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A Bibliometric Analysis on Development of Sub-Lingual Epidermoid Cyst in Children

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Keywords: Advanced Diagnostic method; Intra oral cyst; Sub-lingual Epidermoid Cyst; Surgical excision; Plunging ranula; Pediatric Patient.

ABSTRACT

Various intra oral cystic lesions are encountered during routine dental check-up or reported by patients and is commonly observable finding during clinical practice. Sublingual epidermoid cyst is a type of benign subcutaneous cystic lesion, derived from the ectoderm, and is rare and develops beneath the tongue or floor of the mouth. Most of the time, these cysts are found asymptomatic, however are reported with discomfort or complications in a patient if they grow in size. Therefore, a thorough knowledge about the nature of sublingual epidermoid cysts is essential arriving at proper diagnosis and also to provide appropriate treatment. In this article, an extensive literature review of the cases reported so far using PUBMED electronic database search was performed to explore its characteristics, etiological features, symptoms, diagnosis, complications, treatment options and prognosis along with its bibliographic details.

Introduction

Epidermoid cyst is a rare benign cystic lesion which is rarely seen in the floor of the mouth. Being a cystic malformation deriving from the ectoderm, they represent less than 0.01% of all the oral cavity cysts [1-3]. While relatively uncommon, these cysts can lead to various symptoms and may require meticulous diagnosis and treatment planning. In composition, they are filled with keratin, sebum, and other skin debris. As a result, they are considered a type of inclusion cyst [4].

Occurrence of epidermoid cyst are more frequent in the other part of the body like skin, ovaries, testicles (80%), head and neck region (1.6-6.9%) compared to intraoral appearance (0.01%) [2,3]. As their occurrence in the oral cavity is uncommon, there are less publications found in the dental journals especially in pediatric journals. Moreover, occurrence of these cysts in pediatric patients are extremely rare. Literature search revealed hardly countable number of publications and no publications in pediatric journals (Table 1) [3,5-11]. Moreover, it is evident that there is no bibliographic analysis carried out pertaining to this cyst apart from few simple narrative reviews. Therefore, the aim of the present article is to analyses the bibliographic details of the sublingual epidermoid cyst pertaining to its characteristics, etiological features, symptoms, diagnosis, complications, treatment options and prognosis in children. Such detailed bibliometric analysis contributes to the thorough understanding of occurrence of this uncommon lesion in pediatric patients providing insights into their management and the importance of meticulous diagnosis.

Methodology

The detailed electronic database search was carried out using PubMed from 1992 till 2024 for a period of one week using key words (MeSh terms) like 'sub-lingual cysts', 'intraoral cysts in children,' 'epidermoid cysts,' 'soft tissue lesions in oral cavity' and 'cysts in floor of the mouth' in different combinations. Only relevant articles pertaining to sublingual epidermoid cysts and which are in English language were considered for evaluation. Those reported in the head and neck region or other parts of the body, other parts of the oral cavity were excluded from the review. Letter to editor and abstracts were also not considered for the evaluation.

Results

Following the literature search, a total of 43 publications were selected (Figure 1). All publications were tabulated pointing the demographic factors like author, year of publication, age, gender of the patient, journal in which article is published, place of the case published, symptoms, location and treatment approach carried out in chronological order starting from new to old (Table 1) [1-45].

Table 1: *List of published sublingual epidermoid cysts in the English literature retrieved through PUBMED search*

Case No.	Author	Year	Patient age (in years)/Gender	Journal name	Place of the study
1.	Basavanthappa et al [45]	2024	6/Female	Indian Journal of Otolaryngology and Head and Neck Surgery	India
2.	Basla et al [2]	2023	17/Male	Acta Otorhinolaryngology Italy	Italy
3.	Ullah et al [12]	2023	30/Male	Cureus	Pakistan
4.	Dokania V [13]	2023	28/Female	Cureus	India

5.	Datta et al [14]	2020	24/Male	Annals of Maxillofacial Surgery	India
6.	Klibngern et al [15]	2020	26/Female	International Journal of Surgery in Case Reports	Thailand
7.	Thibouw et al [16]	2020	73/Male	National England Journal of Medicine	France
8.	Santos et al [3] (48-year retrospective study)	2020	13 cases (0.08%) 8 cases -Female (61.5%) Female:Male ratio 1.6:1 11 months to 82 years. Mean age 38.2 years	Medicine Oral Pathology Oral Circum Bucal	Brazil
9.	Silveira et al [17]	2019	26/Male	Journal of Oral and Maxillofacial Surgery	Brazil
10.	Brunet-Garcia et al [18]	2018	43/Male	Journal of Clinical and Experimental Dentistry	Spain
11.	Findik et al [5]	2017	10/Male	Journal of Pakistan Medical Association	Turkey
12.	Sahoo et al [19]	2017	1. 55/Male (submental) 2. sublingual	Annals of Maxillofacial Surgery	India
13.	Nishar et al [20]	2016	60/Male		India
14.	Utumi et al [21]	2016	15/Female	Autopsy in Case Reports	Brazil
15.	Gulati et al [22]	2015	16/Male	Iranian Journal of Otolaryngology	India
16.	Soares et al [23]	2015	45/Male	Case Reports in Dentistry	Brazil
17.	Daban et al [6]	2015	3/Female	Case Reports in Dentistry	Spain
18.	Baliga et al [24]	2014	26/Female	National Journal of Maxillofacial Surgery	India
19.	Mirza et al [25]	2014	43/Male	Qatar Medical Journal	Qatar
20.	Tandon et al [26]	2014	23/Female	Journal of Maxillofacial and Oral Surgery	India
21.	Zielinski et al [7]	2014	1. 6/Male 2. 15/Female	Open Medicine (Wars)	Poland
22.	Gordon et al [27]	2013	79/Female	Journal of Maxillofacial and Oral Surgery	USA
23.	Mammen et al [28]	2013	57/Male	Journal of Clinical and Diagnostic Research	India
24.	Kudoh et al [29]	2013	69/Male	Case Reports in Medicine	Japan
25.	Verma et al [30]	2012	16/Female	National Journal of Maxillofacial Surgery	India

26.	Assaf et al [31]	2012	39/Male	In Vivo	Germany
27.	Banerjee et al [32]	2011	61/Female	Journal of Maxillofacial and Oral Surgery	India
28.	Lyngdoh et al [1]	2010	24/Male	Indian Journal of Surgery	India
29.	Anantanarayanan et al [8]	2010	12/Female	Head Neck Pathology	India
30.	Patil et al [33]	2009	28/Male	Cases Journal	India
31.	Tsirevelou et al [9]	2009	1. 14/Female 2. 35/Female	Cases Journal	Greece
32.	Fung et al [34]	2008	50/Male	Hong Kong Medical Journal	Hong Kong
33.	Kandogan et al [10]	2007	11/Male	Journal of Medical case Reports	Turkey
34.	Jham et al [35]	2007	25/Male	Journal of the Canadian Dental Association	USA
35.	Yilmaz et al [4]	2006	1. 35/Male 2. 4/Female	Journal of Laryngology Otology	Turkey
36.	Seah et al [36]	2004	26/Male	Annals Academy of Medicine Singapore	Singapore
37.	Bitar et al [37]	2003	17/Female	European Archives of Otorhinolaryngology	Lebanon
38.	De Ponte et al [38]	2002	18/Male	Journal of Craniofacial Surgery	Italy
39.	Behl et al [39]	2001	22/Male	Medical Journal of Armed Forces India	India
40.	Walstad et al [40]	1998	23/Female	Journal of Maxillofacial and Oral Surgery	USA
41.	Turetschek et al [41]	1995	25/Female	British Journal of Radiology	Australia
42.	Calderson et al [11]	1993	2/Female	Journal of Oral Maxillofacial Surgery	Israel

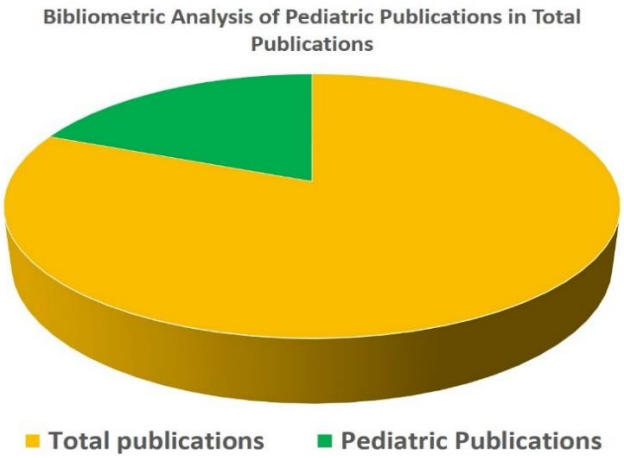

Figure 1: *Bibliometric analysis of Pediatric and total publications*

A total of 60 cases have been reported from total 43 publications, in that majority publications were contributed from India (n=13) followed by Brazil (n=4), USA (n=3) and Turkey (n=3) (Table 1) (Figure 2). Maximum publications were reported in medical journals with only eight reports published in journals related to dentistry such as Journal of the Canadian Dental Association, Case Reports in Dentistry, Journal of Oral and Maxillofacial Surgery, National Journal of Maxillofacial Surgery, Annals of Maxillofacial Surgery, Journal of Clinical and Experimental Dentistry, Medicine Oral Pathology Oral Circum Bucal and Journal of Maxillofacial and Oral Surgery. Not a single publication was published in pediatric dental journals (Table 2).

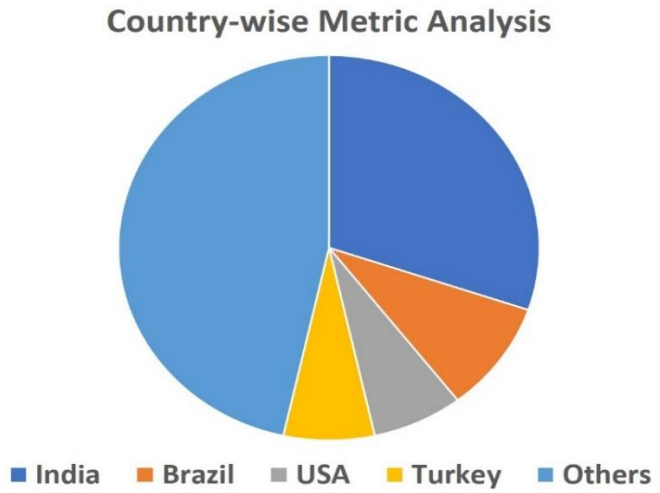

Figure 2: *Country-wise metric analysis about their contribution*

Table 2: *Bibliometric analysis of Journal's contribution towards occurrence of Sublingual Epidermal cysts.*

Sl. No.	Name of the Journal	Total Publication Contribution
1.	Journal of Maxillofacial and Oral Surgery	5
2.	Journal of Oral and Maxillofacial Surgery	4
3.	National Journal of Maxillofacial Surgery	2
4.	Case Reports in Dentistry	2
5.	Cases Journal	2
6.	Cureus	2
7.	Annals of Maxillofacial Surgery	2
8.	Indian Journal of Otolaryngology and Head and Neck Surgery	1
9.	International Journal of Surgery in Case Reports	1
10.	Acta Otorhinolaryngology Italy	1
11.	National England Journal of Medicine	1
12.	Medicine Oral Pathology Oral Circum Bucal	1
13.	Journal of Clinical and Experimental Dentistry	1
14.	Journal of Pakistan Medical Association	1
15.	Qatar Medical Journal	1

16.	Open Medicine (Wars)	1
17.	Journal of Clinical and Diagnostic Research	1
18.	Case Reports in Medicine	1
19.	Indian Journal of Surgery	1
20.	Head Neck Pathology	1
21.	Hong Kong Medical Journal	1
22.	Journal of Medical case Reports	1
23.	Journal of the Canadian Dental Association	1
24.	Journal of Laryngology Otology	1
25.	Annals Academy of Medicine Singapore	1
26.	European Archives of Otorhinolaryngology	1
27.	Journal of Craniofacial Surgery	1
28.	In Vivo	1
29.	Medical Journal of Armed Forces India	1
30.	British Journal of Radiology	1
31.	Iranian Journal of Otolaryngology	1

Discussion

In the present bibliometric analysis about the domain occurrence of sublingual epidermal cysts, literature search was carried out using PUBMED electronic database from inception 1992 till December 2024 and the selected articles were tabulated (Table 1). Following the search, a total of 43 publications reporting total 60 cases have been reported occurring in the sublingual region of the oral cavity.

Evaluation of reported sublingual epidermoid cysts in pediatric patients revealed hardly nine cases in the English literature showing uncommon occurrence of these cysts during childhood or puberty (Table 1). Among these nine cases, six cases were diagnosed in female patients and remaining three in male patients. This finding was contrast to the previous reports showing male predilection found in adult cases [2,3,25]. However, some reports show there is no gender predilection for this cyst [12-15]. The age of the patients in children ranged from 11 months to 14 years old in this review. In case of adult patients, a wide age group is observed ranged from 15 to 87 years with mean age of 34 years in males and 29 in females [2,3]. Only in two cases symptoms like difficult in chewing and speaking was present and in remaining cases there were no symptoms present with the lesion.

Santos et al recently (2020) [3] evaluated the clinical and morphological features of both dermoid and epidermoid cysts of the oral cavity in their 48-year retrospective study in Brazil at an oral diagnosis reference center. Among 15.387 documented cases, only 4 (30.7%) of sublingual epidermal cysts were found and in that they found only one case of epidermoid cyst in an eleven-month-old female infant. Unfortunately, they have not mentioned the treatment approach done in this case.

In children, these cysts usually mimic or be mimicked by dermoid cyst or ranula in that region. It is very difficult to differentiate between plunging ranulas from plunging sublingual epidermoid cyst and pose a great diagnostic challenge as both of them exhibit very similar clinical features [12-14]**.** A ranula is a retention cyst of the sublingual salivary gland, characterized by unilateral transparent lesion occur in the floor of the mouth below the tongue. Even this is more common in children. Therefore, in differential diagnosis of large sublingual cysts, dermoid cyst and simple ranula should be considered. Using only radio imaging studies, it is not possible to distinguish between a dermoid, epidermoid cyst or a simple ranula. As three of them require different treatment strategies, it is important to differentiate one from the other. FNAC investigation guides in

differentiating ranula from epidermoid or dermoid cyst. In addition to this, MRI provides a valuable information of the cyst location thereby helping in selection of appropriate surgical approach [19,27]. In the present case, there was a diagnostic dilemma regarding plunging ranula or sublingual epidermoid cyst as both are common in the midline of the oral cavity in the anterior portion of the sublingual space. When both these swellings in the floor of the mouth dissect through the mylohyoid muscle and produce swelling within the neck, they are referred to as plunging sublingual epidermoid cyst or plunging ranulas. But following histopathological examination, a final diagnosis of plunging sublingual epidermoid cyst was made. Therefore, for proper diagnosis and to rule out from different lesions, a histopathological examination is always mandatory [30,31].

The clinical appearance of these lesions in the oral cavity depends on their size, as well as their anatomical locations. They vary in size from a few millimeters to a few centimeters and literature shows few cases of giant cysts [20,30,32]. In one case it measured 7 cm and, in another case, it measured 13 cm. Lyngdoh et al [1] published a giant sublingual epidermoid cyst which measured 13x13 cms, with visible Wharton's duct openings on both the sides. Radiographic examination revealed even a grossly enlarged mandible in this case. Therefore, early detection and diagnosis of all cystic lesions in children is highly essential [46]. Regarding review of treatment approach, in four cases intraoral approach and in remaining cases extra oral approach was used. In the present case, transoral approach was used. The intraoral approach is more appropriate for sublingual epidermoid cysts and small/medium sized lesions and has the advantage of no external visible scar [43]. However, it is associated with limited exposure and with higher morbidity due to damage to Wharton's duct and other vital structures in the sublingual space. Transcervical approach is required for infra-mylohyoid cysts and large lesions which gives an optimal result.

Conclusion

Sublingual epidermoid cysts are rare in children but can present challenges when they become symptomatic. Understanding their characteristics, causes, symptoms, and available treatment options is crucial for healthcare professionals and individuals including pediatric dentist affected by these cysts. Early diagnosis and appropriate management contribute to a positive prognosis and improved quality of life for those with sublingual epidermoid cysts.

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