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Article *in* Oral and Maxillofacial Surgery · March 2022

DOI: 10.1007/s10006-021-00971-x

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ORIGINAL ARTICLE

Role of coronoidectomy and temporalis myotomy in surgical management of oral submucous fibrosis

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Received: 15 November 2020 / Accepted: 5 May 2021

Abstract

Purpose The involvement of temporalis muscle fibers by oral submucous fibrosis (OSMF) and the procedure of coronoidectomy and temporalis myotomy in the surgical treatment protocol for the disease is a controversy. The primary objective of this study is to evaluate the histopathological changes in temporalis muscle fibers in patients undergoing surgical treatment for OSMF and to authenticate the importance of temporalis myotomy and coronoidectomy in surgical treatment protocol.

Method A 3-year prospective study was conducted to assess the histopathological changes in temporalis muscle in surgically treated OSMF cases. The predictor variables were drawn from demographic characteristics (age and gender) etiology, and mouth opening. The outcome variables were histopathological assessment of temporalis muscle fibers for parameters suggestive of degenerative changes and fibrosis changes at cellular level.

Results Out of 56 patients, 30 patients were had surgical intervention. Twenty-eight (93.3%) were male and 2 (6.6%) were female with a ratio of 14:1. Histopathological examination of temporalis muscle fibers revealed hyalinization of muscle fibers in 80% of the patients followed by loss of striation (73.33%), fragmentation (60%), nucleus internalization (33.33%), infiltration of macrophages and other inflammatory cells (20.67%), multiple nuclei (20%), and swollen muscle fibers (6.67%). Mean preoperative mouth opening was 12.4 and post-operatively 41.3 mm on 1-year follow-up and this was stable on further follow-up.

Conclusion The results of this study suggest involvement of temporalis muscle with disease itself and the justification for coronoidectomy and temporalis myotomy in the surgical protocol was established.

Keywords Oral submucous fibrosis · Oral oncology · Temporalis · Coronoidectomy · Nasolabial flap · Buccal fat pad

Introduction

Oral submucous fibrosis (OSMF) is "a slowly progressive chronic fibrotic disease of the oral cavity and oropharynx, characterized by fibroelastic change and inflammation of the mucosa, leading to a progressive inability to open the mouth, swallow or speak" [1]. It is referred to as a premalignant condition of the oral cavity. Areca nut chewing is common in Southeast Asia and is the etiology for this disease [2]. Affected patients usually present with a chief complaint of inability to open the mouth, burning sensation, pale and rigid oral mucosa, restriction in movements of tongue, shrunken uvula, and dysphagia. These changes are due to marked submucosal fibrosis, reduction in vascularity, and epithelial atrophy [1].

The major concern in advanced stages of disease is severe trismus which has serious health implications such as malnutrition, alteration in speech, and inability to maintain proper oral hygiene. Trismus is due to juxta-epithelial

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Published online: 15 May 2021

hyalinization and fibrosis in the oral mucosa which clinically manifests as stiffened mucosal surfaces [3]. This subsequently causes degeneration, fibrosis, and scarring of the masticatory muscles, further reducing the mouth opening. Resultant hypomobility of the mandible can affect the temporomandibular joint and its adjacent structures which can manifest as internal derangement or ankylosis. Inspection of oral cavity for malignant changes would not be possible in such patients due to compromised access for examination. In very advanced cases, fibrosis can extend into the laryngopharynx, esophagus, and Eustachian tubes, leading to complications like dysphagia and hearing impairment [4].

The surgical procedures in the treatment of OSMF are still a controversy. The need to do coronoidectomy and temporalis myotomy is debated in terms of whether the OSMF disease extends to involve the temporalis muscle or is it due to the secondary fibrosis changes in the muscle. There is no literature available to demonstrate the probable changes in the temporalis muscle fibers in OSMF patients. The primary objective of this study is to evaluate the histopathological changes in temporalis muscle fibers in patients undergoing surgical treatment for OSMF and to authenticate the importance of temporalis myotomy and coronoidectomy in surgical treatment protocol. There is a gap in the literature on the histopathological changes in temporalis muscle fibers in OSMF patients so far.

Materials and methods

A prospective study was conducted at our tertiary craniofacial center from 2017 to 2020. Out of all the OSMF patients who reported to the department, patients of age group 20–60 with groups III and IV OSMF (Andrade and Khanna Classification) [4] and mouth opening less than 20 mm were included in the study group. Patients showing dysplastic features were excluded from the study. The patients diagnosed with OSMF group I and group II were given conservative treatment. Valid consent was obtained from all surgically treated patients. Institutional Review Board (IRB) permission was obtained and has followed Helsinki Declaration guidelines in this study.

A detailed case history was obtained (Table 1) including the demographics, habits, and mouth opening followed by thorough clinical examination. Mouth opening was measured as the inter-incisal distance using a divider and measuring scale. Patients satisfying the inclusion criteria underwent surgical treatment with our standard surgical plan which included fiberoptic nasotracheal intubation, bilateral fibrotomy, extraction of impacted third molars [2, 5], bilateral coronoidectomy and temporalis myotomy (Fig. 1), and reconstruction of fibrotomy defect using nasolabial flap or buccal fat pad [6]. Forceful mouth opening was done intra-operatively using Ferguson's mouth prop, and the patient was put on aggressive mouth opening exercises post-operatively using tongue depressors as well as manually with finger pressure. Temporalis muscle fibers were excised during surgery as a surgical protocol of the treatment, following which these muscle fibers were subjected for histopathologic examination to evaluate the changes like loss of striation, fragmentation, nucleus internalization, presence of multiple nuclei, hyalinization, and macrophage infiltration [7–10]. The data obtained were tabulated and evaluated to quantify the changes in temporalis muscle fibers in these patients. All the surgeries were performed by the same surgeon and the same pathologist evaluated all the samples.

Results

Out of 56 patients who were diagnosed with oral submucous fibrosis, 30 (53.57%) patients who satisfied the inclusion criteria underwent surgical treatment, which comprised the study group.

The age group of the patients ranged from 28 to 55 years with a peak incidence in 30–39 years (n=14, 46.66%). Men were more affected than females with a ratio of 14:1. Seventy-three percent of the study patients were areca nut chewers with or without concomitant tobacco usage.

Histopathological examination of temporalis muscle fibers showed hyalinization of muscle fibers and was the most observed changes (80%), followed by loss of striation (73.33%), fragmentation of muscle fibers (60%), nuclear internalization (33.33%), infiltration of macrophages and other inflammatory cells (2.67%), multiple nuclei (20%), and swollen muscle fibers (6.67%) (Table 2, Fig. 4).

In 24 patients (80%), the reconstruction of fibrotomy defect was done using nasolabial flap and in 6 patients (20%) buccal fat pad to reconstruct the defect (Table 1).

Preoperative mouth opening of the patients ranged from 5 to 20 mm with a mean mouth opening of 12 mm (Fig. 2). Mouth opening increased to 32–44 mm with a mean of 39.53 mm 3 months post-operatively and 35–45 mm on 1-year follow-up with a mean of 41.33 mm (Table 1, Fig. 3). No reduction in mouth opening was noted in their subsequent follow-ups up to 18 months.

Discussion

OSMF, initially described by Schwartz in 1952 [11], is a condition commonly seen in Indian subcontinent. In India, the prevalence of OSMF was found to be 0.2–5% as reported by Pindborg et al. [12]. It is a potentially malignant disorder with an appraised incidence of the malignant transformation to be 2–8% to squamous cell carcinoma (SCC) [11,

Table 1 Demographic statistics and treatment outcome

Patient	Age (years)	Sex	Mo (mm)	Habits	Stage	Recon	Mo 3 M (mm)	Mo 1 year (mm)
1	34	Male	7	Areca nut + Tobacco	IV	Nasolabial	40	44
2	42	Male	20	Areca nut	III	BFP	42	45
3	38	Male	6	Gutka	IV	Nasolabial	42	42
4	50	Male	9	Areca nut	IV	Nasolabial	39	43
5	48	Female	18	Areca nut	III	BFP	38	36
6	33	Male	18	Gutka	III	Nasolabial	39	40
7	34	Male	14	Areca nut	IV	Nasolabial	43	45
8	55	Male	6	Areca nut + Tobacco	IV	Nasolabial	38	38
9	43	Male	11	Areca nut + Tobacco	IV	Nasolabial	44	44
10	28	Male	5	Gutka	IV	Nasolabial	38	40
11	35	Male	18	Areca nut + Tobacco	III	BFP	42	42
12	30	Male	9	Areca nut	IV	Nasolabial	41	43
13	45	Male	16	Areca nut + Tobacco	III	Nasolabial	40	42
14	52	Male	14	Areca nut	IV	Nasolabial	32	35
15	30	Male	15	Gutka	III	Nasolabial	35	38
16	33	Male	18	Areca nut + Tobacco	III	BFP	42	43
17	48	Male	15	Areca nut + Tobacco	III	Nasolabial	42	42
18	51	Male	9	Areca nut	IV	Nasolabial	44	45
19	37	Male	20	Gutka	III	BFP	40	42
20	46	Male	12	Areca nut	IV	Nasolabial	39	39
21	48	Male	15	Gutka	III	Nasolabial	38	39
22	35	Male	17	Areca nut + Tobacco	III	Nasolabial	42	40
23	54	Female	7	Areca nut	IV	Nasolabial	44	43
24	51	Male	9	Areca nut	IV	Nasolabial	35	38
25	35	Male	14	Areca nut + Tobacco	IV	Nasolabial	38	42
26	46	Male	13	Areca nut	IV	Nasolabial	40	44
27	39	Male	12	Areca nut + Tobacco	IV	Nasolabial	43	42
28	28	Male	10	Areca nut	IV	Nasolabial	40	42
29	31	Male	18	Gutka	III	BFP	36	40
30	33	Male	16	Gutka	III	Nasolabial	35	36

MO, mouth opening; BFP, buccal fat pad; Recon, reconstruction; M, month

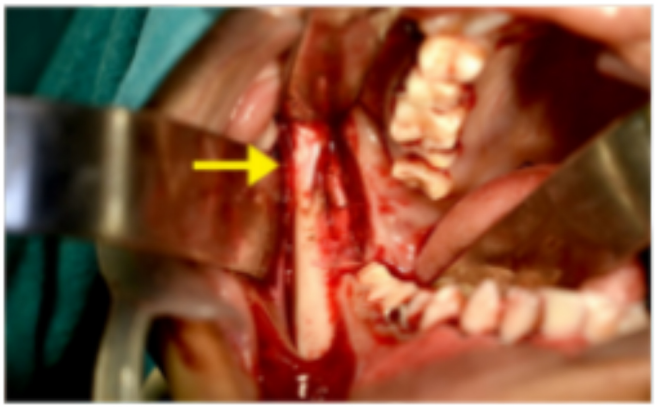

Fig. 1 Intra-operative photograph of temporalis muscle attached to the coronoid process

Table 2 Histopathological changes noted in temporalis muscle fibers

Sl no	Histopathological findings	Number of positive muscle fibers	Percentage (%)
1	Loss of striation	11	73.33
2	Fragmentation	9	60
3	Nucleus internalization	5	33.33
4	Multiple nuclei	3	20
5	Swollen muscle fibers	1	6.67
6	Hyalinization	12	80
7	Infiltration of macrophage and other inflammatory cells	4	20.67

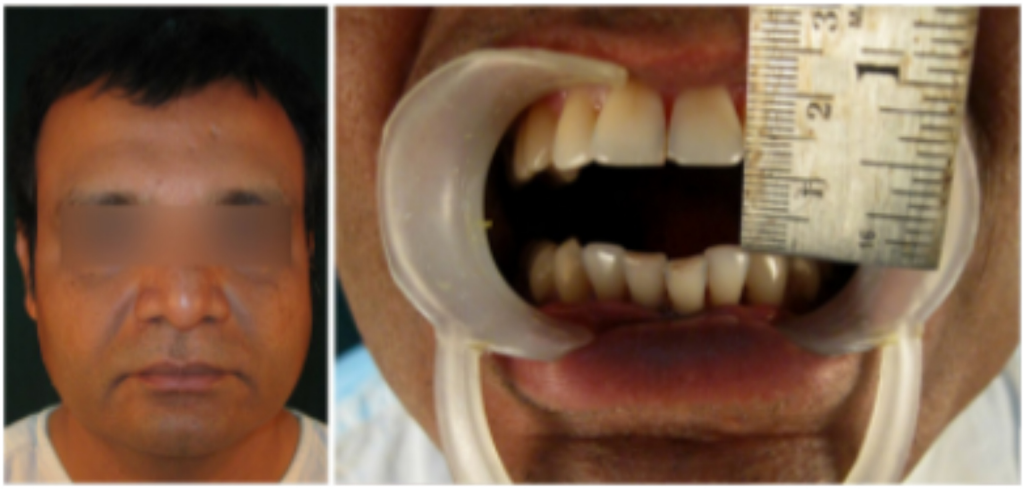

Fig. 2 Preoperative photograph showing restricted mouth opening

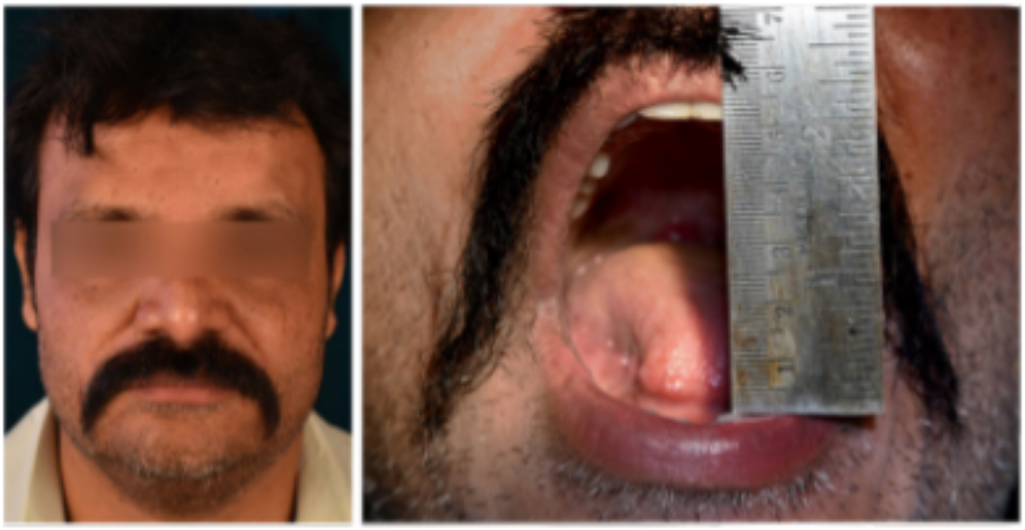

Fig. 3 1-year post-operative follow-up photograph

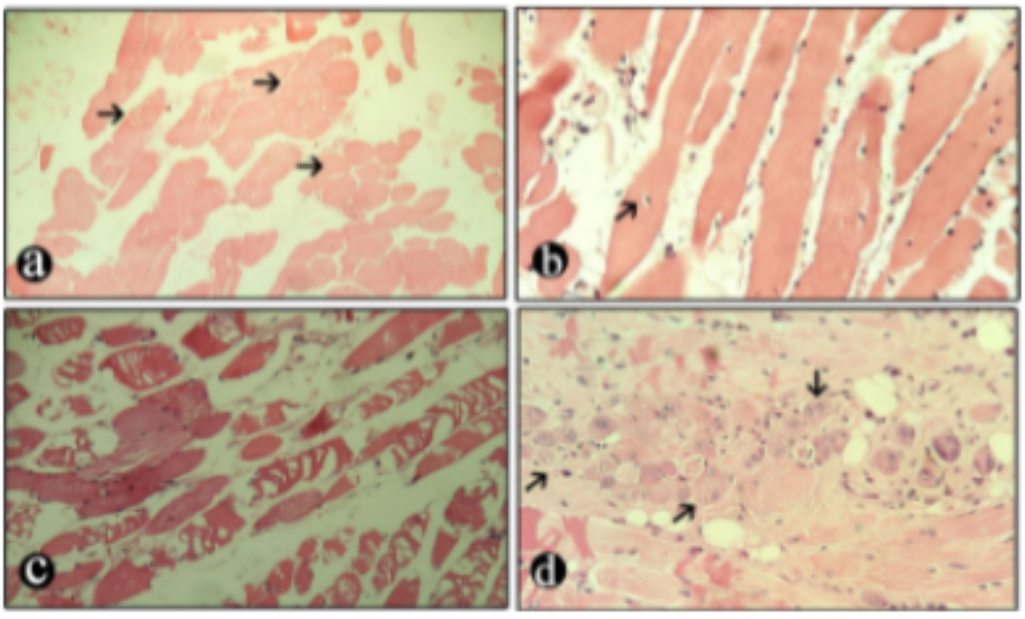

Fig. 4 Photomicrograph temporalis muscle tissue (H&E, 40×) showing hyalinization, loss of striation, and loss of nucleus

13]. Studies suggest that the dysplastic changes noted in the affected patients to be of 25% and the malignant transformation ranges from 3 to 19% [14, 15]. As the disease progresses, thick collagen band formation, hyalinization, and compromise in vascularity occurs and this leads to tissue hypoxia and permeability of epithelium to carcinogens, which is one of the suggested hypotheses for the malignant transformation of OSMF into SCC [4, 15, 16]. In advanced stages of the disease, not only cessation of patient's habits but also surgical intervention is obligatory to stop the disease progression. Surgical treatment has also helped to prevent malignant transformation in our experience. Surgical intervention to increase mouth opening restores the normal function of the oral cavity and aids in the regular inspection of their oral cavity for any malignant changes [17]. The present study was aimed at evaluating the histopathological changes in temporalis muscle fibers in patients undergoing surgical treatment for OSMF and to authenticate the importance of temporalis myotomy and coronoidectomy in surgical treatment protocol. Present study will be the first of this kind to register in the literature.

Younger age group was predominantly affected by the disease showing a peak incidence in 30–39 years. The youngest patient was 28 years and the oldest 55 years with a mean age of 39.8. Increased number of cases were reported <40 years as mentioned in the recent literature [2].

Khanna JN et al. have established the crucial role of areca nut in the etiopathogenesis of OSMF [2]. Present study also showed areca nut chewing was the major habit in the affected patients (73%) and disease was reported in patients chewing only gutka (27%).

Juxta-epithelial hyalinization was noted on histopathological examination of biopsied specimens of buccal mucosa in OSMF patients, known to be the major cause of trismus. The degeneration of masticatory muscles increases the severity of the trismus. Apart from the muscle degeneration due to activity of alkaloids released from areca nut or tobacco, overactivity of masticatory muscles in habitual chewers of these substances results in excessive glycogen consumption and depletion, thus leading to further degeneration, fibrosis, and scarring of muscle fibers [2, 4].

Treatment options for OSMF extend from pharmacological to surgical methods. Surgical intervention (fibrotomy) is the only efficient treatment option in patients suffering from advanced stages of OSMF as suggested by Khanna and Andrade, considering the severity of the trismus and the histopathological findings of secondary muscle degeneration and fibrosis [2].

Coronoidectomy with temporalis myotomy is carried out by few surgeons in all cases while others prefer it only if a minimum of 35 mm of mouth opening is not obtained after bilateral buccal mucosal fibrotomy [18, 19]. Also, the authors who highlighted the importance of these procedures have not provided with evidence for the actual involvement of the temporalis muscle in the disease progression [2, 18]. Degenerative changes in temporalis muscles cause abnormal contraction of these strong muscles [20]. Coronoidectomy along with release of these degenerated muscles from its attachments on mandible will remove muscle interference in carrying out post-operative mouth opening exercises, and thus prevent relapse.

All third molars were extracted during the surgery to facilitate the inset of nasolabial or buccal fat pad and to avoid post-operative trismus due to any infection from impacted third molars. All patients in our study group had third molars and they were extracted intra-operatively.

Histopathological evaluation of the temporalis muscle fibers excised from our study patients revealed various degrees of degenerative changes within the muscle bundles. Similar changes were noted in muscles obtained by punch biopsies of buccal mucosa by El-Labban NG and Canniff JP [20]. Hyalinization is the process whereby tissue degenerates, and in histologic appearance, the muscle cells and collagen fibers lose their detail and appear to fuse and take on a homogeneous, acellular, glassy eosinophilic appearance [4, 21]. Our study showed hyalinization of muscle bundles in 12 cases (80%). The myofibrils in skeletal muscle are composed of actin and myosin filaments, repeated in units called sarcomeres, the basic functional units which form the muscle fiber. The striated appearance of skeletal muscle fiber is due to the sarcomere and it forms the basic machinery for the muscle contraction. Loss of striation (Fig. 4) indicates loss of actin and myosin protein leading to loss of muscle contraction property and this will, in turn, lead to restricted mouth opening [8, 21]. Loss of striation in temporalis muscle bundles was seen in 11 cases (73%).

Fragmentation of muscle bundles, which was observed in 9 cases (60%), is secondary to lack of nutrition due to chronic injury to the tissue like in OSMF. Separation of the muscle bundles proposes that the change is brought by mechanical influences and dissociation of muscle cells that occur before death of these muscle fibers [5]. Skeletal muscle cells are elongated or tubular with nuclei located on the periphery of the cell. Skeletal muscle fibers are multinucleate because they are sincitio [3]. Three cases (20%) in study showed multiple nuclei and 5 showed nucleus internalization (33.33%) which is also suggestive of degeneration of temporalis muscle. Eccentric exercise causes initial sarcolemma injury which leads to subsequent inflammation. Swollen muscle fibers are secondary to inflammation and build-up fluid. In the present study, only one case (6.66%) revealed swollen muscle fibers [8]. In 3 cases (20%), inflammatory cell infiltrate was noted.

Leaving the fibrotomy defects for secondary epithelialization can lead to wound contracture and cause rebound fibrosis. This can be avoided by resurfacing the defect

using an appropriate vascularized tissue of adequate size. The reconstruction options ranges from extraoral, intraoral flaps, microvascular free flaps to alloplasts. Extraoral local flaps used for reconstruction are nasolabial flap and its modifications, temporoparietal muscle or fascia, and platysma flaps [22]. Buccal fat pad is the available intraoral reconstructive option. Combined nasolabial and buccal fat pad has been used as sandwich flap by Ambereen A et al. to overcome the drawbacks of using buccal fat pad or nasolabial flap alone for reconstruction [23]. Also, radial forearm free flap and collagen membranes also can be used for filling the resected defects of bilateral buccal mucosa [22]. Our experience has shown excellent results with the use of buccal fat pad for small to medium sized, more posterior defects, and nasolabial flaps for all other defects [5, 24]. The defect size was large in 80% of the cases treated in our study group; hence, we had to choose nasolabial flap for reconstruction in those patients. Buccal fat pad will be deficient for covering such a large defect especially anterior extend will be left uncovered, which can further lead to refibrosis [23].

To conclude, the present study revealed the significant pathological changes in temporalis muscle fibers of patients with advanced stages of OSMF. Surgical treatment of these patients should aim at preventing the malignant transformation of the disease, providing a disease-free life, and restoring the function of the oral cavity. Thus, we recommend our standard treatment protocol for the management of advanced stages of OSMF which includes the following:

- Fibreoptic nasotracheal intubation
- Bilateral fibrotomy
- Extraction of impacted third molars (if present)
- Bilateral coronoidectomy and temporalis myotomy
- Reconstruction of fibrotomy defect using nasolabial flap or buccal fat pad
- Followed by aggressive post-operative mouth opening exercises

Acknowledgements The authors would like to thank Dr. Niranjan Kumar and Dr. Balaram Naik for the support, encouragement, and facilities provided.

Declarations

Ethical approval Ethical approval is obtained from the Institutional Review Board, 2017/P/OS/47.

Informed consent Informed consent is obtained from all study group patients.

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