Pedunculated Origin of Intraoral Fibrolipoma – Report of A Rare Case and Systematic Quantitative Literature Review

Case Report

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ABSTRACT

Lipoma is a benign mesenchymal tumour that rarely develops intraorally. Intraoral lipoma usually presents as a slow-growing, painless mass, occasionally interfering with speech and mastication. Here we describe a patient with pedunculated intraoral lipoma causing speech and mastication difficulties. A 48-year-old gentleman presented with a slow-growing, painless intraoral mass causing discomfort while chewing and talking. The mass originated from the buccal mucosa through a thin peduncle. We excised the tumour entirely from its root. Histopathologically it was diagnosed as fibrolipoma. We also conducted a systematic quantitative review of the PubMed database following PRISMA guidelines using specific sets of keywords and defined inclusion and exclusion criteria. Total 44 relevant articles were identified comprising of 175 patients. Their demographic and clinical profiles are described. Most common site of origin is buccal mucosa followed by tongue and lips. The presenting symptoms vary according to the intraoral location of the lesion. Pedunculated origin of intraoral lipoma is an extremely rare presentation. It is difficult to diagnose clinically, but a high degree of clinical suspicion helps in dealing with such cases.

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INTRODUCTION

Lipoma is a benign mesenchymal neoplasm composed primarily of mature adipocytes. About 15 to 20% of lipoma arise in the head and neck region[1]. Incidence of intraoral lipoma is even rare (1 to 4% only)[1]. While diagnosing such a case, other tumours of mesenchymal origin such as neurofibroma, schwannoma, lymphoma, haemangioma, and lymphangioma should also be kept in mind^[2]. Lipoma usually presents as a painless, soft to firm palpable swelling that rarely causes significant symptoms other than the cosmetic defect. But intraoral lipoma may occasionally impair the functions of mastication, speech, and even swallowing depending on its intraoral location^[3, 4]. Histologically, lipoma can be classified into classic lipoma, fibrolipoma, chondrolipoma, sialolipoma, spindle cell lipoma and osteolipoma^[1]. Surgical excision of the mass is the treatment of choice. The final confirmation of the diagnosis is made by histopathological examination.

Case Report:

A 48-year-old gentleman presented with a swelling inside the oral cavity on the right side for the last ten years. It was interfering with his mastication and speech.

He had a history of chewing tobacco products for the last 20 years. There was no history of any alcoholism or substance abuse. Externally no swelling was found over the cheek. Intraoral examination revealed a smooth, pink coloured, pedunculated, mobile mass arising from the right side of the buccal mucosa adjacent to the lower gingivolabial sulcus (Figure 1). The mass was smooth surfaced, non-tender, and soft to firm in consistency. It was free inside the oral cavity, and the only attachment was to the buccal mucosa through the peduncle (Figure 2). The adjacent areas of the buccal mucosa, tongue, and gingivolabial sulcus were unremarkable. The rest of the oral, oropharyngeal, and laryngeal structures did not reveal any abnormality. No cervical lymph node was palpable clinically. We performed the surgical resection of the mass from the root of the peduncle under local anaesthesia. The surgical incision was sutured with 3-0 Vicryl (Ethicon Inc, polyglactin 910, cutting edge). It was a 2.2×3.2 cm smooth mucosa-covered mass (Figure 3). The histopathological examination revealed mature adipose tissue interspersed with broad bands of dense connective tissue, thus confirming the diagnosis of fibrolipoma (Figure 4). No recurrence was seen after six months of follow-up.



Fig. 1: Smooth surface, pink coloured mass inside the oral cavity adjacent to the tongue



 $\textbf{Fig. 2:} \ \, \textbf{Intraoral mass arising from the buccal mucosa through a thin peduncle (Black arrow)}$



Fig. 3: Intraoral mass after surgical excision (3.2 * 2.2 cm). Black arrow shows the cut-end of the peduncle.

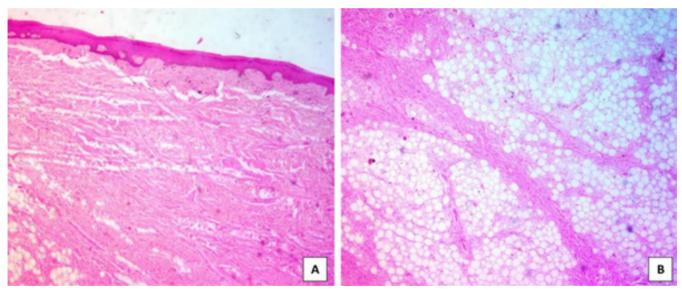


Fig. 4 A: Histological sections revealed stratified squamous epithelium with underlying fibrous connective tissue showing bundles of collagen fibers arranged in a haphazard manner with few lobules of adipocytes (hematoxylin and eosin stain, ×4). B. Deeper areas of the section show mature adipose tissue interspersed by broad bands of dense connective tissue (hematoxylin and eosin stain, ×10).

DISCUSSION

Lipoma is a benign mesenchymal neoplasm composed primarily of mature adipocytes. Incidence of intraoral lipoma is rare (1 to 4% only)^[1].

We conducted an extensive literature search in the PubMed database pursuant to the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) statement. The database was accessed

on 14th July, 2022 using Google chrome browser (Google, USA) using three separate sets of keywords: (fibrolipoma) AND (oral cavity), (fibrolipoma) AND (intraoral) and (fibrolipoma) AND (oropharynx). The collective list of the articles was scanned to remove the duplicates. The exclusion criteria comprised of non-English literature articles, animal studies and articles describing expert opinion rather than patient centric data (Figure 5). We only included those articles describing fibrolipoma in the intraoral locations (oral cavity and oropharynx). Other histologic variants

of intraoral lipomatous tumours were excluded to confine the search results only to 'fibrolipoma'. Lastly, we excluded the articles with incomplete or no information about demographic profile of the patients and/ or location/nature of the lesions. We also scanned the reference section of the selected articles to expand the scope of the search. All the authors independently extracted data from the selected articles and the collective data was compiled.

Thirty case reports and fourteen case series, published between 1954 to 2022 were included in our review^[5-48]. Total number of patients was 175. Mean age of the patients was 49.52 ± 20.26 years with a slight female preponderance. Regarding the duration, nature and dimension of the lesions, data were available for 35, 15 and 39 articles respectively. Median duration of the intraoral fibrolipoma was 24 months (IQR = 47.5). Information was available for 17 pedunculated and 18 sessile lesions. The mean dimension of all the lesions was 2.35 ± 2.39 cm, the largest lesion having a maximum dimension of 16.7 cm (Table 1).

Regarding the intraoral location maximum number of lesions arose from the buccal mucosa (41%), followed by tongue (12%), lips (11%), buccal sulcus (8%), floor of mouth (6%) and palate (6%) (Table 2). The symptoms varied with the location of the lesions. Most of the lesions arising from intraoral subsites remained unnoticed for a long duration, only to be discovered while chewing or swallowing. But the lesions with nasopharyngeal origin presented with progressive nasal obstruction, conductive hearing loss, snoring and breathing difficulties. The largest lesion of nasopharyngeal origin had the maximum

dimension of 6.5 cm that extended from nasopharynx to hypopharynx causing breathing and swallowing difficulties. All the patients were treated by surgical excision of the intraoral lesion by different approaches. Simple intraoral approach was sufficient for the lesions confined to the oral cavity. But for the nasopharyngeal lesions often a combined transnasal endoscopic and intraoral approach was necessary.

Intraoral lipoma may occasionally impair the functions of mastication, speech, and even swallowing depending on its location^[3,4]. In our patient, the lipoma originated from the buccal mucosa close to the lower gingivolabial sulcus. But surprisingly, it was attached to the buccal mucosa only through a thin peduncle. The rest of the mass was mobile inside the oral cavity with the axis of movement around the peduncle. It was very close to the tongue; hence the movements often hampered the tongue movements. It also interfered with the speech and mastication of the patient and was the reason for his discomfort. After surgical excision, it was diagnosed as a case of fibrolipoma in the histopathological sections.

This was a very unusual presentation of intraoral lipoma where the entire attachment was dependent on a thin peduncle, and the rest of the mass was mobile inside the oral cavity. One such case of pedunculated intraoral lipoma was described by Shah KM *et al.*^[49]. This type of intraoral exophytic mass is difficult to diagnose clinically due to its atypical presentation. Therefore, excisional biopsy with histopathological confirmation is the treatment of choice. A high degree of clinical suspicion is warranted while dealing with such cases.

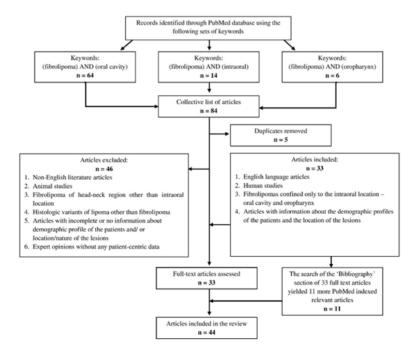


Fig. 5: PRISMA flowchart

Table 1: Demographics and clinical characteristics of patients with intraoral fibrolipoma

Age (years)	$49.52 \pm 20.26 \text{ (Mean} \pm \text{SD)}$
Gender	Male: Female – 9: 10
Duration (months) (Data available for 35 articles)	Median – 24 Interquartile range – 47.5
Nature (Data available for 15 articles)	
Pedunculated	17
Sessile	18
Dimension (cm) (Data available for 39 articles)	2.35 ± 2.39 (Mean \pm SD)

Table 2: Location of the intraoral fibrolipoma

Location	Number of patients
Buccal mucosa	72
Gingiva	8
Buccal sulcus	14
Floor of mouth	11
Lips	19
Tongue	21
Gingivo-labial sulcus	1
Retromolar area	6
Tonsillar fossa	2
Palate	11
Vestibule	5
Nasopharynx	3
Posterior oropharyngeal wall	1
Palpable inside mouth	1

CONFLICT OF INTEREST

There are no conflicts of interest.

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