No age predilection for a disease like OSMF. A case report of 5-year-old child

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ABSTRACT

Background: Oral Sub Mucous Fibrosis (OSMF) is a clinical entity, well known since the time of Sushruta as 'Vidari'. OSMF is a chronic subtle disease that is associated with significant functional morbidity and an increased risk for malignancy. Generally, OSMF is considered as an adult disease, but recently Indian suburb area has reported incidence of 0.2–1.2 percent in pediatric age group as well. The pathogenesis of the disease is not well established, but is believed to be multifactorial. Areca nut chewing is considered as the major etiology amongst all. Children indulge in habit of chewing areca nuts due to its easy availability of different forms of multicolored attractive areca nut pouches in markets. Other factors involved in the consumption of Areca nut are levels of awareness, household environment, peer pressure, low cost, etc.

Case presentation: A 5-year-old boy reported with the complaint of restricted mouth opening since 3–4 months. He had habit of chewing betel nut, a minimum of 2–3 sachets daily since one and a half years. He also complained of intolerance to spicy food. The oral mucosa was pale, opaque, blanched with a reduced mouth opening of interincisal width being 06 mm. Palate was fibrotic with shrunken uvula.

Conclusion: This Case of OSMF in a 5-year-old boy is reported to be the first incidence of OSMF in a child with least interincisal opening of 06 mm.

1. Introduction

Oral Sub Mucous Fibrosis (OSMF) is well known clinical entity, known since the time of Sushruta as 'Vidari'.¹ In the middle era, the condition was first reported by Schwartz in 1952 who termed it as "atrophia idiopathies (tropica) mucosae oris." Joshi in 1953 described it among Indian population in his report and coined the term "sub-mucous fibrosis of the palate and pillars".² Other names that have been proposed were "diffuse oral submucous fibrosis", "idiopathic scleroderma of the mouth", "idiopathic palatal fibrosis", and "sclerosing stomatitis".¹

OSMF has been described by Pindborg and Sirsat in 1966 as 'a chronic subtle disease affecting oral mucosa and occasionally extending to the pharynx and the esophagus, or associated with vesicle formation. OSMF is characterized by a juxta-epithelial inflammation followed by fibro-elastic change of the sub mucosa with epithelial atrophy that lead to stiffness of the oral mucosa and causing trismus'.¹

The factors which have drawn attention as risk factors for OSMF include areca nut or betel nut chewing habit, vitamin B complex

deficiency, long term iron deficiency and excessive consumption of spicy food that accelerates lysyl oxidase activity, which results in fibrosis, majorly caused by areca nut chewing in any form.³

Generally, OSMF is considered as an adult disease, however an incidence of 0.2–1.2 percent in pediatric age has been reported in India.⁴ In recent years increasing chewing habits of betel and areca nut was observed in many parts of India like Gujarat, Maharashtra, Madhya Pradesh and Bihar.³ Ahmad et al. conducted epidemiological and etiological study in schools and colleges and concluded that 13–50% of children have habit of chew areca nuts in different forms because of easy availability of different multicolored attractive areca nut pouches in markets.⁵ Children indulge in habit of chewing areca nuts due to its easy availability of different forms of multicolored attractive areca nut pouches in markets. Other factors involved in the consumption of Areca nut are levels of awareness, household environment, peer pressure, low cost, etc.

Considering early age of involvement and severity, we report a Case of 5-year-old child with interincisal opening of 06 mm.

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Fig. 1. Intraoral photograph showing restricted mouth opening.

Fig. 2. Intraoral photograph showing pale and blanched oral mucosa.

Fig. 3. Extraoral photograph showing 06 mm of interincisal mouth opening.

1.1. Case report

A 5-year-old boy reported to the outpatient department with the complaint of restricted mouth opening since 3–4 months. (Fig. 1) The patient also complained of intolerance to spicy food. History revealed that the patient had habit of chewing betel nut (sweet supari) since one and a half years (a minimum of 2–3 sachets daily). He started the habit of chewing at age 3.5 years and continued it regularly since then. The child revealed that the habit was inculcated in him by his parents. The parents on being asked disclosed that betel nut chewing helps in reducing the appetite. The parents came from very low socioeconomic

Fig. 4. Extraoral photograph showing 08 mm of interincisal mouth opening followed by treatment of one month.

background and worked on daily wage basis as laborer. Both the parents reported of similar habit and were unaware of the ill effects of betel nut chewing.

On intraoral examination, oral mucosa was pale, opaque, blanched (Fig. 2) with a reduced mouth opening of interincisal width being 06 mm. (Fig. 3) Palate was fibrotic with shrunken uvula. The buccal mucosa was rubbery and inelastic, along with palpable fibrous bands and diffuse white lesion of size approximately 15–20 mm along with multiple diffuse areas of melanotic pigmentation extending from the retromolar area to the deciduous canine. On palpation, all the findings were confirmed. Oral hygiene condition was poor with presence of stains and calculus.

1.2. Classification

According to classification given by More et al., our patient falls under the category of S2M4, where S2: Palpable fibrous bands in buccal mucosa and/or oropharynx, with/without stomatitis and M4: Inter-incisal mouth opening less than 15 mm.⁶

1.3. Differential diagnosis

On basis of clinical examination with difficulty in mouth opening, habit of betel nut chewing and intolerance to spicy food, differential diagnosis may be OSMF and localized scleroderma. However, patient had habit of chewing areca nut and absence of any clinical signs and symptoms suggesting scleroderma, the final diagnosis was confirmed as OSMF.

1.4. Treatment

OSMF occurring at such an early age can cause serious consequence, hence the first approach was to motivate child for cessation of the betel nut chewing habit and parental counseling. Combination of medical therapy like vitamin B complex capsules, antioxidants and iron supplements along with opening exercises by ice-cream sticks was taught to

Table 1
OSMF cases reported in pediatric age group all over the world.

Year of report	Authors Details	Age (Years)	Gender (Male/ Female)	Geographic Location/ Country	Mouth Opening (mm)	Etiology/Habit	Habit Influenced by	Frequency & Duration of Habit
1985	Peter A Hayes ⁹	04	Female	Toronto	11	Areca nut (Paan Supari)	Parents and siblings	3-4 times a week since 2 years
1993	Anil S ¹¹	12	Female	Andaman & Nicobar Islands	17	Paansupari	–	Minimums 1 times daily since 5 years
1999	R.K. Munda ¹²	08	Female	Indore, Madhya Pradesh, India	20	Betel Nut (babusupari)	Working Parents	6 months
2001	Binal Shah ¹⁰	11	Female	Bangladeshi residing in UK	–	Supari and betel quid	Parents has habit of Tobacco	Occasionally since 1 year
2002	H Yusuf ¹³	12	Male	Bangladeshi residing in UK	22	Paan Supari	Chewing	Since 4 years
2010	Sumeet Setia ¹⁴	09	Male	Gaziabad, Uttar Pradesh, India	25	Smokeless tobacco	Parents	1-2 packets daily since 2-3 years
2010	Mohaideen Sitheeqe ¹⁵	2.3	Female	Sri Lanka	–	Areca Nut	–	–
2010	Mohaideen Sitheeqe ¹⁵	03	Male	Sri Lanka	–	Betel with Areca Nut	Grandmother	2-3 per day
2010	Mohaideen Sitheeqe ¹⁵	03	Male	Sri Lanka	–	Betel with Areca Nut	–	1-2 per day
2010	Mohaideen Sitheeqe ¹⁵	03	Male	Sri Lanka	–	Areca Nut	Grandmother	2-3 per day
2010	Mohaideen Sitheeqe ¹⁵	03	Female	Sri Lanka	–	Betel with Areca Nut	Grandmother	2-3 per day
2011	Anuradha Agarwal ¹⁶	09	Female	Bhopal, Madhya Pradesh, India	16	Sweet Supari	Parents	3-4 times dailysince 5-6 years
2012	Richa Dhariwal ¹⁷	10	Male	Haryana, India	15	Gutkha	Colleagues	1 year
2012	Richa Dhariwal ¹⁷	12	Female	Haryana, India	19	Pan masala	Grandmother	7 years
2013	Anshula Deshpande ¹⁸	14	Female	Vadodara, Gujarat, India	30	Areca nut & Tobacco	Socio-economic status	2 packets per day since 1 year
2013	Vinay Gupta ¹⁹	11	Female	Lucknow Uttar Pradesh, India	14	Areca nut	Peer pressure	7 years
2013	Vinay Gupta ¹⁹	10	Male	Lucknow Uttar Pradesh, India	13	Areca nut	Peer pressure	6 years
2014	Shivesh Kumar ²⁰	11	Male	Bareilly, Uttar Pradesh, India	15	Sweet supari	Parents	5-6 packets perday for 1 year
2015	Tulasi Lakshmi Duggirala ²¹	09	Female	Maharashtra, India	14	Sweet supari	Peer pressure	9-10 timesince 3 years
2015	Tulasi Lakshmi Duggirala ²²	13	Male	Maharashtra, India	22	Pan Chewing & Gutkha Chewing	Parents	Pan 2-3 times for 4 years & Gutkha 1-2 times a week
2016	Mamta Kamat ²²	12	Male	Bagalkot, Karnataka, India	21	Roasted Tamarind Seeds	Parents	15-20 seeds, two to three times a day since five years.
2017	Vishal Khandelwal ²³	14	Male	Indore, Madhya Pradesh, India	14	Gutkha	Peer pressure	3-4 times a day since 6 years
2018	Sahand Samieirad ²⁴	10	Male	Iran	10	Unknown	Unknown	6 months
2020	Pratik Kariya (Present Case)	05	Male	Maharashtra, India	06	Betal Nut	Parents	2-3 sachets dailysince 2 years

patient and parents, which helped in increase mouth opening.

On one-month follow up visit, minor amount of correction in mouth opening was observed (inter-incisal mouth opening: 08 mm) and patient now could tolerate spicy food. (Fig. 4) Child had completely quit the habit of betel nut chewing. The child is still under observation.

2. Discussion

OSMF is a chronic subtle disease that is associated with significant functional morbidity and an increased risk for malignancy. The pathogenesis of the disease is not well established, but is believed to be multifactorial. Areca nut chewing is considered as the major etiology amongst all. The major alkaloids present in areca nut are Arecolidina, Arecoline, Arecaidine, Guacine and Guyacoline among which Arecoline, the most abundant alkaloid, may have cytotoxic effects on cells and also promote collagen synthesis.⁷

Histologically, OSMF shows the distinctive features of complete loss of rete ridges with severely atrophied epithelium. Various degrees of epithelial atypia may be present. The underlying lamina propria shows severe hyalinization, with homogenization of collagen. Cellular elements and blood vessels might be reduced.⁸

There are many evidences of reported cases across the globe in pediatric age. Hayes (1985) reported first Case of OSMF in a 4-year-old girl.⁹ No sex predilection is established in children but a small number of studies reported areca nut chewing to be more in boys.⁹ Shah et al. (2001) reported a case of OSMF in an 11-year-old Bangladeshi girl living in the UK.¹⁰ Table 1 shows the details of OSMF patients reported in Pediatric age group all over world till date.^{9–24} Reviewing the literature no case has been reported with an interincisal opening less than 10 mm,²⁴ thus making this case to be the first incident case of OSMF in a child with least interincisal opening of 06 mm.

Definite cure of OSMF is still not known. Therefore, the initial treatment starts with the cessation of the betel nut chewing habit. If this is achieved, the early lesions have a good prognosis and might regress with improvement in symptoms. Several conservative approaches such as Oral Antioxidants, Micronutrients and Minerals, Turmeric, Pentoxifylline, Interferon-gamma, submucosal steroid injections of Dexamethasone, Hyaluronic acid (hyalase), Chymotrypsin, Placental extract (placentrix), milk from immunized cows, Intralesional injections of stem cells, physiotherapy, regular mouth-opening exercises and local heat therapy work with little success if the disease is diagnosed early and termination of habit is achieved completely. Surgical care is indicated in patients with severe trismus, dysplasia or neoplasia. Surgical modalities include Myotomy, Coronoidectomy and simple excision of fibrous bands, mucosal or non-vascularized split thickness grafts.

3. Conclusion

Oral submucous fibrosis is one of the most commonly occurring premalignant conditions in the Indian subcontinent. Number of children with oral submucous fibrosis is increasing in India due to easy and cheap availability of psycho-stimulating products like betel-nut, areca nut and tobacco. Pediatric dentists should be aware of possibility for occurrence of OSMF at young age. Early diagnosis can be instrumental in deciding the course of outcome. This would also be helpful to reduce the morbidity and mortality associated with this condition. In India, lack of awareness and taboos associated with the surgical and interventional treatment modalities, poses a serious need for establishment of a preventive and intervention program for the malicious habit. Increased efforts should be made by societies like Indian Society of Pediatric and Preventive Dentistry (ISPPD), Indian Dental Association (IDA), World Health Organization (WHO) etc. To work towards creating awareness among masses against substance abuse.

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Patient (Parents) consent taken.

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Declaration of competing interest

No conflict of interest.

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