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Surgical Ciliated Cyst of the Jaws: A Systematic Review

Çenelerin Cerrahi Silyalı Kisti: Sistematik Bir İnceleme

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ABSTRACT

Objective: Cysts are fluid-filled lesions located in bone or soft tissue, surrounded by an epithelial-lined capsule. Surgical ciliated cyst (SCC) is a rare, non-odontogenic cyst lined with respiratory epithelium, formed as a result of traumatic implantation of the sinus or nasal mucosa into the jawbone especially maxilla and it was included in the latest WHO classification in 2022. The present study was aimed to systematically review the cases of surgical ciliated cysts.

Method: A literature review on the search engine Web of Science, PubMed, Google Scholar and Clinical Key was conducted, without date restrictions including search terms "surgical ciliated cyst maxilla" or "surgical ciliated cyst mandible". The records were examined in detail as full text according to inclusion and exclusion criteria. Inclusion criteria; lesions that were definitively diagnosed as SCC as a result of histopathological examination. Exclusion criteria; lesions without a history of surgical procedures.

Results: The total number of records generated was 150 and after the duplicates were removed, 85 records left. These records were carefully examined according to inclusion and exclusion criteria and as a result a total of 39 records and 49 cases of surgical ciliated cysts were included in the systematic review. 84% of the cases were seen in and around the maxilla, and very few were in the mandible.

Conclusion: SCC is not a new entity; maxillofacial radiologists, surgeons and pathologists should be familiar with the clinical, radiologic, histopathologic features and medical history of the SCC cases.

Keywords: Respiratory mucosa; jaw; cyst

ÖZ

Amaç: Kistler kemik veya yumuşak dokuda yer alan, epitelle kaplı bir kapsülle çevrelenmiş sıvı dolu lezyonlardır. Cerrahi siliyer kist (CSK), sinüs veya nazal mukozanın travmatik olarak çene kemiğine, özellikle maksillaya implantasyonu sonucu oluşan, solunum epiteli ile döşeli, odontojenik olmayan nadir bir kisttir ve 2022 yılında WHO sınıflandırmasına dahil edilmiştir. Bu çalışmada, cerrahi siliyer kist vakalarının sistematik olarak gözden geçirilmesi amaçlanmıştır.

Yöntem: Web of Science, PubMed, Google Scholar ve Clinical Key arama motorunda, tarih sınırlaması olmaksızın "cerrahi siliyer kist maksilla" veya "cerrahi siliyer kist mandibula" İngilizce arama terimlerini içeren bir literatür taraması yapılmıştır. Kayıtlar, dahil etme ve hariç tutma kriterlerine göre tam metin olarak detaylı bir şekilde incelenmiştir. Dahil edilme kriterleri; Histopatolojik inceleme sonucunda kesin olarak CSK tanısı konan lezyonlar. Dahil edilmeme kriterleri; cerrahi işlem öyküsü olmayan vakalar.

Bulgular: Oluşturulan toplam kayıt sayısı 150 olup, kopyalar çıkarıldıktan sonra 85 kayıt kalmıştır. Bu kayıtlar dahil etme ve hariç tutma kriterlerine göre dikkatlice incelenmiş ve sonuç olarak toplam 39 kayıt ve 49 cerrahi siliyer kist vakası sistematik derlemeye dahil edilmiştir. Vakaların %84'ü maksilla ve çevresinde, çok azı ise mandibulada görüldü.

Sonuç: CSK yeni bir lezyon olmamakla birlikte; maksillofasiyal radyologlar, cerrahlar ve patologlar CSK'nin klinik, radyolojik ve histopatolojik özelliklerine ve hasta öyküsüne vakıf olmalılardır.

Anahtar kelimeler: Solunum mukozası; çene; kist.

INTRODUCTION

Cysts are fluid-filled lesions located in bone or soft tissue, surrounded by an epithelial-lined capsule. Intrabone cysts are most commonly seen in jaws. Jawbone cysts may originate from odontogenic or non-odontogenic epithelial sources. Surgical ciliated cyst (SCC), which was included in the latest WHO classification in 2022, is not actually a new entity. This cyst is a rare, non-odontogenic cyst lined with respiratory epithelium, formed as a result of traumatic implantation of the sinus or nasal mucosa into the jawbone. It occurs as a result of the implantation of sinus epithelium into the bone with contaminated instruments during operations such as orthognathic operations, sinus operations, and trauma. It is also known as postoperative maxillary cyst or respiratory implantation cyst. It is more common in the 5th and 6th decades and no gender distinction is observed.[1] It is most common in the posterior maxilla, and is rarer in the mandible. Histopathologically, the cyst is lined with ciliated pseudostratified columnar epithelium

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and mucous cells are common. Previous surgical history is an important criterion for diagnosis. Treatment is simple enucleation and recurrence is rare.

In this article, the authors aimed to systematically review the cases of surgical ciliated cysts, which have recently been included among the non-odontogenic cysts of the jaws by WHO.

METHODS

A literature review on the search engine Web of Science, PubMed, Google Scholar and Clinical Key was conducted, On September 19, 2023 without date restrictions including search terms "surgical ciliated cyst maxilla" and "surgical ciliated cyst mandible". The flow-chart for review process is seen on Figure 1. The total number of records generated was 150 and after the duplicates were removed, we had 85 records left. 18 records excluded based on the title and abstract review and 67 records left. These records were examined in detail as full text according to inclusion and exclusion criteria. Inclusion criteria; lesions that were definitively diagnosed as SCC as a result of histopathological examination. Exclusion criteria; cases without a history of surgical procedures. Based on these criteria, 28 more articles were eliminated and finally 39 records and 49 cases were included (Table 1) in the systematic review. As a result, the remaining records we had covered the years 2005-2023.

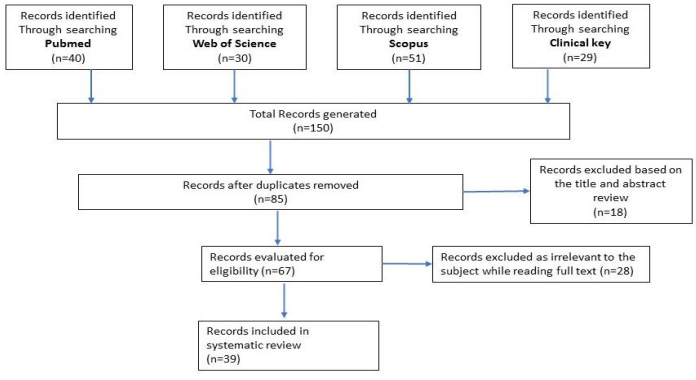

Figure 1. Flow-chart of the review process.

RESULTS

A total of 39 records and 49 cases of surgical ciliated cysts were included in the systematic review. General characteristics of the cases are seen in Table 1. Table 2 summarizes the general descriptive characteristics of the cases. Accordingly, the mean age of the patients with surgical ciliated cyst was 45.02±13.65 (min: 20, max: 76) years. 26 (53.06%) of the patients were male, 19 (38.77%) were female, and the gender information of 4 (8.17%) patients could not be obtained. When the localizations of the cases were examined, 39 of the surgically ciliated cysts were in the maxilla, 8 in the mandible, and 2 around the maxilla (1 medial canthal, 1 infratemporal). Namely, 84% of the cases were seen in and around the maxilla. When the surgical stories were examined, the patients had past stories such as maxillary sinus surgery, genioplasty, traumatic tooth extractions, rhinoplasty, trauma, orthognathic surgery. The average time from surgical history to diagnosis was 14.57±11.78 years, with a minimum of 5 months and a maximum of 56 years. When the dimensions of the lesions were examined, the maximum size was 4.9 cm, although size information was not available for each lesion. While 4 of the lesions appeared radiologically multilocular, the majority were in the form of unilocular radiolucency. Additionally, multifocal lesions were observed in 5 patients. When the treatment methods are examined; It was observed that 38 (77.55%) of the cases were treated with enucleation, 5 (10.20%) with curettage, and 3 (6.12%) with marsupialization. No information could be obtained about the treatment method of 3 (6.12%) cases. Three cases showed recurrence after treatment.

DISCUSSION

Jaw bones are likely to contain epithelial cell residues, whether due to odontogenesis, proximity to oral mucosa or the closure of embryonal clefts. For this reason, they are the bones with the most cysts compared to other bones in the body. Cysts seen in the jaws are classified as odontogenic and non-odontogenic and the majority of them are of odontogenic origin. SCC is a non-odontogenic cyst seen in jaws and is included the in the latest classification of WHO. Although this lesion was actually a previously known lesion, it was included in the classification in 2022. The lesion is most common in the 5th-6th decades and patients have a history of previous surgery.[1]

It occurs as a result of the inclusion of the respiratory epithelium into the jaw bones, especially after surgical procedures involving the maxilla. It is stated that SCC develops in 3-20% of patients who undergo maxillofacial surgery.[2] The lesion, which radiologically appears as well-circumscribed radiolucent defects, is histopathologically lined with respiratory epithelium. SCC was first reported by Kubo in 1927.[3] The oldest case found in this systematic review was an SCC of the hard palate reported by Hayhurts et al in 1993.[4] In this case, the authors presented a case of SCC occurring in the hard palate in association with the nasal mucosa in a patient who underwent LeFort 1 osteotomy. However, since we could not access this article in full text and could not obtain the patient's data, we could not include it in the revision table.

SCC usually occurs between the ages of 40 and 60.[1] Indeed, in the systematic analysis of the cases in the literature, we found that the majority of patients were in this age range. The mean age of the patients was 45.02±13.65 years. The youngest patient with SCC was 20 years old[5] and the oldest patient was 76 years old.[6] In this systematic review, we found that the majority of SCC cases reported in the literature were males.

SCC is also known as post-operative maxillary cyst and is more common in the maxilla. This is due to the close proximity of the maxilla to airways such as the nasal mucosa and sinus mucosa. In the mandible, it occurs when instruments contaminated with respiratory mucosa invade the mandible during large multifocal operations. The vast majority of cases in the literature were in and around the maxilla also.[5-35] One case was in the medial canthus[36] and one case in the infratemporal fossa.[37] These can also be considered to be around the maxilla. The majority of SCC cases in the maxilla had a history of orthognathic surgery, sino-nasal surgery, sinus augmentation. Four patients had a history of traumatic tooth extraction[6,16,28,33] one patient had a history of apicoectomy[16] and two patients had a history of zygomaticomaxillary fracture.[14,21] It has been demonstrated that during the healing of comminuted zygomaticomaxillary fractures, the torn sinus mucosa becomes trapped between the bone edges of the maxilla and degenerates into the cyst.[14] Also in the maxilla, one of the cases of SCC reported by Archambault et al.[31] occurred after cleft palate surgery and one after implant surgery.

In the systematic review, we identified 8 cases reported in the mandible.[2,3,31,38-41] All of these patients had a history of orthognathic surgery, one case[38] had a history of genioplasty and rhinoplasty. In fact,

Table 1. Surgical ciliated cyst cases included in the systematic review.

	Published	Gender	Age	Surgical procedure	Location	Dimension and radiographic appearance	Time between surgery and diagnosis	Treatment and prognoses
Cai et al.	2015	Male	21	LeFort1/Genioplasty	Mnd	2 lesions unilocular	28 months	Curettage
Leung,el al.	2012	Female	45	Caldwell luck	Mx/left/premolar molar	unilocular/alv.region/below max.sinus	1 year	Enucleation
Leung,el al.	2012	Male	28	Caldwell luck	Mx(right/anterior	2.1x1.2x1.3 cm unilocular	12 years	Enucleation
Leung,el al.	2012	Female	49	Caldwell luck	Mx/left/premolar molar	3.4x1.7x2.1 unilocular	25 years	Enucleation
Li et al.	2014	Male	72	Rhinoplasty/Genioplasty	Mnd/Anterior	3 cm unilocular	56 years	Curettage
Li et al.	2014	Male	42	LeFort1/SSOste	Mnd/right/ramus	2.7x0.9 cm unilocular	18 years	Curettage recurrence
Bourgeois and Nelson	2005	Female	27	LeFort1/genioplasty	Mnd/Anterior	2 areas/0.8x0.8-1.0x1.0 cm unilocular	4 years	Enucleation
Theofilou et al.	2021	Female	48	LeFort1/SSOste	Mx/right/left	2 lesions/2,3x1,8x1,7 cm-2x2x1,3 cm unilocular	13 years	Enucleation
Theofilou et al.	2021	Female	50	LeFort1/SSOste	Mx/right/tuber	2 lesions/1,1x0,9x0,6 cm-1,1x1x0,9 cm unilocular	10 years	Enucleation
Theofilou et al.	2021	Male	20	LeFort1	Mx/right/left	2 lesions/0,7x0,6x1,6 cm-3x,16x0,9 cm unilocular	3 years	Enucleation
Lafuente-Ibanez de Mendoza et al.	2021	Male	67	Sinus augmentation	Mnd/right/posterior	1.2x1.0.x1.0 cm unilocular	2 years	Curettage
Youn et al.	2022	Male	42	Orthognathic surgery/genioplasty	Mnd/Anterior	>2 cm unilocular	24 years	Enucleation
Yamamoto et al.	2017	Female	55	Sinus augmentation	Mx/left	bilocular	9 years	Enucleation
Ramakrishnan et al.	2020	Female	76	Tooth extraction	Mx/left	3,5x3,1x3,4 cm unilocular	5 months	Enucleation
Kwack et al.	2023	Male	40	Lefort1	İnfratemporal fossa/right	2.8x5.2x3.8 cm unilocular	25 years	Marsupialization
Cordero-Garcia et al.	2023	Female	41	LeFort1/SSOste	Mx/anterior	4.3x3.7x3.7 cm unilocular	20 years	Enucleation
Ragsdale et al.	2009	Male	30	LeFort1/	Mnd/extend.right to left	large /anterior unilocular	16 years	Enucleation
Cano et al.	2009	Female	56	Caldwell luck	Mx/right	?	3 years	Enucleation
Adachi et al.	2016	Female	44	Caldwell luck	Mx/left	trilocular	21 years	Enucleation
Golaszewski et al.	2019	Female	28	LeFort1	Mx/right/anterior	unilocular	5 years	Enucleation
An and Zhang	2014	Male	42	Midfacial fracture	Right/medial canthal	2.5 cm unilocular	10 years	Enucleation
Peighoun,Sahand et al.	2022	Male	73	Nasal polyp surgery	Mx/left/posterior	?	40 years	Enucleation
Toyoshima et al.	2014	Female	56	Zygomaticomaxillary fracture	Mx/left	multilocular	30 years	Enucleation
Tanio et al.	2019	Male	25	LeFort1	Mx/right/posterior	unilocular(15 mm)	5 years	Enucleation
Redman and Rodriguez	2020	Male	53	Tooth extraction	Mx/left/anterior	2.5x3 cm unilocular	?	Enucleation
Redman and Rodriguez	2020	Male	47	Apicoectomy	Mx/left/posterior	2.5x3 cm unilocular	23 years	Enucleation
Fernandes et al.	2013	Male	63	Mx.sinus surgery	Mx/right	?	15 years	Enucleation
Koo Min Chee et al.	2014	Male	42	Bimaxillary osteotomy	Mx/right	?	20 years	Enucleation
Moe et al.	2013	Male	42	LeFort1	Mx/right/posterior	1.3x1.5 cm unilocular	20 years/2 years rady.dia	Enucleation
Coviello et al.	2017	Male	53	LeFort1	Mx/anterior/central	2.5x4.0x1.5 cm unilocular	12 years	Enucleation
Vieira et al.	2022	Female	51	Zygomatic fracture	Mx/right/zygomatico-orbital	?	10 years	Enucleation
Vieira et al.	2022	Male	43	LeFort1	Mx/left/anterior	?	12 years	Enucleation recurrence
Siwach et al.	2020	Female	65	Caldwell luck	Mx/right/posterior	?	30 years	Enucleation
Soares et al.	2021	Female	39	LeFort1	Mx/right/left	2 lesions 4,9x2,4 cm/1,9x1,3 cm unilocular	6 years	Enucleation
Soares et al.	2021	Male	38	LeFort1	Mx/right/anterior	?	4 years/first diagn.2 year	Enucleation recurrence
Lim and Ngeow	2017	Male	43	Mx sinus surgery	Mx/right/posterior	3,5x3,1x3,4 cm unilocular	30 years	Enucleation
Yang et al.	2018	Female	35	Bimaxillary osteotomy	Mx/bilateral /posterior	?	10 years	Marsupialization
Pakravan and Nafarzadeh	2017	Male	49	Mx sinus surgery	Mx/left/posterior	4x1.5 cm unilocular	30 years	Curettage
Park and Lim	2021	Male	56	Sinus augmentation	Mx/left/posterior	?	18 years/13 years small	Marsupialization
Alamri et al.	2022	Male	39	Tooth extraction	Mx/left/posterior	0,93x0,78 cm unilocular	?	Enucleation
Shakib et al.	2009	Female	28	LeFort1	Mx/anterior	2.5x1.9x2.2 cm unilocular	7 years	Enucleation
Amin et al.	2014	Male	53	Mx sinus surgery	Mx/right	0,6 cm unilocular	10 years	Enucleation
Archambault et al.	2017	?	?	Cleft surgical repair	Mx/anterior	?	?	?
Archambault et al.	2017	?	?	Implant surgery	Mx/posterior	?	?	?
Archambault et al.	2027	?	?	Orthognathic surgery	Mnd/	?	?	?
De Arruda et al.	2017	?	47	Odontogenic cyst surgery	Mx/left/posterior	?	9 months	Enucleation
Silva et al.	2020	Female	32	Tooth extraction	Mx/left/posterior	1.3 cm unilocular	?	Enucleation
De Pinho et al.	2022	Female	51	Zygomatic fracture	Mx/right/zygoma	?	10 years	Enucleation
Da Silva Leonel et al.	2022	Male	25	Orthognathic surgery	Mx/right	?	4 years	Enucleation

Mnd: Mandible, Mx: Maxilla

Table 2. The summary of the general descriptive characteristics of the cases.

Age	45.02±13.65(min: 20, max: 76)
Gender	26 (53.06%) male, 19 (38.77%) female, 4 (8.17%) unclear
Location	39 maxillary, 8 mandibular, 1 medial canthal, 1 infratemporal
Time between surgery and diagnosis	14.57±11,78years (min: 0.42, max: 56)
Treatment	38 (77.55%) enucleation, 5 (10.20%) curettage, 3 (6.12%) marsupialization, 3 (6.12%) unclear

the occurrence of SCC in the mandible is quite rare. Tissues containing airway epithelium can be transplanted into the mandible during operations involving both maxilla and mandible, such as Le Fort I osteotomy and genioplasty. Cai et al.[3] proposed that transplantation of respiratory epithelium attached to the graft had proliferated in the favorable healing environment of the grafted site. Nastri and Hookey[42] first reported a respiratory tract epithelium in the wall of a cyst in the mandible in 1993 after grafting of autogenous bone. Li et al.[38] reported two cases of SCC in the mandible. One of the cases had a history of rhinoplasty and genioplasty, the other had a history of LeFort I osteotomy, turbinectomy and mandibular sagittal split osteotomy in the same session. Bourgeois at al.[39] also reported SCC in the mandible in a patient with a history of Lefort I osteotomy and genioplasty. Ragsdale et al.[41] and Archambault et al.[31] also reported mandibular SCC after orthognathic surgery. Lafuente-Ibanez de Mendoza et al.[2] presented a case of SCC in a 67-year-old man after surgical treatment of an endo-periodontal radiolucent lesion in the mandibular posterior region. Interestingly, the patient had no history of major maxillofacial surgery.

In the literature, there were several other cases of cysts lined with ciliated epithelium without any history of surgery, trauma or sinusitis.[43-45] Since there was no surgical history in these cases, we did not accept them as SCC and did not include them in the review. All of these cases were in the maxilla. Epithelial remnants possibly due to anatomical proximity to the sinuses during embryonic life may be responsible. It was stated that small segments of sinus mucosa may become trapped in the bony margins of the maxilla sometimes.[14]

Odontogenic cysts such as dentigerous cysts, odontogenic keratocyst and glandular odontogenic cysts should be considered in the differential diagnosis of SCC.[38] Cysts generally appear as unilocular, regularly circumscribed radiolucent lesions. SCC likewise presents as regularly circumscribed unilocular radiolucency. Peighoun et al.[13] presented a case of SCC mimicking a residual cyst or odontogenic keratocyst. The definitive diagnosis could only be made by histopathologic examination. The majority of cases in the literature were unilocular radiolucent lesions, with a few cases appearing multilocular.[8,11,14,23] Considering the size of the available cases, the largest case was the lesion in the anterior maxilla after Leforrt I osteotomy reported by Cordero Garcia et al.[9]

The interval between the history of surgical operation and the diagnosis of the lesion ranged from 5 months to 56 years. Of course, the factors that initiate cystic degeneration of the epithelial cell are still not clearly known today. The p53 gene has been implicated in the formation of many jaw cysts.[46] For example, it is interesting that non-odontogenic developmental cysts formed from epithelial remnants of embryonal life occur at an advanced age. In the occurrence of SCC here, the interval between the surgical history and the diagnosis of SCC was quite variable. It may be that some people are genetically more prone to cyst formation.

When the treatment modalities of the cases in the literature were evaluated, it was seen that they were mostly treated with enucleation. Only five lesions were treated with curettage and three with marsupialization. Recurrence was seen in three cases.[21,23,38]

In conclusion, maxillofacial radiologists, surgeons and pathologists should be familiar with the clinical, radiologic and histopathologic features of the newly classified SCC. This lesion should be considered in the differential diagnosis of lesions in the maxilla and even in the mandible. The medical history and previous operations of the patients should be examined in detail.

Ethics Committee Approval: Since it is a review study, there is no ethics committee approval.

Informed Consent: No patient data was used for the study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – F.Ç.; Design - F.Ç.; Supervision - F.Ç.; Resources - F.Ç.; Materials - H.L.T.D.; Data Collection and/or Processing - H.L.T.D.; Analysis and/or Interpretation – F.Ç.; Literature Search - F.Ç.; Writing Manuscript - F.Ç.; Critical Review - F.Ç.; Other – H.L.T.D.

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