

REVIEW

Imaging features of epidermoid cyst located in the floor of the mouth: case report and narrative review of literature

Caratteristiche imaging di una cisti epidermoide nel pavimento orale: case report e review narrativa della letteratura

Nicoletta Basla¹, Domenico Sfondrini², Marina Francesca Achilli¹, Michele Catalano¹, Francesco Sanvito¹, Maria Gabriella Sala³, Stefano Marelli², Marco Benazzo^{4,5}, Lorenzo Preda^{1,3}

¹ Radiology Unit, Department of Clinical, Surgical, Diagnostic and Pediatric Sciences, University of Pavia, Pavia, Italy; ² Maxillo-facial Surgery Unit, Fondazione IRCCS Policlinico San Matteo, Pavia, Italy; ³ Radiology Unit 1, Fondazione IRCCS Policlinico San Matteo, Pavia, Italy; ⁴ Otorhinolaryngology Unit, Department of Clinical, Surgical, Diagnostic and Pediatric Sciences, University of Pavia, Pavia, Italy; ⁵ Otorhinolaryngology Unit, Fondazione IRCCS Policlinico San Matteo, Pavia, Italy

SUMMARY

Epidermoid cysts are benign slow-growing developmental lesions resulting from ectodermal tissue that can localise anywhere in the body. Only 7% of all epidermoid cysts are located in the head and neck area and, together with dermoid cysts, account for less than 0.01% of all oral cavity cysts. Herein, we present the case of a 17-year-old male diagnosed with a sublingual epidermoid cyst and a review of the literature on this rare localisation, focusing on clinical and imaging features, management and outcomes. All the articles reporting epidermoid cysts located in oral cavity were searched using PubMed/Medline, discarding all those not located in head and neck district, not mentioning diagnostic imaging, not defined as epidermoid cysts by a histopathological diagnosis, and those for which only an abstract was available. In literature there were 35 articles, from 1993 to 2020, that meet the aforementioned requirements, for a total of 38 patients and 39 cysts. For each article, age, sex, onset time of symptoms, location, size and surgical access were analysed. Particular attention was paid to the radiological investigation used in order to understand the most common imaging aspects and also rare cases with unusual presentations.

KEY WORDS: epidermoid cyst, mouth, diagnosis, review of reported cases

RIASSUNTO

Le cisti epidermoidi sono lesioni benigne, a lenta crescita, derivanti dal tessuto ectodermico e possono localizzarsi in qualsiasi parte del corpo. Solo il 7% si localizza nel distretto testa-collo e, sommate alle cisti dermoidi rappresentano meno dello 0,01% di tutte le lesioni cistiche della cavità orale. Le cisti epidermoidi localizzate all'interno della cavità orale sono rare e generalmente rinvenute in pazienti giovani adulti, senza significativa prevalenza di genere. È presentato il caso di un maschio di 17 anni, con diagnosi di cisti epidermoide sublinguale, e una revisione della letteratura su questa rara localizzazione, puntando l'attenzione sulle caratteristiche cliniche, sull'imaging radiologico, il trattamento scelto e i dati istologici. Tutti gli articoli analizzati nella revisione della letteratura sono stati reperiti tramite database PubMed/Medline; sono stati scartati i casi di cisti non situate nel distretto testa-collo, gli articoli dove non è menzionata alcuna diagnostica strumentale, o dove la diagnosi istologica di cisti epidermoide non è stata confermata.

In letteratura sono riportati 35 articoli, dal 1993 al 2020, che soddisfano i suddetti requisiti, per un totale di 38 pazienti e 39 cisti. Per ogni articolo sono stati analizzati: età e sesso, tempo di insorgenza dei sintomi, posizione e dimensioni, e accesso chirurgico. Particolare attenzione è stata rivolta all'indagine radiologica scelta, al fine di comprendere gli aspetti di imaging più comuni e anche casi rari con presentazioni insolite.

PAROLE CHIAVE: cisti epidermoide, bocca, diagnosi, revisione dei casi in letteratura

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Correspondence

Nicoletta Basla

Radiology Unit, Department of Clinical, Surgical, Diagnostic and Pediatric Sciences, University of Pavia, viale Camillo Golgi 19, 27100 Pavia, Italy
Tel. +39 0382 503761; + 39 0382 501315
E-mail: nicoletta.basla01@universitadipavia.it

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Introduction

Dermoid cysts are malformations lined with squamous epithelium and usually the term “dermoid” is used indistinctly to define dermoid cysts, epidermoid cysts, and teratoid cysts. These can be classified according to the epithelial components and the degree of differentiation of the elements that compose them ^{1,2}.

Epidermoid cysts are coated with simple squamous epithelium and have a fibrous wall. True dermoid cysts (or compound cysts) consist in an epithelial-lined cavity with keratinization and with skin appendages in the cyst wall, like sebaceous and sweat glands, hair and hair follicles. Finally, teratoid cysts (or complex cysts) may be covered in different types of epithelium and contain derivatives of ectoderm, mesoderm and endoderm, such as bone, muscle, gastrointestinal and respiratory tissue ^{2,3}.

The differential diagnosis of dermoid cysts of the floor of the mouth includes mucus extravasation phenomenon, embryological anomalies, infection (such as acute bacterial infection/cellulitis of the oral floor, sialadenitis, viral lymphadenitis), infectious and non-infectious granulomatous disease, non-granulomatous inflammatory disease, benign or malignant tumour of the salivary glands, normal fat tissue in the submental/mandibular area, HIV-related lymphadenopathy ^{3,4}.

The aetiology of the dermoid and epidermoid cysts is unknown, although several theories have been proposed ⁵. Medial epidermoid cysts of the floor of the mouth can be the result of entrapped ectodermal tissue of the first or second branchial arches, which fuse during the 3rd or 4th week of gestation. A second theory is a surgical or accidental event conditioning a traumatic implantation of epithelial cells into deeper tissues. Finally, another hypothesis is that these cysts are a variant of thyroglossal duct cysts ⁵. Usually they present as solitary masses, but can sometimes be multiple ⁵. Epidermoid cysts located in the oral cavity are rare and usually found in young adult patients without significant gender prevalence. Only 7% of all the epidermoid cysts are located in the head and neck area which, together with dermoid cysts, account for less than 0.01% of all oral cavity cysts ⁶.

Epidermoid cysts are rarely located in the floor of the mouth, primarily in the sublingual, submaxillary, or submandibular spaces ⁷. When located in the lower part of the neck, they must be differentiated from thyroid nodules ⁸.

Usually they are asymptomatic, but in some cases they can interfere with mastication, swallowing, speaking and breathing. Moreover, an infection can occur but rarely causes a life-threatening situation ⁴.

For diagnosis, ENT surgical evaluation and imaging of

first (ultrasonography US) and second levels (computed tomography CT, or magnetic resonance MRI) are required, together with cytologic examination following fine-needle aspiration biopsy (FNAB) ⁷.

Surgical excision is the treatment of choice and the approach depends on the location and size of the lesion ⁹.

The access can be with an extraoral incision if the lesion is located under the geniohyoid muscle, or with an intraoral approach if it is located above the mylohyoid muscle ⁷.

Prognosis is good, with a very low incidence of recurrence. Malignant changes have been recorded in dermoid cysts, but not in the floor of the mouth, with a 5% rate of malignant transformation for the teratoid type ⁷.

We present a rare case of epidermoid cyst located in the sublingual space, and discuss the radiological findings by collecting and comparing other evidence from the literature.

Case report

A 17-year-old teenager presented to his general practitioner complaining of a submental swelling, appearing approximately one year earlier, with an increase in size in the last six months. After physical examination, in the suspicion of submental lipoma, an ultrasound (US) examination was requested, along with ENT surgical evaluation for possible excision.

The physical examination conducted by the ENT specialist in August 2021 revealed a 5 cm soft swelling, fixed with respect to the deep planes, regular contours, without tenderness.

US showed a 40 mm wide and inhomogeneous cystic lesion. The cyst content was hypoechoic, with some hyperechoic spots. No apparent vascularisation was identified with colour Doppler US. These findings were considered suspicious for lipoma or cyst with particulate content (Fig. 1A).

A second level imaging MRI scan (Figs. 1B-G) was performed. The images showed a well-circumscribed, non-enhancing, cyst. The cyst content was homogeneously hyperintense on T2 weighted images, and iso-hypointense on T1 weighted images. Diffusion Weighted Imaging (DWI) and the corresponding Apparent Diffusion Coefficient (ADC) map did not reveal a restriction of water molecule diffusion. No signal suppression was identified on fat-saturated images.

The lesion was located medially under the geniohyoid muscle, contacted the inferior side of the mylohyoid muscle, and, laterally, the belly of the digastric muscle.

In consideration of the imaging features, we posed a hypothesis of epidermoid cyst.

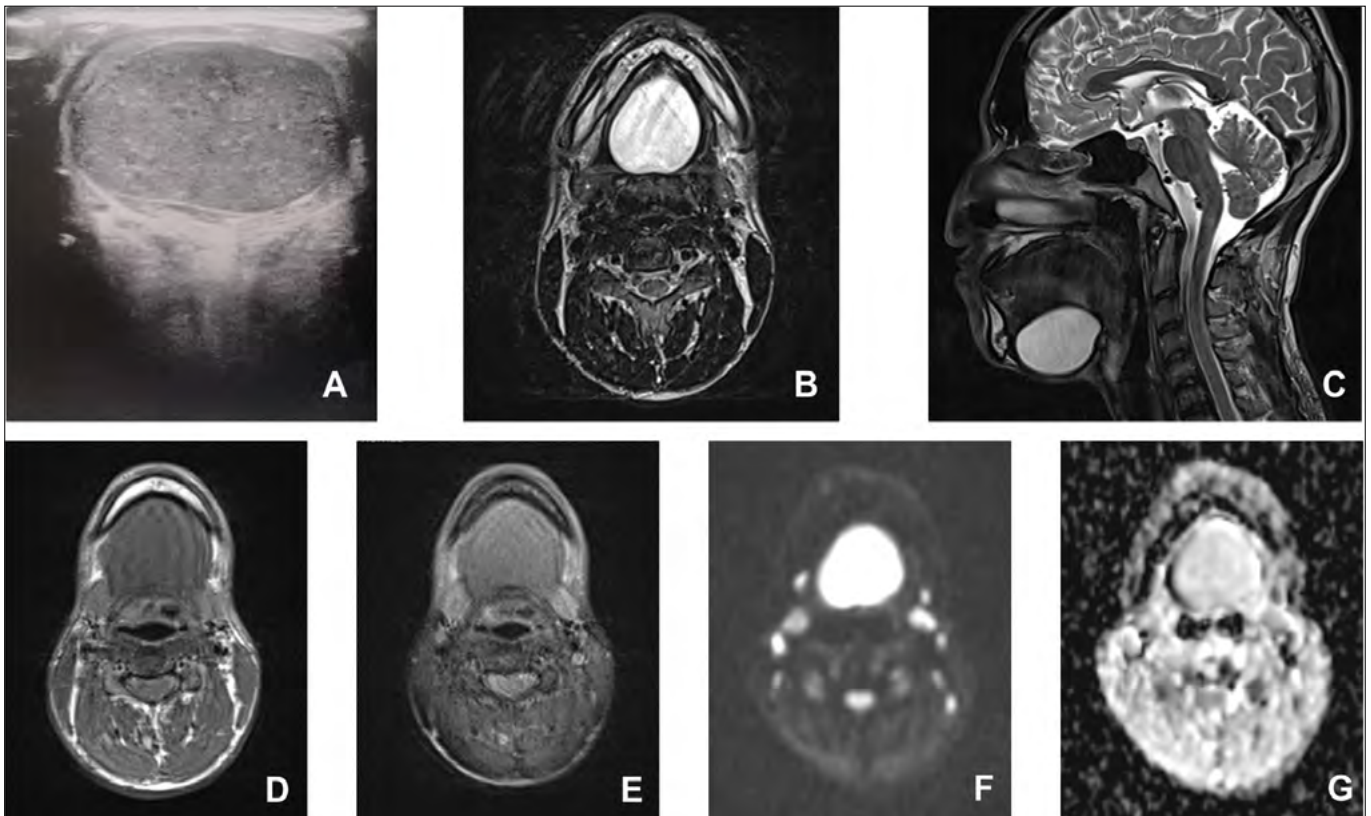


Figure 1. The images above show, respectively, in the first row sonographic appearance of the cyst, with posterior enhancement artifact and hyperechoic spots (A). In T2w images on axial (B) and sagittal (C) plans it appears homogeneously hyperintense. It appears slightly hypointense in T1w (D) and isointense in T1w Fat Sat images (E). There is no pathological restriction of signal on diffusion weighted images (F) and the respective apparent diffusion coefficient map (G). As seen on the sagittal plane, the lesion is located under the geniohyoid muscle, dislocating inferiorly the mylohyoid muscle.

Other possible differential diagnoses were excluded. An infectious hypothesis was discarded because the US did not demonstrate vascularisation of the lesion. A lipoma was ruled out since it did not have fatty components. The hypothesis of teratoid cyst was not convincing, considering its homogeneous features. Given the location along the median line of the oral floor, the hypotheses of ranula and thyroglossal duct cysts were also considered; however, in accordance with the ultrasound appearance they were subsequently discarded. In fact, the thyroglossal duct cyst in the suprahyoid area is typically located in the median area in relation to the foramen cecum. Usually the epicentre is at the base of the tongue or in the posterior part of the floor of the oral cavity. The ranula is located in the anterior portion of the sublingual space, usually lateralised. It has the appearance of a “horseshoe-shaped” cystic formation. The diving/plunging ranula develops mainly in the submandibular space with an anterior tail in the sublingual space. Furthermore, both the thyroglossal duct cyst and the ranula appear anechoic or hypoechoic, presenting slight hyper-echogenicity only in cases of infection.

Surgery was performed under general anaesthesia, with an extraoral incision, because the cyst was located under the geniohyoid muscle.

The surgical act was a median cervical incision extended for about 5 cm, detachment and isolation of the neoformation, removed by blunt way. The procedure ended with haemostasis, placement of suction drain, irrigation and layered suturing.

On macroscopic evaluation, the surgical specimen appeared as a yellowish/white smooth oval mass, measuring approximately 50 × 40 × 45 mm, soft in consistency and cystic in nature (Fig. 2A). Histopathological microscopic examination of the surgical specimen showed a cystic formation with pultaceous content, with histological features typical for epidermoid cyst as shown in Figures 2B, C. Clinical evaluation carried out 6 months after surgery showed no residual disease.

Review of the literature

Although there are manuscripts in the literature concerning

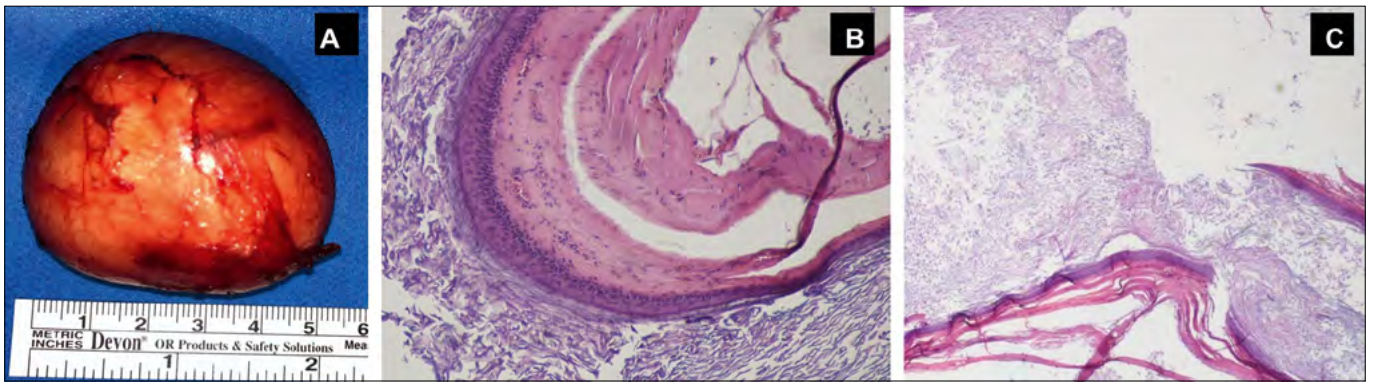


Figure 2. Surgical specimen: the cyst was entirely removed and has intact and well-defined walls (A). Histopathological microscopic appearance of the surgical specimen: cystic formation with pultaceous content (B, C).

cystic lesions of the head and neck district, there are no reviews that specifically deal with epidermoid cysts in the oral floor, focusing in particular on the diagnostic approach and therapeutic management.

All the articles published up to December 2021 reporting epidermoid cysts located in oral cavity were searched on PubMed/Medline, using as keywords “epidermoid cyst submental”, “epidermoid cyst oral cavity”, “epidermoid cyst of the floor of the mouth”.

All of the following were discarded: lesions not located in head and neck district, articles not discussing diagnostic imaging techniques, absence of histopathological diagnosis of epidermoid cysts, or absence of a full-text manuscript. Our search yielded 35 articles, in a time frame of 27 years (1993-2020), including 39 lesions in 38 patients¹⁻³⁷. Table I summarises the main data on the articles retrieved from the literature. Of 38 patients, 21 were male (55.3%) and 17 female (44.7%). Age range was 2-79 years; the mean age was 30 ± 20 years (34 in males and 29 in females).

Only in one case, reported by Sahoo et al.¹⁴, a 55-year-old woman presented with two cysts.

The mean onset time of the cysts was about 40 months. The most common symptoms reported were difficulty in swallowing (11 patients) and speech (10 patients), 5 patients presented with dysphagia, and only 3 with dyspnoea; finally, 15 patients were asymptomatic.

Sorting by anatomic position, when specified, all the 19 cysts were classified according to their position relative to geniohyoid and mylohyoid muscles in three categories:

- above the geniohyoid and mylohyoid muscle: 7 cysts;
- between geniohyoid and mylohyoid muscles: 7 cysts;
- beneath geniohyoid and mylohyoid muscles: 5 cysts.

Of these, in only 2 cases were the cysts included in both categories 2 and 3, as they herniated through the mylohyoid

muscle. In one case the cyst herniated through the geniohyoid muscle.

Figure 3 briefly outlines the possible anatomical locations and the relationships with the muscular structures, also guiding the surgical approach.

Regarding the first diagnostic approach, in 15 cases (38.46%) US was performed, in 14 (35.90%) a CT scan, and only in 9 cases (23.08%) an MRI. In several cases, additional in-depth diagnostic investigations were performed. Out of a total of 56 diagnostic investigations carried out, US and MRI were both performed in 16 cases (41%), and CT in 25 cases (64.1%), basing the percentage on the total number of cysts.

By US, the findings were: in 3 cases hypoechoic aspect, in 2 cases hyperechogenic aspect without ever finding an inhomogeneous appearance, where specified.

The most common appearance of epidermoid cyst in a CT scan was a hypodense round lesion, found in 21 cases.

While in MRI the epidermoid cysts were described as isohypointense in T1 weighted images and hyperintense in T2 weighted images, Zielinski et al.¹¹ and Sahoo et al.¹⁴ reported 3 cysts that were hyperintense in T1, Zielinski et al.¹¹ and De Ponte et al.⁵ reported 2 cases of hypointensity in T2.

Due to the wide time range of the studies reported in the literature, we do not have specific data about the appearance of the lesion in DWI sequences and in the respective ADC maps, nor in post-contrast T1w sequences.

As is known, surgical excision of lesions is definitive therapy and this depends on the relationship with the suprahyoid muscles. Of the 31 cases for which the type of surgical strategy was known, 17 cysts were excised with an intraoral approach, while 14 were approached using an extraoral technique. Of these, 3 cysts were treated both intra- and extra-orally. Only for 14 out of 39 cysts

Table I. Summary of the main data of the clinical cases from the articles selected in the literature.

Author	Gender	Age (years)	Location	Symptomatology	Size (mm)	First imaging (all imaging)	Time onset (months)	Treatment
Kandogan et al., 2007 ¹	M	11	Sublingual	Difficulty chewing and swallowing of solid foods	40 x 35	RM	36	Extra-oral
Utumi et al., 2016 ¹⁰	F	15	-	No	45	CT (CT - MR)	6	Extra-oral
Zielinski et al., 2014 ¹¹	M	6	-	No	27 x 25 x 19	US (US - MRI)	3	Extra-oral
Zielinski et al., 2014 ¹¹	F	15	-	No	29 x 26 x 24	US (US - CT)	6	Extra-oral
Baliga et al., 2014 ¹²	F	26	-	Altered speech and functional difficulty	30 x 30	US	36	Intra-oral
Patil et al., 2009 ²	M	28	-	No	30 x 20 x 20	US	5	?
Silveira et al., 2019 ¹³	M	26	Sublingual	Speech and swallowing difficulties	70 x 70	US	24	?
Sahoo et al., 2017 ¹⁴	F	55	Submental	Difficulty in speech and swallowing	40	US (US - CT - MR)	6	Intra-oral
Sahoo et al., 2017 ¹⁴			Sublingual	-	30	US (US - CT - MR)		
Verma et al., 2012 ¹⁵	F	16	-	Difficulty in chewing and swallowing of solid foods	70 x 50 x 45	CT (CT - MR)	5	Intra-oral
Mirza et al., 2014 ⁴	M	43	-	No	83 x 77 x 50	US (US - CT - MR)	-	Extra-oral
Lohaus et al., 1999 ¹⁶	F	19	-	No	50 x 50 x 38	US (US - CT - MR)	3	Extra-oral
Bitar et al., 2003 ¹⁷	F	17	Herniating through the mylohyoid, from the floor of mouth to the thyroid cartilage	Difficulty in chewing and swallowing solid food	80 x 51 x 47	CT (CT - MR)	-	Extra-oral
Calderon et al., 1993 ¹⁸	M	2	Sublingual	No	30 x 20	US	-	Intra-oral
Thibouw et al., 2020 ⁶	M	73	Sublingual	Difficulty in speaking	70 x 40 x 35	CT (CT - MR)	0.13	Intra-oral
Pascual et al., 2015 ¹⁹	F	3	Sublingual	No	20 x 15	CT (CT - MR)	2	Intra-oral
Soares et al., 2015 ²⁰	M	45	-	Limitation in mouth opening and speech, dysphagia, dyspnoea	50 x 30 x 20	CT (CT - MR)	-	Intra-oral
De Ponte et al., 2002 ⁵	M	18	-	Difficulty breathing, swallowing and speaking	45	RM	-	Intra-oral
Tsirevelou et al., 2009 ⁷	F	14	Over the geniohyoid muscle	No	14 x 12	CT (CT - MR)	10	Intra-oral
Tsirevelou et al., 2009 ⁷	F	35	-	Dysphagia, dysarthria, dyspnoea on exertion	21	CT (CT - MR)	6	Extra-oral
Datta et al., 2020 ²¹	M	24	Submandibular	Right-sided neck swelling	50 x 30	US (US - CT - MR)	8.5	Both
Findik et al., 2017 ⁹	M	10	Submandibular	No	40 x 30 x 40	RM	-	Extra-oral

continues ►

Table I. Summary of the main data of the clinical cases from the articles selected in the literature (*follows*).

Author	Gender	Age (years)	Location	Symptomatology	Size (mm)	First imaging (all imaging)	Time onset (months)	Treatment
Klibngern et al., 2020 ²²	F	26	Left sublingual space	No	65 x 32 x 25	RM	6	Intra-oral
Assaf et al., 2012 ²³	M	39	Right sublingual region	Dysphagia, progressive snoring during sleep, occasional shortness of breath	50	RM	-	Intra-oral
Tandon et al., 2014 ²⁴	F	23	From sublingual area to thyroid notch	Difficulty in swallowing, mastication, and speech	80 x 50	CT (CT - MR)	36	Intra-oral
Nishar et al., 2016 ²⁵	M	60	Submandibular region	No	103 x 81	US (US - CT - MR)	360	Extra-oral
Gulati et al., 2015 ²⁶	M	16	Left sublingual space	Difficulty in swallowing	62 x 60 x 57	RM	3	Both
Yilmaz et al., 2006 ²⁷	F	34	Left sublingual region	No	75 x 45 x 60	RM	120	Intra-oral
Yilmaz et al., 2006 ²⁷	M	35	Suprahyoid region	Difficulty in swallowing	50 x 70 x 80	RM	240	Extra-oral
Brunet-Garcia et al., 2018 ²⁸	M	43	Ssublingual space	Difficulty in swallowing, mastication, and speech	80 x 50	TC	24	Intra-oral
Behl et al., 2001 ²⁹	M	22	Sublingual space	Dysphagia, difficulties in speech	100 x 80	TC	-	Extra-oral
Turetschek et al., 1995 ³⁰	F	25	Sublingual space	Swelling, increasing difficulties in speech	60 x 50	US (US - CT - MR)	-	Intra-oral
Gordon et al., 2013 ³¹	F	79	Sublingual	Swelling	29 x 27 x 53	CT (CT - MR)	-	Extra-oral
Kinzer et al., 2006 ³²	M	40	-	No	55 x 45 x 25	RM	-	?
Kudoh et al., 2013 ³³	M	69	Submental	No	40 x 30 x 25	CT (CT - MR)	24	Intra-oral
Banerjee et al., 2011 ³⁴	F	61	Submandibular	Swelling, supine dyspnoea, almost unable to speak or eat anything including fluid	68 x 48	US (US - CT - MR)	-	Intra-oral
Mammen et al., 2013 ³⁵	M	57	Under the platysma anterior to the right-submandibular gland	Mildly painful, progressively increasing, swelling	38 x 30	US (US - CT - MR)	3	Extra-oral
Fung et al., 2008 ³⁶	M	50	Submental	Discomfort with swallowing	-	US (US - CT - MR)	Since childhood	Both
Walstad et al., 1998 ³⁷	F	23	Submental	Difficulty with swallowing and speech	48 x 27 x 35	CT (CT - MR)	-	Extra-oral

was specified the location between the muscles of the oral floor. Moreover, even when known, the surgical approach was not always planned according to the location of the lesion, for example in Kandogan et al. ¹, despite the cyst was in the sublingual space, an extra-oral approach was chosen.

Discussion

Epidermoid cysts located in the oral cavity are uncommon and mostly found in young adult patients (30 ± 20 years old) without significant gender prevalence.

Review of the literature showed that the main symptoms

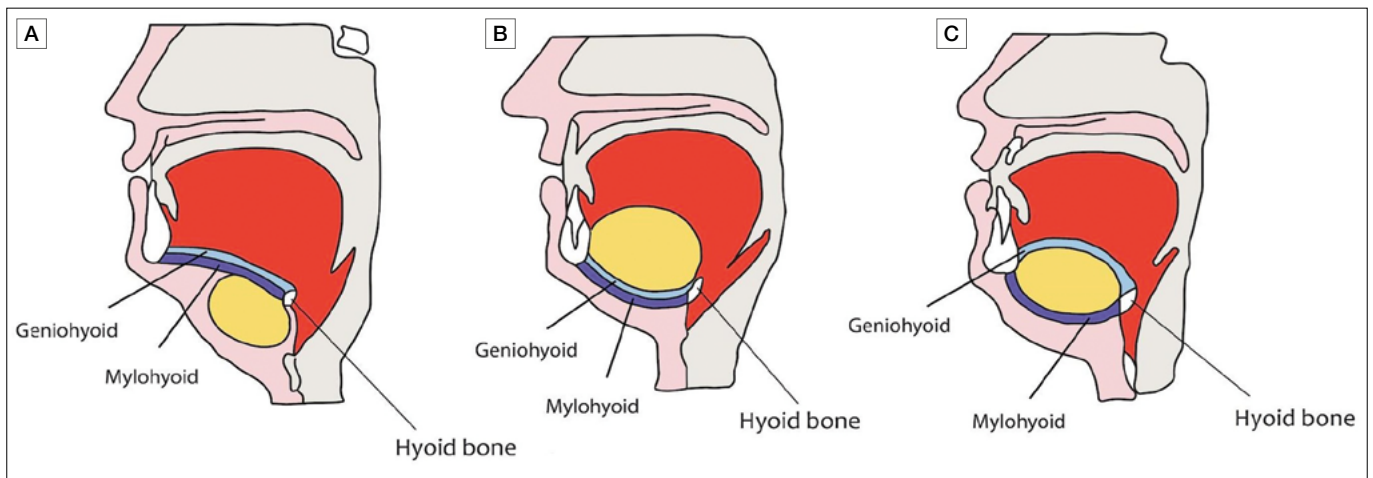


Figure 3. Different locations of epidermoid cysts of the oral cavity: under the mylohyoid and geniohyoid muscle (A); above the mylohyoid and geniohyoid muscle (B); among the mylohyoid and geniohyoid muscle (C).

Table II. Indications on when to use different imaging techniques and iconographic features of epidermoid cysts.

Indications		Characteristics of epidermoid cyst
US	First-line imaging method to detect the lesion and to formulate the main diagnostic hypothesis	Cystic well-defined lesions with variable imaging features: homogeneous hypoechoic content, hypoechoic with some hyperechoic spots, or hyperechogenic aspect. No vascularisation with colour Doppler
CT	Only if MRI is not available	Homogeneously hypodense or hypodense with hyperdense components. Sometimes with post-contrast peripheral enhancement
MRI	Second level diagnostic investigation, best imaging method to evaluate the location of the cyst and establish the surgical planning, in consideration of the position between the geniohyoid and mylohyoid muscles	Well-circumscribed, non-enhancing, cystic lesion, with homogeneous content hyperintense on T2w and iso-hypointense on T1w images, without restriction of water molecule diffusion. No signal suppression in fat-saturated images

are difficulties in swallowing and speaking, but almost half of patients complained of no symptoms.

The most interesting data emerged from review of literature is that MRI was performed in only 9 of 39 cases, although it allows precise evaluation of the position of the mass with the muscles of the oral floor (in particular mylo-hyoid and genio-hyoid) that is essential for pre-operative planning. Moreover, despite the young age of most patients, CT was performed in many more cases than MRI, and together with the US was the first line method. In 4 cases, all three imaging modalities were used.

The reasons for the lack of use of MRI can be due to the intrinsic disadvantages of the method, such as its high cost and absolute and relative contraindications of execution.

The imaging features of the lesions were found to be quite overlapping in all patients among diagnostic investigations, in particular the hypodense aspect in CT, iso-hypointense aspect in T1 weighted sequences and hyperintense aspect in T2 weighted sequences of MRI.

Considering the great advantages of MRI such as the non-use of ionising radiation, the ability to discriminate be-

tween cystic and solid lesions, reproducibility of the imaging aspects of epidermoid cysts due to the literature review, and finally the ability to accurately describe the localisation of the lesion among muscular structures surrounding the oral floor, this should be considered at least as a imaging of second level, after preliminary US examination, which is adequate for a first evaluation.

Table II presents a summary of US, CT, MRI indications and cyst epidermoid primary imaging features.

Conclusions

Epidermoid cysts of the oral cavity are rarely seen and even if they are uncommon, it is necessary to include them in the differential diagnoses among lesions of the head and neck district. As in our case, they frequently present as a solitary lesion. Otolaryngological examination and US represent the first step before surgery. MRI should be performed as a second level imaging modality, because it is essential to characterise the lesion and for surgical planning⁴. When MRI is not available or is contraindicated, CT can be use-

ful. Surgical removal of a sublingual epidermoid cyst is the treatment of choice and the approach depends on the size and location of the lesion. In fact, whether extraoral or intraoral incision is preferred depends on the position of the lesion in relation to the muscles of the floor of the mouth (primarily mylohyoid and geniohyoid). Therefore, the radiological report should describe the relationship between the mass and these suprahyoid muscles.

Conflict of interest statement

The authors declare no conflict of interest.

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Author contributions

MGS, DS, LP, MB: conception and design; NB, MA, MC, MGS, DS, MB, SM: provision of study materials or patients; NB, MA, MC: collection and assembly of data; NB, MA, MC: data analysis and interpretation; all authors: manuscript writing.

Ethical considerations

The research was conducted ethically; the patient has filled out informed consent before carrying out the instrumental investigations, in accordance with current European regulations, agreeing to voluntary participation in the study and publication of data in anonymous form.

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