ONLINE FIRST

CASE REPORT

Idiopathic Oral Submucous Fibrosis in an 8-Year-Old Pakistani Female: A Case Report

Araib Tahir[1*], Samir Azeem[1], Syeda Sadia Kanwal[1], Hijab Fareed Khan[1], Amna Rehman[1]

[1]*Liaquat College of Medicine and Dentistry, Karachi, Sindh-Pakistan*
Correspondence: *dr.araaibtahir@hotmail.com*
doi: 10.22442/jlumhs.2025.01242

ABSTRACT
Idiopathic oral submucous fibrosis in children is an infrequent entity, in particular in regions like India and Pakistan, where the use of areca nut is culturally widespread. Unlike the usual OSF cases due to areca nut or tobacco consumption, idiopathic OSF occurs without any identifiable etiological factors, and poses differential diagnostic and therapeutic challenges. It is a case report of an 8-year-old female child presenting in KKF Dental OPD, a Project by Liaquat College of Medicine and Dentistry from Karachi, Pakistan, who had a minimal mouth opening of 11 mm with bilateral fibrous bands in the buccal mucosa and did not give a history of any areca nut or tobacco habit. Her condition deteriorated due to poor follow-up despite primary management by doing jaw exercises, dietary modification, and applying aloe vera gel. The clinical, psychological, and social implications of OSF in the pediatric age group are reported upon, with an emphasis on early diagnosis and management by a multi-disciplinary team to prevent impairment and malignant transformation. Various medical, surgical, and physiotherapeutic treatments such as corticosteroids, proteolytic enzymes, nutritional support, fibrous band excision, and reconstructive surgery are applied to the patients. This case calls for increased awareness about idiopathic OSF in children, particularly in regions with a higher prevalence level, and calls for the implementation of broad, culture-specific measures to reduce this problem. Etiology will need further clarification to optimize its management protocols for such rare cases of debilitating afflictions.

KEYWORDS: Idiopathic oral submucous fibrosis, pediatric OSF, Karachi, areca nut-free OSF, multi-disciplinary management, rare oral disease.

INTRODUCTION

Oral submucous fibrosis (OSF) is a long-term condition that is marked by progressive mucosal lining fibrosis of the upper digestive system involving the oral cavity, oropharynx, and often the upper third of the oesophagus, leading to spicy food intolerance, a burning feeling in the mouth, xerostomia, and restricted mouth opening, tongue movement, swallowing, or phonation. Idiopathic oral submucous fibrosis in the pediatric population is a rare and alarming phenomenon that needs careful attention. Though the majority of cases of OSF are associated with areca nut-related products, there is a small chunk of cases that occur without any distinguishable etiological aspects and are thus categorized as idiopathic.

The worldwide incidence of (OSF) in children is not well defined in the literature. Even though OSF is largely seen as a potentially malignant disorder associated with chewing areca nut and tobacco consumption among adults[1], there are occasional reports of OSF presentation in children that are seemingly idiopathic. For instance, Talla H 2019[2] also provided a case of OSF in a 5-year-old child, where they described an uncommon presentation in an age group not classically exposed to the usual causative factors of adults. Sitheeque M 2010[3] wrote about depigmentation of the oral mucosa among Sri Lankan preschool children and proposed that it could be an early clinical manifestation of OSF, which suggests that the disease process can start in childhood even in the absence of recognized risk factors like areca nut and tobacco chewing.

In India, the prevalence of OSF is particularly high, with studies indicating that it can occur even in kids. For instance, a case report documented the occurrence of OSF in a five-year-old child, highlighting the upsetting drift towards areca nut consumption among school-aged children[4]. The cultural acceptance of chewing areca nut and related products contributes to the early onset of this condition, which has traditionally been associated with older adults who have long-standing habits of tobacco and areca nut use[4,5]. The systematic review by Jain A 2019[5] emphasizes the increasing frequency of OSF in pediatric patients, noting that a total of ten cases were identified, underscoring the need for greater awareness and preventive measures in this demographic.

The literature shows inadequate data on idiopathic OSF, with only a few case reports and small case series supporting this condition. One of the cases reported was about a 5 year old Indian young girl who suffered with OSF without any history of areca nut or tobacco consumption[6]. The authors emphasized taking OSF under consideration as one of the presumptive diagnoses especially in very young patients and taking appropriate early treatment to prevent its further progression.

Another case report is described from India, highlighting a 9-year-old female patient with idiopathic OSF, underlining the rarity of this condition in the pediatric age group[6]. The etiology of idiopathic OSF is still unknown, and several factors may contribute to it, such as genetic predisposition, autoimmunity, and nutritional deficiencies.

In Pakistan, the condition is concerning. The influence of risk habits, such as areca nut and tobacco chewing, is noteworthy, with studies representing that OSF is among the most common possibly malignant disorders (PMDs) in the country[7,8]. The demographic factors influencing the incidence of OSF in Pakistani children include socio-economic status, cultural practices, and access to education regarding the risks linked with these habits[7]. To date, a review of the literature reveals no reported cases of idiopathic oral submucous fibrosis in Pakistani children.

The clinical implications of OSF in children are enormous. It is not only an impairment of oral function but also carries psychological and social consequences since it shows changes and limitations of functions with a visible impact. Early diagnosis and intervention will be critical to

preventing further advancement of the disease and risk of malignant transformation, which has been as high as 25.77% in untreated cases[9].

The rationale for selecting the case of idiopathic oral submucous fibrosis (OSF) was noticed because of its unusual presentation in an 8-year-old child without any identifiable risk factors like areca or tobacco chewing. The reason behind its selection is that it is a rare condition, and no reported cases of idiopathic OSF in Pakistani children are available. It emphasizes the therapeutic and diagnostic dilemma of treating such presentations in the pediatric age group, which is not well represented in the literature.

CASE PRESENTATION

The patient first presented to KKF Dental OPD, a project of Liaquat College of Medicine and Dentistry, Karachi on February 21, 2024, as illustrated in **Picture 1**. We educated her on several treatment lines, such as exercises, diet modification, and aloe vera gel. So, we advised her several treatments, including mouth exercises, dietary changes, and aloe vera gel application topically. However, she did not maintain follow-up appointments and came to us in December 2024 with further decreased mouth opening, now going down to 11–10 mm. Then, we discussed a few more treatments with her.

Patient Information

Age/Gender: 8-year-old female

Residence: Johar, Karachi

Presentation Date: February 21, 2024

Primary Complaint

Pain in the lower right side of the jaw for the past 2–3 days, along with limited mouth opening, persisting for 3–4 months.

History of present illness

The pain was localized in the jaw area, non-radiating, and related to spicy food intake. No other significant history was found about any illness or acute trauma.

Medical History

The patient also recalled no significant medical history concerning the cardiovascular, gastrointestinal, or respiratory systems. She was never hospitalized or had no history of blood transfusion.

Clinical Examination

Extraoral Findings

No facial asymmetry, swelling, or deformity was seen.

No sign of muscle atrophy or other visible abnormalities.

Intraoral Findings

Initially, the mouth opening was measured at about 15 mm in February 2024, but it was limited to 11 mm on the second visit in December. (**Figure 2**)

Bilateral fibrous bands were observed extending from the tonsillar pillars to the buccal mucosa.

Dental caries in the lower right and left primary second molars (lower D and E). (**Figure 3**)

Investigations

Oral Examination: No observable predisposing habits, such as areca nut or betel nut use, were noted. OPG X-ray, CBCT, and Blood Reports (CBS, ESR) were part of the investigation. (**Figures 4, 5, 6**) shows no significant findings.

Further investigations, such as biopsy and histopathological analysis, are necessary to confirm the diagnosis and exclude other causes like congenital disorders or trauma-related fibrosis, which our team was unable to do because the patient was not cooperative.

Diagnosis

Idiopathic Oral Submucous Fibrosis (OSF).

Classification

According to the classification proposed by More CB et al.[10], (**Table I**) the patient falls into 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.

Table I: Classification of OSF

Clinical staging	S1: Stomatitis and/or blanching of the oral mucosa
	S2: Palpable fibrous bands in buccal mucosa and/or oropharynx, with/without stomatitis
	S3: S2 with palpable fibrous bands in any other parts of the oral cavity
	S4A: S1/S2/S3 with any potentially malignant lesions (Leukoplakia/ Erythroplakia, etc.)
	S4B: Any one of the above stages, along with oral carcinoma
Functional staging	M1: Inter-incisal mouth opening up to or >35 mm
	M2: Inter-incisal mouth opening between 25 mm and 35 mm
	M3: Inter-incisal mouth opening between 15 mm and 25 mm
	M4: Inter-incisal mouth opening less than 15 mm

TREATMENT PLAN

Immediate Management

We advised the patient to address carious teeth through restorative treatment or pulpectomy based on pulp status, due to limited mouth opening it was not favorable for the patient to get treated for carious teeth.

We prescribed analgesics to manage pain in the lower right jaw. In long-term management, we advised physiotherapy to improve mouth opening (e.g, jaw exercises).

Nutritional counseling to ensure a balanced diet and adequate intake of essential vitamins and minerals, especially iron and B-complex vitamins. Monitor for signs of disease progression or complications.

Followup

Regular follow-up visits are conducted to monitor mouth opening and fibrous band progression.

Reevaluate for any underlying systemic or environmental factors contributing to development of OSF. However, the patient was not motivated to attend regular follow-up visits, primarily due to dental anxiety and the long distance between the dental OPD and their residence. Her father further explained that, despite undergoing the prescribed treatment, there was no noticeable improvement in her symptoms**,** making it difficult for him to take time off repeatedly to bring her for ongoing dental follow-ups.

Figure 1: First visit of patient February 2024

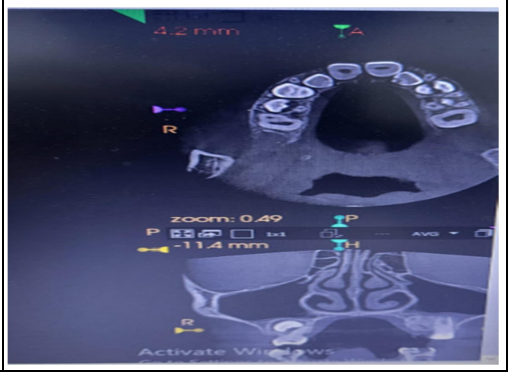
Figure 2: Second visit in December 2024.

Figure 3: Fibrous bands visible in the bilateral buccal mucosa

Figure 4: No significant changes seen on nails or hands

Figure 5: OPG X ray of patient showing no condylar abnormality

Figure 6: CBCT of the patient showing no abnormality of asymmetry or septal defect



DISCUSSION

OSF is treated with different lines of treatment in Pakistan. It is characterized by fibrous bands bilaterally compromising the functional needs, the esthetics and progressive fibrosis of the oral mucosa, leading to severe functional impairment and an increased risk of malignant transformation. The treatment strategies can be largely classified into medical, surgical, and supportive therapies.

Clinical criteria for the diagnosis of OSF have been developed by consensus of histo-pathologists and oral maxillofacial surgeons in a workshop in Malaysia[11]. This clinical criterion consists of the presence of leathery mucosal texture, fibrous bands palpable by hand, loss of appearance of tongue papillae, burning sensation to spicy food in all clinical cases, rigidity of the tongue and blanching of mucosa. These clinical features are found to some extent in all forms of OSF.

Medical Management

The main goal of medical management focused towards improving the symptoms and slow down the disease progression. The interventions used are as follow:

Corticosteroids*:* are the anti-inflammatory drugs, which include dexamethasone and betamethasone. They are prescribed to treat the inflammation and fibrosis of the oral mucosa as intralesional injections[12,13]. However, their long-term efficacy has been questioned; they are used mainly in preservative therapy.

Proteolytic Enzymes*:* Chymotrypsin, collagenase and hyaluronidase are used for their known ability to lower fibrosis, inflammation and tissue repair. They work by the hydrolysis of peptide bonds, which in turn can loosen up fibrous tissue[13].

Nutritional Support*:* Considering the patient situation and related complications with feeding, giving nutritional supplementations such as beta-carotene, zinc, and vitamin A may help improve the general health condition and reduce the complication of OSF[14,15].

Aloe Vera: Some studies have proposed that aloe vera may have valuable properties in managing OSF symptoms due to its anti-inflammatory properties[16,17]. It can help to reduce pain and burning. A study assessed aloe vera as a treatment for oral submucous fibrosis. Seventy-four patients were assigned to two groups: one received systemic and topical aloe vera, and the other received intralesional hydrocortisone and hyaluronidase along with antioxidants. Symptomatic improvement was seen in both groups, with aloe vera providing similar outcomes to conventional treatments. Aloe vera was found to be a safe and effective alternative by the study, and it is suggested that larger-scale, long-term studies be conducted[18].

Surgical Management

In cases where medical management is not enough, surgical intervention may be necessary:

Surgical Excision*:* Excision of the bands is accompanied by coronoidectomy for achieving improvement in the oral functions by regaining the opening of the mouth. Buccal fat pad grafting is often employed to fill defects during surgery, promoting healing and functional recovery[4,19].

Reconstructive Surgery*:* In more advanced cases, reconstructive techniques may be necessary to restore normal anatomy and function[19].

Physiotherapy

Physiotherapy plays a crucial role in managing OSF, particularly in reducing trismus. Specific exercises designed to enhance mouth opening can be beneficial when combined with medical treatment[20,21]. Maintenance or improvement of oral function can be achieved through regular physiotherapy, which is essential for the quality of life of affected individual.

Ayurvedic and Alternative Therapies

Alternate remedies like Ayurvedic treatments, such as Pittashamaka Yoga and gandush oil pulling, have been considered as possible adjunctive therapy in the treatment of OSF[22,23]. While these treatments may not have widespread scientific support, they are part of the cultural background in which OSF is treated.

Multi-disciplinary Approach

We can ensure comprehensive management by merging with multi-disciplinary professionals, including dentists, oral surgeons, nutritionists and physiotherapists. The early stage of the disease can considerably prevent further progression, thus contributing to considerable improvements in the quality of life for patients[24,25].

Limitations

The limitations of this case report reflect several significant limitations. Inadequate investigation was a major issue, given that patient non-cooperation denied crucial diagnostic investigations like biopsy and histopathology, thus compromising diagnostic accuracy. Secondly, the absence of identifiable etiological factors means that the etiology of oral submucous fibrosis in this case remains unknown, making it even harder to understand and manage. Lack of cooperation from patients for follow-up also resulted in disease progression, hence curtailing continued monitoring and effective management. Finally, the conclusions are drawn from one case, limiting generalizability to larger pediatric populations and highlighting the importance of more extensive studies.

CONCLUSION

This case report draws attention to the paucity and clinical relevance of idiopathic oral submucous fibrosis among children, especially in areas such as Pakistan, where no previous cases have been reported. It also emphasizes the singular diagnostic and management challenge of idiopathic presentations and underscores the necessity of increased vigilance among clinicians for early detection and intervention. This case is a worthwhile reminder of the need for multi-disciplinary input and culturally sensitive preventive interventions in managing this crippling disease, while at the same time creating possibilities for additional studies on its pathogenesis and best management.

Conflict of Interest: The author states no conflict of interest.
Financial Disclosure/Grant Approval: No funding agency was involved in this research.
Data Sharing Statement: The corresponding author can provide the data proving the findings of this study on request. Privacy or ethical restrictions bound us from sharing the data publicly.

AUTHOR CONTRIBUTION

Tahir A:	Conception, design and write-up of manuscript
Azeem S:	Final approval of version to be published
Kanwal SS:	Agreement to be accountable of all aspects of work
Khan HF:	Revising it critically
Rehman A:	Final Approval

REFERENCE

1. Zeng L, Peng H, Yu H, Wang W, Duan C, Fang C et al. Expression Profiles of tRNA-derived Small RNA and Their Potential Roles in Oral Submucous Fibrosis. Journal of Oral Pathology and Medicine. 2021;50(10):1057-66.
2. Talla H, Reddy PVR, Mudavath M, Bathina T. Oral Submucous Fibrosis in a 5 Year Old Child. Contemporary Clinical Dentistry. 2019;10(4):679.
3. Sitheeque M, Ariyawardana A, Jayasinghe RD, Tilakaratne WM. Depigmentation of Oral Mucosa as the Earliest Possible Manifestation of Oral Submucous Fibrosis in Sri Lankan Preschool Children. Journal of Investigative and Clinical Dentistry. 2010;1(2):156-9.
4. Talla H, Reddy PR, Mudavath M, Bathina T. Oral Submucous Fibrosis in a 5 Year Old Child. Contemporary Clinical Dentistry. 2019;10(4):679.
5. Jain A, Taneja S. Oral Submucous Fibrosis in Pediatric Patients: A Systematic Review and Protocol for Management. International Journal of Surgical Oncology. 2019;2019:1-6.
6. Agrawal A, Sarkar P, Shigli K. Oral Submucous Fibrosis in a 9-Year-Old Indian Girl. BMJ Case Reports. 2011;2011(sep28):bcr0820114588-bcr.
7. Rashid S, Manzar S, Kazmi F, Shahzad HB, Jan ZA, Qureshi MM. Influence of Risk Habits on Demographic Factors and Its Impact on Oral Submucous Fibrosis. PJMHS. 2021;15(11):3058-61.
8. Siddiq M, Shakeel J, Haider SM, Siddiq S. Mean Lipid Profile Among Patients Suffering From Oral Sub-Mucous Fibrosis Reporting in a Tertiary Care Hospital of Karachi, Pakistan: A Descriptive Study. Annals of Punjab Medical College. 2021;15(1):12-6.
9. Tamang PS, Bajgai GP, Pokhrel HP. Distribution of Oral Submucous Fibrosis Cases and Malignant Transformation Rate Among Bhutanese Patients Treated at Jigme Dorji Wangchuck National Referral Hospital, Bhutan. International Healthcare Research Journal. 2021;4(12):OR7-OR10.
10. More CB, Das S, Patel H, Adalja C, Kamatchi V, Venkatesh R. Proposed clinical classification for oral submucous fibrosis. Oral oncology. 2012;48(3):200-2.
11. Zain R, Gupta P, Warnakulasuriya S, Shrestha P, Ikeda N, Axell T. Oral lesions associated with betel quid and tobacco chewing habits. Wiley Online Library; 1997.
12. Alshadwi A, Bhatia I. Excision of Oral Submucous Fibrosis and Reconstruction With Full Thickness Skin Graft: A Case Study and Review of the Literature. Case Reports in Dentistry. 2012;2012:1-5.
13. Wollina U, Verma S, Ali FM, Patil K. Oral Submucous Fibrosis: An Update. Clinical Cosmetic and Investigational Dermatology. 2015:193.
14. Aggarwal A, Shetti A, Keluskar V, Bagewadi A. Estimation of Serum Beta Carotene Levels in Patients With Oral Submucous Fibrosis in India. Journal of Oral Science. 2011;53(4):427-31.
15. Dhariwal R, Mukherjee S, Pattanayak S, Chakraborty A, Ray J, Chaudhuri K. Zinc and Vitamin a Can Minimize the Severity of Oral Submucous Fibrosis. BMJ Case Reports. 2010;2010(may11 1):bcr1020092348-bcr.
16. Anuradha A, Patil B, Asha V. Evaluation of Efficacy of Aloe Vera in the Treatment of Oral Submucous Fibrosis – A Clinical Study. Journal of Oral Pathology and Medicine. 2016;46(1):50-5.
17. Shanbhag VKL. Aloe Vera in the Management of Oral Submucous Fibrosis. Oncobiology and Targets. 2017;04:1.

18. Anuradha A, Patil B, Asha VR. Evaluation of efficacy of Aloe vera in the treatment of oral submucous fibrosis–a clinical study. Journal of Oral Pathology & Medicine. 2017;46(1):50-5.
19. Rajan B, Ebenezer V, Kumar B, Muthumani. Surgical Management of OSMF With Coronoidectomy and Buccal Fat Pad Replacement - A Case Report. Biosciences Biotechnology Research Asia. 2014;11(2):883-6.
20. Nidhi N, Srikrishna K, Kumar V. Role of Physiotherapy in the Management of Oral Submucous Fibrosis: A Case Control Study. International Journal of Contemporary Medical Research [Ijcmr]. 2019;6(1).
21. Chitlange NM. Physiotherapeutic Approach in Oral Submucous Fibrosis: A Systematic Review. Cureus. 2023.
22. Nayak A, Fiaz S, Bhatnagar SS. Ayurvedic Management of Oral Submucous Fibrosis -a Conceptual Study. Irjay. 2021;04(03):123-8.
23. Solanki V, Unadkat RS, Mehta CS, Baraiya HP, Bineesh EP. An Ayurvedic Approach on Oral Submucous Fibrosis and Its Management – A Review. Asian Pacific Journal of Health Sciences. 2022;9(4):117-22.
24. Rao NR, Villa A, More CB, Jayasinghe RD, Kerr AR, Johnson NW. Oral Submucous Fibrosis: A Contemporary Narrative Review With a Proposed Inter-Professional Approach for an Early Diagnosis and Clinical Management. Journal of Otolaryngology - Head and Neck Surgery. 2020;49(1).
25. Khalam SA, Kumar L, Zacariah RK, Kurien NM, Raj P. Oral Submucous Fibrosis: A Clinical Review. Madridge Journal of Dentistry and Oral Surgery. 2017;2(1):28-30.