

**DURATION OF CONVERSION OF ORAL SUBMUCOUS FIBROSIS INTO
MALIGNANCY: CASE REPORT AND LITERATURE REVIEW**

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ABSTRACT

Oral submucous fibrosis (OSF) is a disease of oral mucosa that has high risk for conversion into malignancy i.e. OSF is a potentially malignant disorder. It is a multi factorial disease but most commonly associated with areca nut chewing. Clinically it presents with restricted mouth opening, burning sensation on hot and spicy food consumption, blanching and fibrosis of the oral mucosa. Histologically it shows juxtaepithelial inflammation and progressive fibrosis in the lamina propria and deeper connective tissue. Here we report a case that was converting from OSF to malignancy in a very short time period i.e. 6 months just because of negligence of patient regarding cessation of habit and treatment.

KEYWORDS: Oral submucous fibrosis, areca nut, malignancy, potentially malignant disorder.

INTRODUCTION

OSF was first described by Schwartz^[1] in 1952 as atrophica idiopathica mucosa oris. Joshi subsequently used the term oral submucous fibrosis (OSF) in 1953 for the disease and in the literature for the first time; the cases were reported from Bombay^[2] and Hyderabad^[3] amongst Indian population. Precancerous nature of OSF was first postulated by Paymaster in 1956. Oral submucous fibrosis (OSF) is a disease of oral mucosa that has high risk for conversion into malignancy i.e. OSF is a potentially malignant disorder. Gupta et al^[4] (1980) reported 2.3% malignant transformation of OSF, According to Pindborg et al^[5] (1984) the rate of conversion of OSF in to malignancy is 4.5%. However Murti et al^[6] (1985) reported 7.6% malignant transformation rate of OSMF in his study. These studies reflected that the malignant conversion of OSF increases with years.

Though conversion rates have been studied in the past there are no recent studies to assess if the conversion rates have changed over the years. Interestingly there are no reports or studies evaluating the duration taken for conversion of OSF to malignancy. It could be hypothesized that this would depend on a multitude of factors like other associated habits, patients age, other associated systemic diseases, duration of areca nut

chewing habit as well as other habits, the maximum mouth opening etc.

We present here a case report in which the approximate duration of conversion has been documented and could serve as basis for future studies on duration of malignant transformation of OSF.

CASE REPORT

Case history and examination

A 50 years old male patient reported with chief complaint of restricted mouth opening since 5 to 6 years. Patient had history of burning sensation on spicy and hot food consumption since 10 years and history of Guthka (a commercial product of betel nut) chewing for 30 years, 8-10 times per day, no other relevant medical, surgical and social history was present.

Physical examination reveals thin build (fig 1a), other vitals were in normal range. On extra oral examination mouth opening was restricted (fig 1b), maximum mouth opening and maximum tongue protrusion were 7 mm (fig 1c), and 17 mm (fig 1d) respectively. Patient had difficulty in blowing cheeks and had 1mm cheek flexibility. Sub mandibular lymph nodes were palpable and mobile.

Intraoral examination revealed tender erythematous atrophic epithelium (fig 1e and 1f), blanching on

application of pressure throughout the oral mucosa, circumoral fibrosis i.e. in the labial and Commissural mucosa, vertical fibrous bands were palpable in bilateral buccal mucosa, pterygomandibular raphae area, palatal mucosa, faucial pillars, floor of mouth. Marble appearance was seen on buccal mucosa (fig 1e).



Fig 1a-Frontal view.

Fig 1b-Restricted mouth opening.

Fig 1c-Maximum mouth opening 7 mm measured by vernier caliper in between incisal edges of 11 and 41.

Fig 1d- Maximum tongue protrusion 17 mm measured by vernier caliper in between incisal edge of 11 and tip of tongue on maximum protrusion.

Fig 1e- Erythematous atrophied labial mucosa and marble appearance of buccal mucosa.

Fig 1f- Erythematous atrophied labial mucosa and supra erupted 18.

On the basis of history and clinical examination provisional diagnosis of oral submucous fibrosis (OSF) was made. The patient was counselled regarding cessation of habit. On routine blood investigation hemoglobin (HB) was 7.1 gm/dl, other blood findings were in the normal range.

Patient was referred to the general medicine department for the management of hemoglobin (HB) concentration and oral and maxillofacial surgery department for the surgical management of the OSF as the patient had very less mouth opening. Patient had refused the treatment because of the fear of surgery.

After 6 months patient again reported with complain of swelling on the left side face since 15 days. Patient had history of tobacco chewing since last 1 month.

Extraoral examination reveals diffuse swelling on left side of face, tender, soft in consistency, size was about 5x4 cm, anteriorly extended up to left corner of mouth posteriorly blended with the body region of mandible, superioinferiorly extended superiorly from an imaginary line joining the left corner of mouth to tragus of left ear to inferiorly up to inferior border of mandible (fig 2a). Left side sub mandibular lymph nodes were enlarged, fixed, non tender, multiple in number. The greatest dimension of lymph node was around 1.5 cm. right side sub mandibular lymph node was mobile.



Fig 2a- Extraoral examination reveals diffuse swelling on left side of face anteriorly extended up to left corner of mouth, superioinferiorly extended superiorly from an imaginary line joining the left corner of mouth to tragus of left ear to inferiorly up to inferior border of mandible

Fig 2b- Intraoral examination revealed the ulceroproliferative growth of approximate size 5x3 cm with creamy white color sloughed over the surface involving buccal mucosa from the corner of mouth to posterior buccal mucosa and retro molar pad area on left side, vestibular mucosa and attached Gingiva in respect to 33, 34,35,36,37 and 38.

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Differential diagnosis, investigations and treatment

On the basis of history and clinical examination provisionally the lesion was diagnosed as malignancy. Differential diagnoses were squamous cell carcinoma, necrotizing sialometaplasia, low grade mucoepidermoid carcinoma. Complete Blood Count (CBC), Renal Function Test (RFT), Liver Function Test (LFT) and Ultrasonography (USG) were advised. HB was 9.5 gm% (13-18 gm%), Total leukocyte count (TLC) was slightly raised i.e. 14,400/cu.mm (4500-10,000/cu.mm), MCV, MCH, MCHC and PCV were reduced viz. 74.44 fl (80-100 fl), 19.43 pg (27-37 pg), 20.10 gm/dl (30-36 gm/dl) and 36.4% (40-54%) respectively. Hematological findings were suggestive of Mild Dimorphic

Hypochromic Anemia and Leukocytosis. RFT AND LFT were normal.

Ultrasonography reveals there is evidence of 4.9x3.4 cm heteroechoic predominantly hyperechoic mass lesion noted in left cheek extending deep into buccal mucosa. The lesion shows few anechoic cystic areas within. Lesion shows internal vascularity within. There is evidence of few, reactive, non necrotic lymph nodes are noted, measuring 1.0 cm in level II region, 1.0 cm in level IV region of right lobe of thyroid and 1.4 cm in level IV region of left lobe of thyroid. These USG findings were suggestive of left oral mass lesion and bilateral cervical lymphadenopathy. Biopsied tissue was taken from the lesion and sent for histopathology, it was suggestive of moderately differentiated squamous cell carcinoma.

Patient was admitted for surgery. Before surgery patient was stabilized by given medications as follows: Tablet combiflam STAT, Tab Pregabalin 75 mg with sip of water, Injection Monocef 1.2 gm bid, Injection Metro 100 ml tds, Injection DynaparAQ tds, Injection Rantac 50 mg bid, injection Emset 4 mg bid, NEB Doulin + Budecort qid, Bro- zedex Syrup 5 ml tid, Injection Clexane 0.4 ml Subcutaneous and 2 point Pack Cell Volume.

Primary tumor removal with left hemimandibulectomy and modified radical neck dissection (MRND) of left side were advised. Left hemimandibulectomy measuring 7 cm along with alveolus with attached buccal mucosa measuring 5x4 cm was done and overlying skin measuring 7x7 cm with a nodule of size measuring 4x3 cm were removed. After dissection there was a grey white ulceroproliferative tumor measuring 4.4x4x3.2 cm on the left lower gingivobuccal, gingivolingual sulcus and buccal mucosa. The maximum tumor thickness was 3cm and that's abuts the overlying skin. Attached level IB with salivary gland measures 3x2x1 cm, lymph nodes were removed largest was measured 1.3 cm.

Anterior and posterior bony cut margins were shaved. Anterior posterior, lingual, floor of mouth and skin cut margins were sent for histopathology. Fibular bone graft was used for left mandibular reconstruction with bone plates and Pectoralis Major Myocutaneous Flap (PMMC) was placed for reconstruction of the soft tissues.

Histopathological examination suggested squamous cell islands and few keratin pearls shows by arrow in fig 3 suggestive of moderately differentiated squamous cell

carcinoma and dense fibrosed connective tissue suggestive of oral submucous fibrosis (fig 3). Histological TNM staging was pT4aN0Mx. Paresthesia in the lower left side of lip was the postoperative complication.

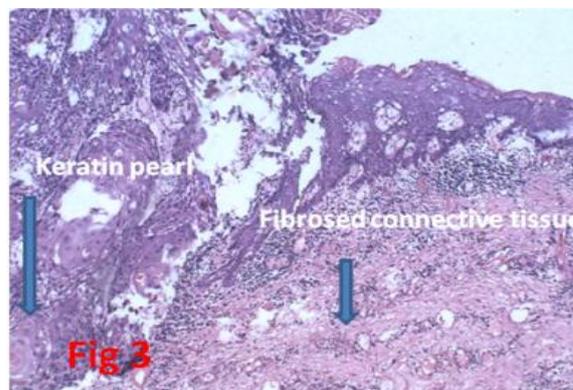


Fig 3: Reveals squamous epithelial island with few keratin pearl and densely fibrosed connective tissue.

Outcome and Follow up

No recurrence was seen after 6 months, patient is under regular follow up after every three months.

DISCUSSION

Worldwide oral cancer is sixth most common cancer having different geographic distribution, frequently seen in Indian and south-east Asian countries.^[7] Globally it is accepted that OSF is an Indian disease^[7]. Most commonly malignancy seen in oral mucosa is Oral Squamous Cell Carcinoma^[8] (OSCC). In our case report also the biopsy report was of moderately differentiated squamous cell carcinoma. Oral cancer in India is mainly due to chewing habits of various commercial products of areca nut and tobacco.^[9] In our case report patient when reported for the first time had the Guthka (commercial areca nut preparation) chewing habit and proper counseling regarding habit cessation was given to patient. After that he started tobacco chewing. Prediction of W.H.O regarding death cause by tobacco in India may exceed 1.5 million annually by 2020.^[10] OSMF has one of the highest rates of premalignant transformation among all premalignant lesion and condition.^[11]

But in our case report patient develops squamous cell carcinoma in the background of OSF in a shorter period of only 6 months (duration was counted from the initial diagnosis when patient was reported OPD for the first time). That is much lesser than earlier studies reported (table 1).^[12,13,14,15]

Table: 1.

Year	Author	Mean time of malignant transformation of OSF (months)	Reference
2017	Yang et al.	30	[12]
2013	Lian et al.	37.42	[13]
2007	Hsue et al.	52.3	[14]
2014	Wang	65.39	[15]

Proposed justification for increase prevalence of OSF^[16] are easy availability of tobacco and areca nut product, cheaper in cost, multicolored attractive packaging, low socio-economic status could be the reason for the use of these products, promotion of these products as a breath fresheners could be the reason of attraction, increased use of these product because very poor awareness regarding disease caused by these products and during many occasion in India areca nut distributed for chewing as a social, cultural and religious belief.

Use of alcohols, smoking, chewing tobacco and chronic irritation of atrophied mucosa by root stump or sharp cusp or faulty restoration could be the reason for malignant transformation of Oral Submucous Fibrosis. More studies are needed for justification of the above statement.

For control of epidemics of OSF and its conversion into malignancy following action could be taken.^[12] Tobacco and areca product should be ban for marketing by government, screening of the patients should be done on mass level for early detection of disease and prompt treatment and increase public awareness regarding deadly effect of the tobacco and areca nut products by television, news papers, pamphlets, radio, street plays, skits, face to face campaigning.

CONCLUSION

OSF is a chronic debilitating symptomatic disease can easily be diagnosed at earlier stage so we can take earlier precautions i.e. proper counseling and awareness regarding the ill effect of tobacco and areca nut chewing and treatment before its progression into advanced stages or in malignancy.

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