

## CASE REPORT

# Novel Treatment Approach of Oral Submucous Fibrosis in a 6-year-old Girl: A Case Report

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### ABSTRACT

**Background:** Oral submucous fibrosis is characterized by stiffness of oral mucosa, blanching and functional limitation, and areca nut predisposition is considered to be one of the main etiological factors. In recent years, there is an increasing prevalence of OSMF in the Indian subcontinent owing to increased consumption of smokeless tobacco products. Very few cases of pediatric OSMF are reported in PubMed literature. Oral submucous fibrosis has a malignant transformation rate of 7–13% and hence, it is important to intervene at an appropriate stage and manage it well in time.

**Aim and objective:** To report a case of oral submucous fibrosis (OSMF) in a 6-year-old Indian girl along with its management and follow-up.

**Case description:** A 6-year-old girl of Indian origin was diagnosed with OSMF and we have used sesame oil pulling as a novel treatment approach and observed good results with long-term follow-up. We have also reviewed PubMed literature for cases of pediatric OSMF reported till date.

**Conclusion:** A timely diagnosis and intervention becomes necessary in pediatric OSMF to improve oral function and prevent malignant transformation.

**Clinical significance:** It is important to report oral potentially malignant disorder (OPMD) cases in pediatric patients and create awareness through health education programs so that parents and children know about the ill effects of consuming tobacco products.

**Keywords:** Areca nut, Oral Fibrosis, Oral submucous fibrosis, Pediatric.

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### INTRODUCTION

Oral submucous fibrosis (OSMF) is an oral potentially malignant disorder (OPMD) described by Schwartz in 1952 as "Atrophica idiopathica mucosae oris" and by Pindborg as "a chronic, insidious, scarring disease of the oral cavity, often with involvement of the pharynx and the upper esophagus".<sup>1</sup> The use of smokeless tobacco in various forms is very popular in the Indian subcontinent and areca nut chewing is considered as the main risk factor for OSMF.<sup>2</sup> It is characterized by reduced movement and depapillation of the tongue, blanching and leathery texture of mucosa, trismus, and inability to eat, associated with the formation of vesicle due to juxtaepithelial inflammatory reaction followed by fibroelastic changes in lamina propria and epithelial atrophy.<sup>3</sup> The prevalence of OSMF in the Indian subcontinent has been estimated to range from 0.2 to 2.3% in males and 1.2 to 4.6% in females, with a broad age range from 11 to 60 years.<sup>4</sup> In recent years, there is an increased prevalence of OSMF in the younger population mainly due to household environment, peer pressure, and attractive pouches of areca nut products. Oral submucous fibrosis has a high malignant transformation rate of 7 to 13% and therefore, its early diagnosis and treatment becomes crucial in pediatric patients.<sup>5</sup> To date, there is no definitive treatment modality and is mainly aimed at symptomatic relief and prevention of its progression to oral cancer. Very few cases of pediatric OSMF have been reported in the literature.

Therefore, we present here a case of OSMF in a young Indian girl of 6 years along with treatment and follow-up. This is the first case of pediatric OSMF wherein sesame oil pulling has been used as a treatment modality and significant improvement evident.

### CASE DESCRIPTION

#### Case History and Presentation

A girl aged 6 years reported with her mother to the Department of Oral Medicine and Radiology of a Tertiary Care Dental Hospital

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**Conflict of interest:** None

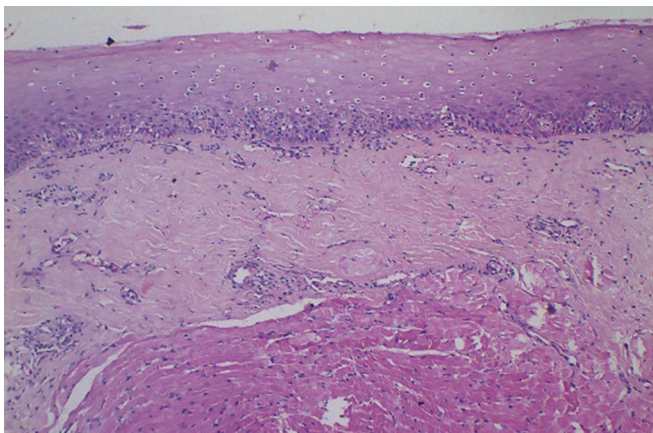
with the chief complaint of inability to open mouth along with a burning sensation of mucosa while taking spicy food for 6 months. The medical history was non-contributory. The patient reported a habit of chewing areca nut 3 to 4 times a day for the past 4 years. A long-standing history of chewing areca nut was present in her grandmother who infused this habit in her granddaughter not knowing the harmful effects of the same. General physical examination was non-contributory. Extraoral examination revealed a restricted mouth opening of 9 mm. Intraoral examination revealed blanching of upper and lower labial mucosa, bilateral buccal mucosa, and palate. There was depapillation of the anterior dorsal surface of the tongue (Fig. 1). The tongue movements were restricted and tongue protrusion was 28.52 mm. Vertically thick fibrous bands were palpable on bilateral buccal mucosa, pterygomandibular raphe, hard and soft palate. Positive history of chewing areca nut along with clinical findings led to a provisional diagnosis of OSMF.

#### Diagnosis

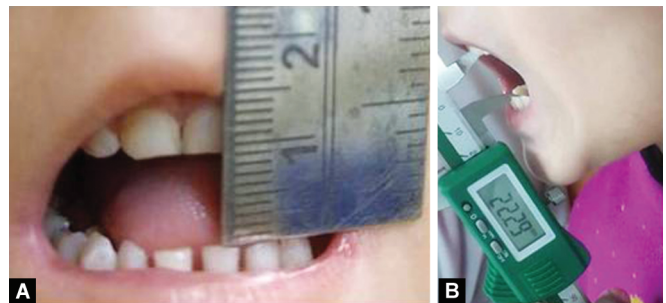
An incisional biopsy was performed from the right buccal mucosa and histopathological examination showed parakeratinized



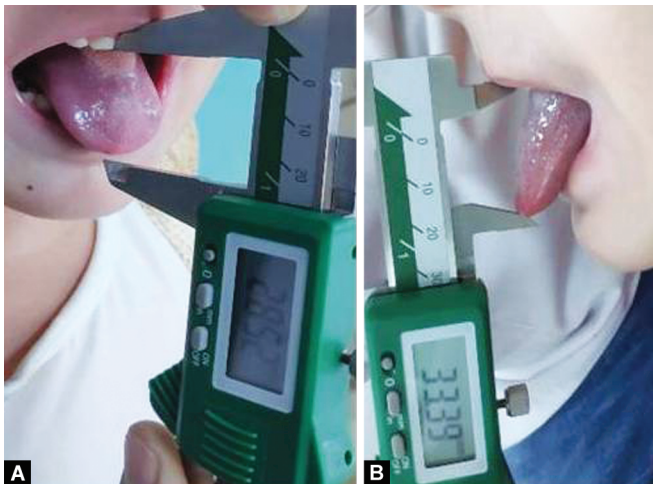
**Figs 1A to C:** Blanching on the right buccal mucosa, left buccal mucosa and palate, and depapillation of anterior dorsal surface of tongue



**Fig. 2:** Microphotograph of patient suggestive of oral submucous fibrosis (10x H&E)



**Figs 3A and B:** Pre- and posttreatment values of mouth opening



**Figs 4A and B:** Pre- and posttreatment values of tongue protrusion

stratified squamous epithelium with a short blunt to flattened rete ridges with stromal tissue composed of densely bundled collagen fibers, and juxtaepithelial hyalinization (Fig. 2). The overall picture was suggestive of highly advanced OSMF.

### Management and Follow-up

The patient and her mother were counseled regarding the adverse effects of chewing areca nut and the potential to cause oral cancer. The patient was advised to stop chewing areca nut. She was treated with sesame oil pulling for 5 minutes daily along with topical application of triamcinolone acetonide (0.1%) and curcumin tablet 300 mg once daily. The patient was given a sesame oil sachet and was instructed to perform oil pulling every day on an empty stomach by emptying the entire 10 mL of oil and swishing it in the mouth for at least 5 minutes without swallowing the oil. This was followed by spitting the oil and brushing the teeth. The patient was refrained from eating anything for at least 30 minutes. The patient was followed up at a regular interval of 1 month up to 6 months followed by follow-up at an interval of 3 months thereafter. She has stopped chewing areca nuts and showed improvement in symptoms. After 3 years of follow-up, the mouth opening is 22.29 mm (Fig. 3) and tongue protrusion is 33.39 mm (Fig. 4). Patient is still under follow-up.

### DISCUSSION

Oral submucous fibrosis is a chronic disease associated with functional limitation and potential for malignant transformation. In recent years, areca nut chewing is thought to be the main etiological factor involved in the pathogenesis of OSMF. Various other factors implicated in its etiopathogenesis are nutritional deficiency, consumption of chilies, genetic predisposition, and immunological process.<sup>6,7</sup> Arecoline, arecaidine, guvacoline, and guacine are the

**Table 1:** Review of pediatric OSMF cases reported in PubMed till date

S.no.	Author and year	No. of cases	Age	Sex	Habit	Mouth opening	Ethnicity	Treatment
1	Hayes et al. <sup>1</sup> 1985	1	4	F	Areca nut	11 mm	Indian	Vitamin supplements, stretching exercises
2	Anil et al. <sup>9</sup> 1993	1	12	F	Areca nut	15 mm	Indian	Submucosal placental extract
3	Mundra et al. <sup>10</sup> 1999	1	8	F	Betel nut	20 mm	Indian	Conservative treatment
4	Yusuf et al. <sup>2</sup> 2002	1	12	M	Areca nut	21 mm	Bangladesh	Mouth opening exercises + conservative treatment
5	Agrawal et al. <sup>11</sup> 2002	1	9	F	Areca nut	16 mm	Indian	Intralesional Injection of placental extracts alone, a combination of dexamethasone and hyaluronidase weekly for 8 weeks
6	Dhariwal et al. <sup>5</sup> 2012	2	10	M	Gutkha	15 mm	Indian	Oral iron supplements, vitamin A, and zinc acetate
			12	F	Paan masala	19 mm	Indian	Oral vitamin A and zinc acetate syrup
7	Deshpande et al. <sup>6</sup> 2013	1	14	F	Areca nut and tobacco	30 mm	Indian	Oral vitamin B complex, vitamin A, E, and C
8	Gupta et al. <sup>12</sup> 2013	2	11	F	Areca nut	14 mm	Indian	Mouth opening exercises
			10	M	Areca nut	13 mm	Indian	Not reported
9	Duggirala et al. <sup>13</sup> 2015	3	9	F	Areca nut	14 mm	Indian	Oral vitamin A, vitamin E for 3 months, and ferrous fumarate tablets for 1 month
			13	M	Paan	22 mm	Indian	Oral vitamin A, vitamin E, and zinc acetate syrup for 3 months + intralesional injection of corticosteroid for 2 months
10	Talla et al. <sup>14</sup> 2019	1	5	F	Betel nut	15 mm	Indian	Topical steroid, multivitamin syrup, diluted tulsi water, physiotherapy

M: Male, F: Female

main alkaloids in the areca nut with arecoline being the most abundant that causes collagen synthesis and fibrosis.<sup>6,8</sup> There is an increasing prevalence of OSMF in the Indian subcontinent mainly due to the increased habit of smokeless tobacco consumption among younger generations. From Table 1, it can be seen that majority of children had a habit of chewing areca nut.<sup>1,2,9,11-13</sup> In the present case series also, the patient had a habit of consuming areca nut. Areca nut has a sweet flavor and because of its mouth freshening action, it is consumed by the children in larger amounts and for longer periods of time.<sup>13,15</sup> Various factors are involved in increasing the consumption of tobacco products among children. Family environment, cultural habits, low awareness, easy availability, low cost, peer pressure, etc., play a major role.<sup>12,13</sup>

There are around 14 cases of OSMF in children in PubMed literature as shown in Table 1. The age group ranged from 4 to 14 years with the first case being reported by Hayes et al. in 1985.<sup>1</sup> In recent years, there appears to be a predisposition of OSMF in adult females. However, no sex predilection is reported in children but some studies have reported areca nut consumption to be more common among males.<sup>16,17</sup> In the review of literature, it can be seen that the majority of children are females with only four males. All except one patient in Table 1 are of Indian origin, thereby implying that OSMF in a pediatric age group is more common in the Indian subcontinent.

Diagnosis can be done on basis of clinical signs and symptoms such as burning sensation on consuming spicy foods, blanching and stiffening of the mucosa, with reduced oral functions such as swallowing, reduced mouth opening, and sometimes accompanied by vesicle formation. A characteristic feature is the formation of fibrous palpable bands on the buccal mucosa, labial mucosa, and soft palate.<sup>13,18</sup> The association of OSMF with the development of oral cancer highlights the importance of patient counseling and education as the first step in its management. With the increasing number of OSMF cases in the pediatric population, it is essential to educate the parents and children about the ill effects of consuming tobacco. This can be done through educational campaigns, schools, newspapers, multimedia, and enforcing legislation to ban the sale of tobacco products to young children.

The goal of the treatment in OSMF is an alleviation of symptoms, improved oral functioning, and prevention of malignant transformation. Treatment ranging from the cessation of habit, medical, and non-medical interventions has been employed for the management of OSMF but so far, a definitive treatment modality has not been able to completely cure the disease and a combination of treatment strategies is used. Cessation of habit forms the most vital step in management. Among medical interventions, micronutrient supplements such as vitamin A, B complex, C and E, copper and zinc supplements, and iron have been used alone

or in combination and the trials have shown good results with marked improvement.<sup>19-22</sup> Antioxidants such as lycopene, beta-carotene, curcumin, spirulina, and aloe vera have been tried with the rationale that these scavenge free radicals, and reactive oxygen species generated due to areca nut.<sup>23</sup> Lycopene has been used for the management of other OPMDs as well and in OSMF, trials have shown improvement in burning sensation and mouth opening.<sup>24,25</sup> Curcumin is also being investigated for its role in preventing fibrosis by decreasing the levels of type I and III collagen in OSMF and has shown good results.<sup>26</sup> Steroid has been used as topical therapy such as betamethasone and triamcinolone or as intralesional injection of dexamethasone in combination with hyaluronidase.<sup>27,28</sup> It is believed that steroids reduce inflammation in OSMF. Various other therapies that have been used include placental extracts, interferon-gamma, pentoxifylline, and isoxsuprine.<sup>22</sup> Surgical interventions are reserved for cases that do not respond to conservative treatment and in advanced cases of OSMF.

Literature on the role of sesame oil pulling and oral health is limited. Patel et al. have used sesame oil pulling for the management of OSMF along with other Ayurveda therapies.<sup>29</sup> They found statistically significant improvement in mouth opening, burning sensation, blanching, and ulceration of the mucosa. Sesame oil according to Ayurveda has the properties of healing, anti-inflammatory, and strength enhancing and along with the technique of oil pulling; it works as a physiotherapeutic measure to improve stiffness.<sup>29</sup> Oil pulling involves sipping, sucking, and swishing the oil in the mouth for 10–15 minutes and is preferably done on an empty stomach.

In the present case, we have employed sesame oil pulling and curcumin for management and we have seen a marked improvement in mouth opening, tongue protrusion, and burning sensation. It was well tolerated by the patient with no side effects. To the best of our knowledge, this is the first case of pediatric OSMF with sesame oil pulling as the management strategy.

## CONCLUSION

A timely diagnosis and intervention becomes necessary in pediatric OSMF to improve oral function and prevent malignant transformation.

It is important to report OSMF cases in pediatric patients and create awareness through health education programs so that parents and children know about the ill effects of consuming tobacco products. Sesame oil pulling and curcumin can be used for the management of OSMF as it shows promising results and has no side effects.

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