



Review Article

Paediatric oral submucous fibrosis – The neglected pre-malignancy of childhood



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ABSTRACT

Oral submucous fibrosis in children is one of the devastating consequences of areca nut addiction in children. Areca nut is the fourth commonest psychoactive agents used worldwide. However, regrettably, unlike other addictions like tobacco and alcohol, its use in children has become socially acceptable at least in a few countries. Consequently, children as young as two years have been noticed to be addicted to areca nuts. It has been a menace in most Asian countries and with increasing population migration, this has picked up even in the west. In order to understand the social factors leading to areca nut addiction in children and prognostic outcomes of paediatric oral submucous fibrosis, this review has been undertaken.

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1. Introduction

Areca nut chewing is the fourth frequently used psychoactive substance, next only to nicotine, ethanol and caffeine [1]. It is also

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the most significant causative factor for oral sub mucous fibrosis (OSMF), a pre-malignant condition of oral cavity [1]. With more than 600 million people using arecanut worldwide, the prevalence of OSMF is increasing at an alarming rate.

Paediatric OSMF is a very rare entity as most cases of OSMF are seen in middle aged populations [2,3]. Lack of information regarding the sociocultural practices of arecanut chewing makes diagnosis and treatment of this condition even more challenging. Recently, cases of paediatric OSMF have been documented in western countries too. This review was conducted to analyze paediatric OSMF and social circumstances leading to it.

2. Materials and methods

An exhaustive literature review was done using electronic databases (PubMed and Medline) and all relevant publications in English language reporting OSMF in children between 1952 and 2016 were included in our study. The following search terms were used: (1) “paediatric” OR “Child” AND “oral submucous fibrosis”; (2) “betel nut” OR “arecanut” AND “child” OR “paediatric”. Cases aged less than or equal to 15 years with detailed oral examination findings suggestive of OSMF were included. Further, an additional manual search was performed by cross-referencing the retrieved cases. The following data were collected – age, gender, social history, arecanut-related history, clinical examination findings, staging, treatment and outcomes. Studies with doubtful diagnosis, incomplete, insufficient and inconsistent information were excluded from our study. Classification proposed by More et al. was utilized for staging [4]. The classification consists of two parts:

A. Clinical staging

S1: Stomatitis and/or blanching of 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 oral cavity.

S4A: S1/S2/S3 with any potentially malignant lesions (Leukoplakia/Erythroplakia etc.)

S4B: Any one of the above stage along with oral carcinoma.

B. 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.

All patients were staged using both clinical and functional staging. For instance: S3M3, S2M4 etc.

2.1. Statistical analysis

Data was entered and analyzed on SPSS 17. Frequencies and percentages were computed for categorical variances.

3. Results

Data were collected from 13 studies and a cumulative of 18 cases of paediatric OSMF was found.

3.1. Demographics

Gender wise distribution showed female preponderance with a male to female ratio of 3:5. Most of the patients were females (55%). The mean age of presentation was 11.11 years (4–15 years). The youngest patient was a four year old girl. The oldest patients were a

15-year old boy and a similarly aged girl. Gender-wise distribution showed female preponderance with a male to female ratio of 3:5. Though most cases were seen in Asian countries especially India, three (16.66%) cases were observed in immigrant population in western countries in United Kingdom and Canada. Four (22.22%) children were reported to be employed as factory workers, domestic help and cow herd. They had an average age of 11.5 years.

3.2. Clinical characteristics

Reduced mouth opening or trismus and burning sensation while eating spicy foods were the commonest symptoms. Other commonly reported complaints were discomfort in mouth, recurrent oral ulcers, reduced tongue movements, dysphagia, loss of taste and protrusion of tongue. The mean duration of symptoms was 16.47 months (5d - 4years).

3.3. Arecanut-related history

All patients consumed arecanuts in one form or the other. Supari was the most common form chewed. The mean age of initiation was 7.40 years (2–13 years). Some consumed it 15 times a day while some chewed nearly five to six packets a day. The mean duration of consumption was 42.64 months (6–84 months). The site of placement of the arecanut product was mentioned in two cases. Buccal vestibule was used in one and the other case chewed it preferably from one side of mouth. In both, clinical findings were more pronounced on the side and site used for chewing. Thirteen patients knew at least one family member who chewed arecanuts. Rest of the details of arecanut consumption has been tabulated in Table 1.

3.4. Spices-related history

Only two cases reported excessive spice consumption in daily foods. They were advised to reduce spice consumption. The rest of the cases neither reported any spice intake history nor advised any dietary restrictions.

3.5. Examination findings

On general physical examination, one child was noted to be undernourished and another was thin. On oral cavity examination, the mean inter-incisional distance on maximal mouth opening was 16.45 mm (11–30 mm)*. [*Two cases had a reported mouth opening of 15–20 mm. For the purpose of calculation of mean, 18 mm was taken for both these cases]. Most cases showed pale oral mucosa, fibrotic bands and loss of mucosal elasticity. Other commonly observed oral findings were contracted uvula, whitish lesions and minor aphthous ulcers, erythematous lesions, melanotic pigmentation, leathery mucosa, gingivitis, Depapillated tongue, poor oral hygiene, dryness of mouth, caries teeth, fluorosis and pericoronitis. One patient was also noted to have a slightly deviated mandible to the side preferably used for arecanut chewing.

3.6. Staging

Most patients belonged to advanced stages (S3/4 and M3/4 based of classification described in methodology) as shown in Table 2.

3.7. Investigations

Most cases (88.88%) underwent a combination of various

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