.7%) according to articles. There was also a significant relationship between incidence of SMF with frequency and duration of the Areca nut products abuse

[5,8,14-23]. In most of the literatures, SMF has been reported as a bilateral condition [4,14,19,20,23]; however the authors reported here a rare case of pediatric SMF with unilateral fibrosis.

Common SMF clinical and pathological signs and symptoms include limitation of mouth opening and trismus, pale and firm mucosa with wide fibrous bands in submucosal layer [4]. These firm and fibrous bands are most commonly seen in most parts of oral cavity and oropharynx such as the buccal mucosa, soft palate, and the lips mucosal layer respectively [35].

Since the mouth opening limitation or trismus might be also occurred in TMJ ankylosis [6,8,10,23,27], so the proper definitive diagnosis between SMF and ankylosis would be essential to prevent misdiagnosis and mistreatment [1,5,8,14,17]. These are two distinct diseases with different surgical and medical treatment obviously.

There is no approved definitive treatment for SMF as it has high recurrence rate and tends to relapse in most of cases [2,25].

Several conservative treatments were reported to have low success rate in SMF management [5,8]. A review of the literature regarding the treatment of SMF suggests that medical treatments in patients with maximum mouth opening more than 22 mm (MMO>22 mm) might be effective. These medical treatment include antioxidants, micronutrients, minerals, interferon gamma prescription and also steroids, hyaluronidase, chymotrypsin, and placentex submucosal injection [1,3,24]. Intra lesion stem cells injection, active and passive oral physiotherapy and topical heat compress therapy are also known to be effective conservative treatment [6,8]. However, in cases with severe trismus and Maximum Mouth Opening less than 22 mm (MMO <22 mm), surgical treatment is suggested according to literatures [1,2,6,25]. These surgical treatment options include fibromyotomy, coronoidectomy, fibrous band excision and also the defect coverage with different methods such as buccal fat pad, amniotic membranes, alloderms and split thickness skin flaps [5,8].

It should be noted that, the surgical treatment without other conjunctive methods might be ineffective in most of patients, as it might complicate the condition more by creating new scar tissues according to literatures [5,8].

In this present study, our first surgical approach including the fibrous tissue band excision and then only primary defect closure was not enough for this lesion treatment, so the first try did not cause satisfactory outcome and the patient returned with trismus again.

As a result after considering the histologic report and entity of lesion, the wide and deep fibrous tissue was excised and removed completely in the second surgery, then the defect conservatively reconstructed with buccal fat pad flap. Meanwhile, other conjunctive therapeutic options such as intra-lesional injections of triamcinolone, prescription of vitamin B complex tablets and both active and passive oral physiotherapy were also performed in order to achieve better treatment outcomes in this patient.

The authors also used a novel and specific adjunctive device which was a modification of microstomia protective lip burn prosthesis. This device reported to use in pediatric lip burning cases in order to decrease fibrosis and therefore prevent the scars reformation effectively. So considering to relapse prevention and also avoiding the recurrence of the lesion and fibrous bands, a unilateral custom made prosthesis was fabricated which had unilateral pad. By the insertion and seating of unilateral pad to covered defect with buccal fat pad, it

had the advantages of scar tissue prevention and fibrosis reformation inhibition in this region.

The patient was followed up for 6 months, and the tissue healing and his maximum mouth opening after this combined treatment (surgical, medical, prosthetic and physiotherapy) were satisfactory.

## Conclusion

The correct diagnosis and treatment planning in submucous fibrosis patients are considered to be as essential factors to successfully achieve the desirable outcomes. Also, it reduces the morbidity and mortality associated with misdiagnosis or mismanagement of this condition. Maxillofacial surgeons are recommended to be familiar with signs and symptoms of SMF as one of trismus etiological probabilities. It is essential for practitioners to be aware of all treatment modalities for this lesion, as well.

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