



OPEN ACCESS

Oral submucous fibrosis in children: an alarming condition and challenges in management

Anshul Rai ,¹ Anuj Jain,² Aakash Arora,³ Tejas Motiwale⁴

¹Dentistry, All India Institute of Medical Science - Bhopal, Bhopal, Madhya Pradesh, India

²Department of Trauma and Emergency Medicine, All India Institute of Medical Science - Bhopal, Bhopal, Madhya Pradesh, India

³Oral and Maxillofacial surgery, Shree Bankey Bihari Dental College and Research Centre, Ghaziabad, Uttar Pradesh, India

⁴OMFS, Sri Aurobindo College of Dentistry, Indore, Madhya Pradesh, India

Correspondence to

Dr Anshul Rai;
anshulrai007@yahoo.co.in

Accepted 3 March 2021

SUMMARY

Oral submucous fibrosis (OSF) is a well-documented potentially malignant condition. It affects most commonly the adult patients of India and Indian subcontinent, only few cases affecting children have been reported in the literature. This paper presents three cases of OSF below the age of 12 years and reviews the aetiology, clinical presentation, treatment modalities in children with improved follow-up results. Clinical features like restricted mouth opening, burning sensation and history of betel nut chewing helps in the diagnosis of the patients, which can be confirmed by histopathological examination. Conservative management and oral physiotherapy in children help in improved mouth opening. Counselling the children, their friends and parents also plays a vital role. The present paper highlights that children on a larger scale are using tobacco products and further studies are required with larger sample size.

BACKGROUND

Oral submucous fibrosis (OSF) is a potentially malignant disorder that commonly affects the individuals who have a habit of betel nut quid chewing. This premalignant condition predominantly occurs in the Indian subcontinent with a prevalence rate of 0.5% and a female preponderance with a ratio of 3:1.¹ OSF may transform into oral malignancy, specifically oral squamous cell carcinoma with a malignant transformation rate of 7.6% as highlighted in the study conducted by Murti *et al.*² Other studies in the literature suggests that the malignant transformation rate ranges from 7% to 13%.³

An early diagnosis and subsequent treatment may reduce the risk of malignant transformation of the disease, lowering the risk of fatality. Management of this disease is focused towards the combination of preventive and corrective measures. The preventive measures include counselling the patient, refraining from the habit and regular follow-up. The corrective measures include the medicinal treatment with agents like lycopene, micronutrients, steroids, chymotrypsin, hyaluronidase, turmeric and placental extracts along with oral physiotherapy.¹ Extensive cases even require surgical treatment.

Nowadays, children are becoming more addicted to betel nut chewing habit mainly because of the easy access and decreasing parental control over the child. This condition is increasing with an alarming rate, especially in paediatric group and it needs prompt attention for timely prevention. We present three cases of OSF in children who reported to our

institute located in the central part of India. All the patients were below the age of 12 years and showed excellent results with the conservative management after 3-year follow-up.

CASE PRESENTATION

Case 1

A 9-year-old boy presented with a chief complaint of gradual reduction in mouth opening since the last 6 months (*figure 1*). History of present illness revealed decrease in mouth opening since 1 year with inability to eat and maintain proper oral hygiene. He also complained of burning sensation in the mouth since 1 year while consumption of spicy food and decreased salivation since 7–8 months.

The patient gave a history of tobacco and betel nut chewing, 20–25 times per day since 4 years and used to keep it in buccal vestibule on both sides for about 3–4 min before swallowing.

Clinical examination revealed a maximum interincisal distance of 11 mm. On intraoral examination, labial and buccal mucosa were blanched and inelastic, with vertical and circular fibrous bands palpable on both sides, including the lower lip, upper lip, (*figures 2 and 3*) buccal mucosa and retromolar region. Vestibular mucosa, palatal mucosa and floor of the mouth also showed blanching. Tongue had restricted movements.

Histopathologically, H&E-stained section showed juxtaepithelial hyalinisation and collagen fibres were arranged in separate bundles with dense inflammatory infiltrate.

Case 2

An 11-year-old boy presented with a chief complaint of difficulty in eating and restricted mouth opening (*figure 4*) since 1 year. On asking leading questions, the patient gave a history of burning sensation over buccal mucosa while consuming spicy food since 1 year. The patient had a habit of tobacco and betel nut chewing 15–20 times per day since 5 years. He also had a habit of keeping the quid in the buccal vestibule.

On clinical examination, maximum interincisal distance was 13 mm. Intraoral examination revealed palpable fibrous bands over labial and buccal mucosa along with blanching of palate, soft palate and floor of mouth (*figure 5A–C*). Tongue movements were restricted and the uvula was shrunken. Histopathological examination showed moderately hyalinised collagen in connective tissue stroma. Few areas show thickened collagen



© BMJ Publishing Group Limited 2021. Re-use permitted under CC BY-NC. No commercial re-use. See rights and permissions. Published by BMJ.

To cite: Rai A, Jain A, Arora A, *et al.* *BMJ Case Rep* 2021;**14**:e238718. doi:10.1136/bcr-2020-238718



Figure 1 Nine-year-old patient with restricted mouth opening.

bundles. Inflammatory exudate chiefly consists of lymphocytes and plasma cells.

Case 3

A 12-year-old boy presented with difficulty in mouth opening (figure 6) and burning sensation on consumption of spicy food since last 8 months. Maximum interincisal distance was 11 mm, which has increased significantly after conservative management (figure 7). The patient gave a history of chewing tobacco and betel nut pouches 20–30 per day since 6 years. On intraoral examination, vertical and circular fibrous bands were palpable over lower lip, upper lip, labial and buccal mucosa.

Histopathologically, H&E-stained section showed collagen fibres were arranged in separate bundles with dense inflammatory infiltrate, also showed juxtaepithelial hyalinisation.

INVESTIGATIONS

Incisional biopsy was performed intraorally from the buccal mucosa region under local anaesthesia in all the three patients for the confirmation of diagnosis. Complete blood counts of the



Figure 2 Fibrous bands and blanched labial mucosa of lower lip.



Figure 3 Fibrosis bands seen on upper labial vestibule and mucosa.

patients were done in order to evaluate the haematological status of the patients.

DIFFERENTIAL DIAGNOSIS

Clinically restricted mouth opening in children suggests the initial diagnosis of condylar fracture, temporomandibular joint ankylosis and jaw deformities. But proper history of betel nut and tobacco chewing habits and intraoral examination led to the provisional diagnosis of OSF, which was confirmed following histopathological examination.

TREATMENT: MANAGEMENT PROTOCOL FOR PAEDIATRIC PATIENTS

Treatment plan for all the patients.

1. Discontinuation of the habit with complete cessation of not only areca nut consumption, but also other irritants, such as hot and spicy foods.
2. Mouth opening exercises with wooden spatulas and Heister's jaw opener. In cases where the patients have mobile teeth and/or missing teeth, a modified Heister's jaw opener⁴ can be used.



Figure 4 Restricted mouth opening in case 2 patient.

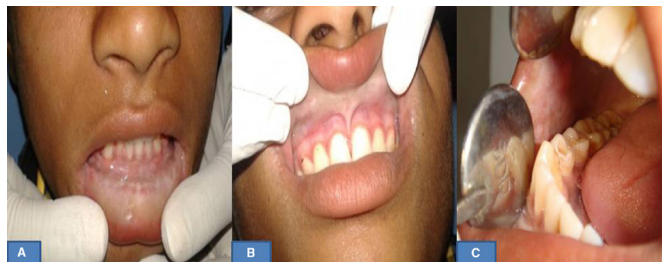


Figure 5 (A) Fibrous bands on lower labial mucosa and vestibule.(B) Fibrous bands on upper labial mucosa and vestibule. (C) Fibrous bands and blanching on buccal mucosa.

3. Antioxidants (tablet Antoxid, once a day for 1 month) and topical application of steroids (Kenacort 0.1% ointment, two times per day for 1 month) and topical application of turmeric paste two times per day was prescribed to the patients.
4. Counselling of the parents and friends regarding the ill-effects and sequelae.

OUTCOME AND FOLLOW-UP

All the patients showed excellent improvement at 3-year follow-up. Their mouth opening increased along with reduction in burning sensation.

Overall, the quality of life of patients improved. Children became less irritable, and regularly attended the school and performed well.

DISCUSSION

The OSF is a disease of obscure aetiology, but it is a known fact that areca nut chewing plays an important role in causing OSF. More than 80% of patients diagnosed with OSF reported with history of chewing areca nut.^{4,5} According to Canniff *et al* the human leucocyte antigen (HLA)-DR typing in OSF patients suggests that there might be an HLA-linked genetic susceptibility for areca nut alkaloids and tannins.⁶



Figure 6 Restricted mouth opening and fibrous bands on lower labial mucosa in case 3 patient.



Figure 7 Improved mouth opening of case 3 patient.

The disease is increasing in the paediatric age group with an alarming rate. Old literature lacks paediatric cases of OSF; however, they are increasing in recent times. The oldest case reported was the case of a 4-year-old girl.⁷ Jain *et al*¹ had conducted a review in 2019 with cases below 10 years of age and presented with a protocol for management of this premalignant condition in paediatric population. Talla *et al*⁸ and Kariya *et al*⁹ both reported a case of OSF in 5-year-old child. Recently, More *et al*¹⁰ conducted an analysis of 36 cases of OSF in the paediatric population. In the present report, the patients were boys aged 9–12 years. All of them were students attending school in the village area, which suggests that other students might also be suffering from OSF. As all the patients gave the history of chewing tobacco and betel nut in a group, it is recommended to carry out a research among students attending school on a larger scale in rural areas of India to identify the quantum of the paediatric population affected with the disease.

OSF is a potentially malignant condition and up to 26% of the patients showed transformation into squamous cell carcinoma,¹¹ which makes it essential to educate the community about the ill-effects of chewing tobacco and betel nut. As there is no cure for the condition, education and prevention seem the only way to reduce the risk of OSF.

It is very difficult to closely monitor patients belonging to paediatric age group. Hence, the role of family members is of paramount importance to keep a close watch on patient's daily activities and counsel them as well as their friends regarding the ill-effects of OSF. Unlike temporomandibular joint ankylosis,¹² the reduction of mouth opening in OSF is gradual. Hence, a close monitoring on child's mouth opening should also be done.

Cessation of patients habits, education regarding the ill-effects of disease and close monitoring of the oral mucosa are the essential components of the treatment of OSF. Surgical intervention becomes essential in severe cases and cases not responding to medicinal treatment.¹³ It is also the duty of schools to put a complete ban on shops that sell tobacco and betel nut to children.

In villages, it is a common practice to eat tobacco and betel nut, it is the duty of elders to not only restrict its use in front of the children, but also to put a complete ban on tobacco and betel nut sale to children in villages. Schools should also take

necessary actions in the form of oral cavity check-up camps, and education programmes for teachers and children regarding the ill-effects of tobacco and betel nut use.

Learning points

- ▶ Oral submucous fibrosis is increasing in the paediatric age group.
- ▶ Early diagnosis gives good results.
- ▶ Proper education should be provided to children to enlighten them of the ill-effects of tobacco and betel nut chewing.
- ▶ Counselling of parents and friends are very important.
- ▶ A long-term follow-up of the patient is very important.
- ▶ Government should make a rule regarding restriction on selling tobacco and betel nut to individuals below the age of 18 years.

Twitter Anshul Rai @Dr Anshul Rai and Aakash Arora @draakasharora

Contributors AR, AJ and AA: Performed the management of the patients, maintained the follow-up of the patients and helped in physiotherapy exercises. TM: Helped in writing the manuscript and performed critical review of the literature. All authors are the guarantors of the manuscript.

Funding The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent for publication Parental/guardian consent obtained.

Provenance and peer review Not commissioned; externally peer-reviewed.

Open access This is an open access article distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work

is properly cited and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>.

ORCID iD

Anshul Rai <http://orcid.org/0000-0002-7571-6447>

REFERENCES

- 1 Jain A, Taneja S. Oral submucous fibrosis in pediatric patients: a systematic review and protocol for management. *Int J Surg Oncol* 2019;2019:3497136:1–6.
- 2 Murti PR, Bhonsle RB, Pindborg JJ, et al. Malignant transformation rate in oral submucous fibrosis over a 17-year period. *Community Dent Oral Epidemiol* 1985;13:340–1.
- 3 Tilakaratne WM, Klinikowski MF, Saku T, et al. Oral submucous fibrosis: review on aetiology and pathogenesis. *Oral Oncol* 2006;42:561–8.
- 4 Rai A, Bholra N, Agrawal B, et al. A modified technique for post operative physiotherapy in edentulous patients. *J Maxillofac Oral Surg* 2012;11:247–8.
- 5 Sinor PN, Gupta PC, Murti PR, et al. A case-control study of oral submucous fibrosis with special reference to the etiologic role of areca nut. *J Oral Pathol Med* 1990;19:94–8.
- 6 Canniff JP, Batchelor JR, Dodi IA, et al. HLA-typing in oral submucous fibrosis. *Tissue Antigens* 1985;26:138–42.
- 7 Hayes PA. Oral submucous fibrosis in a 4-year-old girl. *Oral Surg Oral Med Oral Pathol* 1985;59:475–8.
- 8 Talla H, Ravindranath Reddy PV, Mudavath M, et al. Oral submucous fibrosis in a 5 year old child. *Contemp Clin Dent* 2019;10:679–81.
- 9 Kariya P, Khobragade V, Sura S, et al. No age predilection for a disease like OSMF. A case report of 5-year-old child. *J Oral Biol Craniofac Res* 2020;10:153–7. Apr-Jun.
- 10 More CB, Rao NR, Hegde R, et al. Oral submucous fibrosis in children and adolescents: analysis of 36 cases. *J Indian Soc Pedod Prev Dent* 2020;38:190–9.
- 11 Borle RM, Nimonkar PV, Rajan R. Extended nasolabial flaps in the management of oral submucous fibrosis. *Br J Oral Maxillofac Surg* 2009;47:382–5.
- 12 Jain A, Rai A. Modified temporalis myofascial flap for management of temporomandibular joint Ankylosis: JAIN technique. *J Maxillofac Oral Surg* 2020;42.
- 13 Rai A, Datarkar A, Rai M. Is buccal fat pad a better option than nasolabial flap for reconstruction of intraoral defects after surgical release of fibrous bands in patients with oral submucous fibrosis? A pilot study: a protocol for the management of oral submucous fibrosis. *J Craniomaxillofac Surg* 2014;42:e111–6.

Copyright 2021 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <https://www.bmj.com/company/products-services/rights-and-licensing/permissions/>
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

Customer Service

If you have any further queries about your subscription, please contact our customer services team on +44 (0) 207111 1105 or via email at support@bmj.com.

Visit casereports.bmj.com for more articles like this and to become a Fellow