Choristomas of Oral Cavity: Origins and Derivates (Literature Review)

Oral Kavitenin Koristomaları: Kökenler ve Türevler (Literatür İncelemesi)

Myroslav GONCHARUK-KHOMYN[a],
İzzet YAVUZ[b,c],
Alessandro Leite CAVALCANTI[d]

[a]Uzhhorod National University Medical Faculty, Department of Public Health and Humanitarian Disciplines, Uzhhorod, UKRAINE
[b]Dicle University Faculty of Dentistry, Department of Pediatric Dentistry, Diyarbakır, TÜRKİYE
[c]Dean of Harran University Faculty of Dentistry, Şanlıurfa, TÜRKİYE
[d]State University of Campina Grande-Paraiba, School of Dentistry, Department of Dentistry, Campina Grande, BRAZIL

Yazışma Adresi/*Correspondence:*
Myroslav GONCHARUK-KHOMYN
Uzhhorod National University Medical Faculty, Department of Public Health and Humanitarian Disciplines, Uzhhorod, UKRAINE
myroslav.goncharuk-khomyn@uzhnu.edu.ua

ABSTRACT Choristoma represents proliferation of histologically normal tissues with no neoplastic pattern but heterotopic localization. Search of literature sources associated with formulated topic of oral cavity choristomas was provided via PubMed database (https://pubmed.ncbi.nlm.nih.gov/) and Google Scholar search engine (https://scholar.google.com/). Search algorithm using Mesh-terms in PubMed was provided as following: (“choristoma”[MeSH Terms] OR “choristoma”[All Fields]) AND (“mouth”[MeSH Terms] OR "mouth"[All Fields] OR “oral”[All Fields]). 64 articles were included in this review, out of which 3 were solid literature reviews (4,69%), 16 were case reports with accompanied literature review (25,0%), 44 were case reports (68,75%), and 1 was systematic review (1,56%). Oral cavity choristomas could be developed from bone, cartilage, brain tissue, glia, gastric mucous, epidermis, and tooth-like structures, or have salivary and sebaceous gland origin. Osseous and cartilaginous types of choristomas have been the most frequently reported within available literature in the form of clinical cases and associated reviews. Choristomas of oral cavity and such of oral cavity origin, but located within other organs and systems, characterized with low level prevalence, nevertheless, risk of such lesions development should always be considered during the process of differential diagnosis. Deficiency of systematized data regarding oral cavity choristomas limiting possibilities for prognosis regarding their recurrence or potential malignization, even though only individual clinical reports demonstrating choristomas relapses after full excision and their potential for getting infected or irritated.

Keywords: Choristoma; oral manifestations

ÖZET Koristoma, neoplastik patern olmayan ancak heterotopik yerleşimli histolojik olarak normal dokuların proliferasyonunu temsil eder. Formüle edilmiş oral kavite koristomları konusuyla ilgili literatür kaynaklarının araştırılması, PubMed veritabanı (https://pubmed.ncbi.nlm.nih.gov/) ve Google Akademik arama motoru (https://scholar.google.com/) aracılığıyla sağlandı. PubMed'de Mesh terimlerini kullanan arama algoritması şu şekilde sağlandı: (“koristoma”[MeSH Terimleri] VEYA “koristoma”[Tüm Alanlar]) VE (“ağız”[MeSH Terimleri] VEYA “ağız”[Tüm Alanlar] VEYA “oral” [Tüm alanlar]). Bu derlemeye 64 makale dahil edildi, bunlardan 3’ü sağlam literatür taraması (%4,69), 16’sı literatür taraması eşliğinde olgu sunumu (%25,0), 44’ü olgu sunumu (%68,75) ve 1 sistematik incelemeydi (%1,56). Ağız boşluğu koristomları kemik, kıkırdak, beyin dokusu, glia, mide mukozası, epidermis ve diş benzeri yapılardan gelişebilir veya tükürük ve yağ bezi kökenliydi. Kemikli ve kıkırdaklı koristoma türleri, mevcut literatürde klinik vakalar ve ilgili incelemeler şeklinde en sık bildirilenlerdir. Ağız boşluğu ve ağız boşluğu orijinli, ancak diğer organ ve sistemler içinde yer alan, düşük prevalans ile karakterize olan koristomlar, yine de, ayırıcı tanı sürecinde bu tür lezyonların gelişme riski her zaman göz önünde bulundurulmalıdır. Oral kavite koristomları ile ilgili sistematik verilerin eksikliği, nüks veya potansiyel malignizasyon ile ilgili prognoz olasılıklarını sınırlamaktadır, ancak sadece koristomaların tam eksizyondan sonra nüksettiğini ve enfekte veya tahriş olma potansiyellerini gösteren bireysel klinik raporlar bildirilmiştir.

Anahtar Kelimeler: Koristom; ağız belirtileri

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Choristoma represents proliferation of histologically normal tissues with no neoplastic pattern but heterotopic localization.[1-6] Another definition of choristoma includes aggregation of histologically/microscopically normal cells

or tissues within organ or tissues which are unusual/abnormal for such cells conglomerates.[4-7] First report about choristoma was provided by Berry, who in 1890-1892 described cartilaginous choristoma, while by some other literature sources in 1885 Zahn presented the first case of cartilaginous conglomerate within oral soft tissues.[8-13] In 1913 osseous choristomas of the tongue were firstly described as lingual osteomas.[4,14] Due to the Chou et al. several classes of choristoma could be distinguished: salivary gland choristomas (either central or gingival), cartilaginous, osseous, lingual thyroid, lingual sebaceous, glial, gastric mucosal (either cystic or solid).[1,15,16]

Considering the rare prevalence of choristomas and their predominant representation in the form of case reports within scientific literature there is a reasonable need for providing relevant review on this topic, which could help to systematize currently available data, highlight main clinical features obtained within nearest years and compare them with previously reported outcomes, accent some new observations not issued previously and describe some new diagnostic approaches.

OBJECTIVE

To systematize available data dedicated to the choristomas of oral cavity and allocate key aspects of pathogenesis, prevalence parameters, clinical characteristics and diagnostics of such lesions through detailed retrospective analysis of the literature.

MATERIALS AND METHODS

Search of literature sources associated with formulated objective was provided via PubMed database (https://pubmed.ncbi.nlm.nih.gov/) and Google Scholar search engine (https://scholar.google.com/). Search algorithm using Mesh-terms in PubMed was provided in the following form: (“choristoma”[MeSH Terms] OR “choristoma”[All Fields]) AND (“mouth”[MeSH Terms] OR “mouth”[All Fields] OR “oral”[All Fields]). Search within Google Scholar was held by the such keywords as “choristoma”, “oral” and “oral cavity”. Criteria of publication date was not used. Only publications in English or at least with English abstract/summary were comprehended in the final sample for further content-analysis. Grouped amount of literature sources was investigated due to the standard manual content-analysis protocol, categories of which considered aspects of pathogenesis, prevalence parameters, clinical characteristics and diagnostics of oral cavity choristomas. For the evidence-expansion purpose we have additionally used Connected Papers service (https://www.connectedpapers.com/), which helped to analyze related, prior and derivates works for all targeted publications (Figure 1).

Microsoft Excel 2019 (Microsoft Office, 2019) software was used for the systematization and categorization of data and formulation of intercourses and links between different publications’ facts and evidences regarding relevant and significant information.

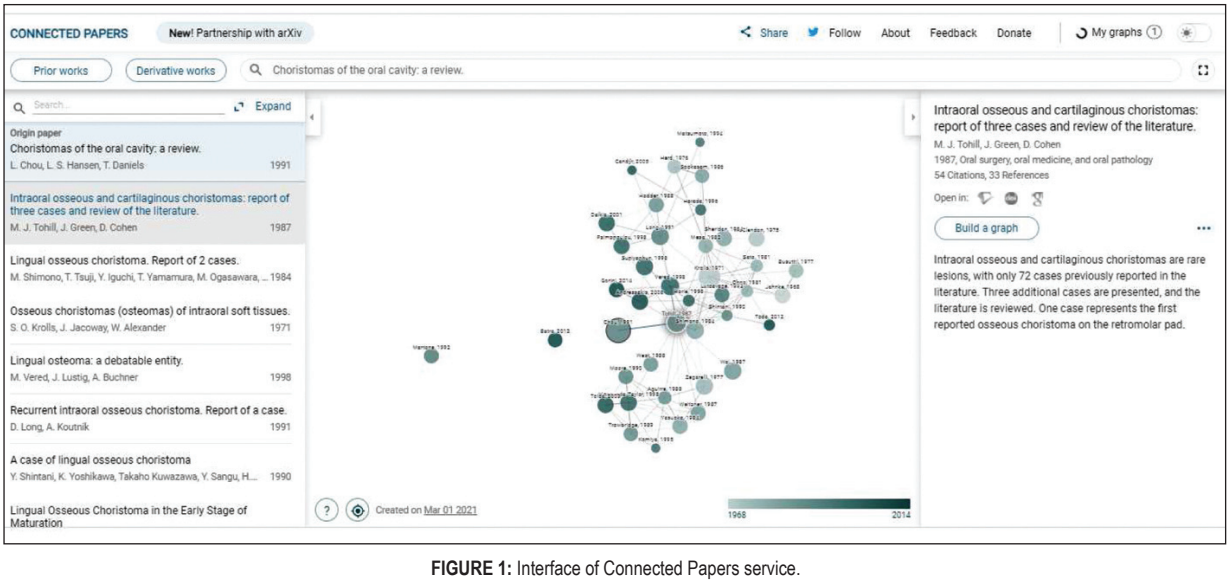

FIGURE 1: Interface of Connected Papers service.

RESULTS

2201 results of the search were found and distributed as follows: 591 in PubMed and 1610 in Google Scholar (Figure 2).

After removing duplicates, 1803 publications remained, of which 416 were selected by analyses of titles and summaries/abstracts. Other 1387 articles were excluded, because they were not associated with the topic of choristomas of oral cavity. All of 416 articles identified were analyzed in full manner. After full reading of article texts 102 of them were excluded because of not presenting information due to the specifically formulated review objective. Out of 314 completed articles accessed by eligibility, 250 were excluded because of duplicating results, or demonstrating data analogically or similarly represented in other articles, or not representing results relevant to up-to-date actual continuity regarding oral cavity choristomas. At the end, 64 articles were included in this review, out of which 3 were solid literature reviews (4,69%), 16 were case reports with accompanied literature review (25,0%), 44 were case reports (68,75%), and 1 was systematic review (1,56%) (Figure 3).

DISCUSSION

ORIGIN AND DEVELOPMENT OF ORAL CAVITY CHORISTOMAS

At the early research stages of choristomas as targeted investigation topic such lesions were presumed to be either "developmental, neoplastic or reparative by their nature".[17] Oral cavity choristomas could be developed from bone, cartilage, brain tissue, glia, gastric mucous, epidermis, and tooth-like structures, or have salivary and sebaceous gland origin.[18] Osseous and cartilaginous types of choristoma have been the most frequently reported within available literature in the form of case reports and associated reviews.

Relevant theories of choristomas' development include following: possible differentiation of pluripotent precursor cells from the mesenchyme; chronic inflammatory, trauma or other factors of irritation, which could cause the release of osteogenic or chondrogenic substances within normal tissue with further de novo tissue formation (considering possibility of progenitor mesenchymal cell to proliferate in different well-organized tissue); spontaneous embryonic residuals proliferation.[1,3,4,6,19] Trauma theory projected on lingual choristomas specifically has been supported by analogical mechanism of "myositis ossificans" formation within different organs and systems of the body.[4,2,20-23] Possible development of lingual choristomas presumed to have associations with movement of tongue during speech articulation that may lead to inflammation progression with further calcium deposition. But results of histological examination revealed that choristomas are formed of well-developed bone tissue, but not just from calcium elements.[3] Krolls et al. also mentioned that such choristomas could possibly occur because of blood vessel calcium transmission on the background of long-standing somatic pathology existence (such as hyperparathyroidism).[24] Etiology of lingual osseous choristomas may also be associated with metabolic alteration, ossification of branchial arch remnants, epignathus formation and degeneration of ossifying fibroma.[25]

Possible pathogenesis variants for osseocartilaginous types of choristomas includes proliferation of embryogenic remnants, which have preserved their differentiation po-

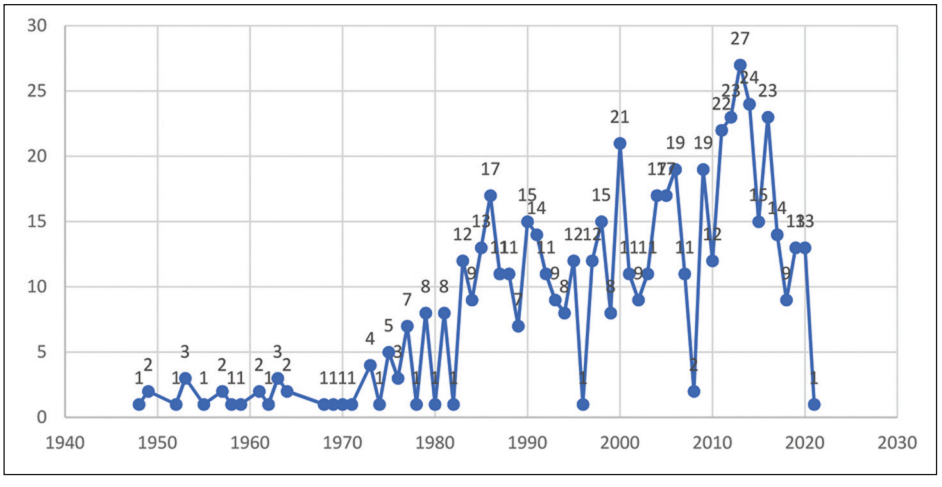

FIGURE 2: Distribution of publications found within PubMed database dedicated to the topic of choristomas of oral cavity.

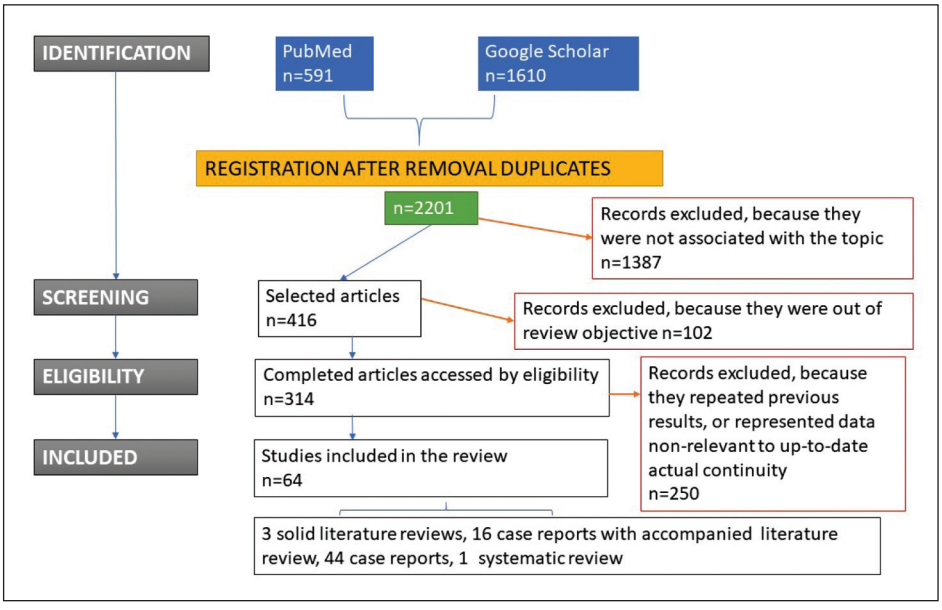

FIGURE 3: Flowchart of selection procedure for the articles included in the review.

tential; mesenchymal primary cell derivation; metaplasia caused by irritation of different nature; mixed neoplasms; neoplasms or teratomas associated with cartilage tissue.[26] Also, cartilaginous choristoma have been associated with chromosomal aberrations within 12q13-q15.[27]

EPIDEMIOLOGICAL CHARACTERISTICS OF ORAL CAVITY CHORISTOMAS

Age of patients with cartilaginous choristoma varies from 10 to 80 years.[28] Osseous choristomas of the tongue were registered among patients aged 5-89 years old with mean of 28,9 years. In the systematic review patients' age range for osseous lingual choristomas was 5-73 years with mean of 28,1 years.[6] Most cases of osseous lingual choristomas were registered specifically among age group of 20-29 years.[6]

Due to the literature data male/female ratio of choristomas prevalence locates at the level of 19:50.[29] Shareef et al. specified that 70% of all reported cases of osseous tongue choristomas were registered among female patients [6]. In Heinz's publication authors also highlighted female predilection for osseous choristomas of tongue with male/female ratio at the level of 16 to 44.[30] Basically, osseous choristomas of the tongue registered among females in 2,7-4 times higher than among males.[31] Generally due to the literature data osseous choristomas demonstrated greater prevalence among females, while cartilaginous and osseocartilaginous types characterized with analogical registration frequency among both sexes.[26]

CLINICAL AND HISTOLOGICAL FEATURES OF ORAL CAVITY CHORISTOMAS

Analysis of 38 cases of lingual choristomas revealed next distribution of their localization over tongue: 67,5% - dorsum, 25% - lateral aspects, 7,5% - mid-third.[21] During the analysis of the literature in 2019 osseous choristomas of the tongue were located in 88,2% cases at the posterior 1/3, in 7,4% cases - in middle 1/3, in 2,9% cases - at lateral borders, in 1,5% - at the floor of the tongue.[32] Due to the data obtained in systematic review most lingual osseous choristomas locate on posterior or lateral aspects of the tongue with base or pedunculi connecting it to the tongue surface.[6] Based on the previous reports osseous choristomas of the tongue were prevalently located at the dorsum aspect often aligning among circumvallate papillae or near foramen caecum.[6] Turan et al. noted that in most cases lingual osseous choristomas are located behind foramen caecum.[31] Tongue base also have been reported as potential localization of osseous choristomas in few studies.[3,30,33-36] Localization of the osseous choristomas within tongue dorsum may be explained by the following embryogenetic predisposition: lesions usually appears between the anterior 2/3 of tongue, which is formed from 1st brachial arch, and posterior 1/3 of the tongue, which is formed from 3rd brachial arch. During the development of such structures, as incus, malleus and hyoid bone, some mesenchymal cells with bone developing potential may get trapped within projection of the tongue.[3] From the other point of view foramen

caecum is the region of thyroid gland' anlage development, remnants of which may be responsible for further cartilaginous or osseous proliferation.[26] Also, chondromatous proliferation at the projection of tongue midline could be explained by the incomplete involution of embryonic cartilaginous tissue of the lingual septum

In 1931 a case of bilateral osseous choristomas of the tongue was described, except which all other reports demonstrated unilateral lesion localization.[14,31] Osseous lingual choristomas have been considered as self-limiting in growth,[6] and frequently they are asymptomatic, while in some cases patients may complain on initiation of vomiting reflex, nausea, gagging sensation and possible dysphagia, manifestations of which could be depended on specific localization and size of the lesion.[29] Due to the systematic review more than 50% of patients with osseous choristomas of the tongue also demonstrated some associated symptomatology (gagging-68%, dysphagia-28.9%, swelling of the tongue-15%, pain-8,0%, changes of speaking function-5%).[6] Benamer et al. stated that lingual osseous choristoma in 25,8% cases clinically characterized just by the subjective feeling of lump on the tongue, while only 6,9% cases were associated with dysphagia, 5,1%-with gagging, 3,4%-with irritation, 3,4%-with nausea.[4]

During the choristoma localization at the base of tongue patients complain on lump and foreign body sensation. In study of Maqbool et al. lingual osseous choristoma caused upper airway obstruction, which was related with the size of the lesion (50 on 40 mm).[37] Analogical outcome of lingual glial choristoma in the form of respiratory distress was presented by Machi et al. among neonate.[38] Another clinical report described situation of severe manifested vomiting symptom caused by lingual osseous choristoma.[39] But in review of the literature provided by Supiyaphun et al., authors have not found associations between choristomas size and presented clinical symptoms.[5]

In all described cases of lingual osseous choristomas which also were supported by histological examinations, mature compact bone was identified within the lesions, while in 75% cases it was also covered with stratified squamous epithelium and in 38% cases accompanied with osteocytes haversian canals presence.[6] Even though individual osteoblasts and osteoclasts could be found during histological examination of lingual osseous choristomas, but no signs of bone modeling-remodeling activity within them were noted in literature yet.[31] Presence of bone marrow or hemopoietic tissues was registered rarely in cases of osseous choristomas of the tongue, because usually intertrabecular spaces are filled with fibrovascular tissue.[21] Sometimes cartilaginous choristomas of the tongue includes elements of bone, adipose tissues and other elements.[40]

Due to the Lindquist et al. only 5 cases of epidermal choristomas of oral cavity have been reported in the literature, while this type of choristomas demonstrated predominant prevalence among male patients in the form of hyperpigmented macule or plaque.[41] Described case of epidermal choristoma of the tongue pointed the attention to such lesion by its potential to get infected and obstruct upper airways.[41] Infected lingual osseous choristoma could cause prominent symptomatic, which in turn could be covered with the use of antibiotics and further excision of the lesion.[2]

Salivary glands choristoma also were found in larynx and bronchus, pituitary gland, parathyroid and thyroid glands, lymph nodes, mediastinum, prostate gland and rectum and even at the buccinator muscles.[42] Choristoma arising from the minor salivary gland was also registered at the area of soft palate.[10] Su Q.Y. et al. described a rare case of salivary gland choristoma in the middle ear.[43] Analogical findings were also reported by other authors.[44-47] Well-designed review of specifically middle ear salivary choristomas was provided by Young and colleagues.[48]

Previously it was suggested that choristomas located within tonsils could be the potential cause of chronic recurrent tonsillitis.[49,50] Several studies reported osseocartilaginous choristomas present within palatine tonsil,[7] while near 3% of chronic tonsillitis cases were associated with cartilaginous choristoma after histopathological examination of specimen received after tonsillectomy.[51]

Osseous choristoma of mandibular vestibule seems to be one of the most recurrent type of choristomas among those previously described in the literature.[52] Recurrence of osseous choristomas within oral cavity could be associated with the fact of iatrogenic trauma during their surgical incision, which causing developing of scar tissues. Latter is characterized with potential to ossification.[52] On the other hand, osseous choristoma could be represented not only by the main singular nodule, but such may be accompanied with smaller heterotopic satellite islands of cells. Remaining of latter may cause further recurrence of pathologic lesion. But in the case presented by Alshawaf and Zahrani osseous choristoma involving the mandibular buccal vestibule has not presented any sign of recurrence for 2 years monitoring.[53]

Few cases of so-called odontogenic choristomas have been reported previously. Such pathologies include presence of well-developed tooth structures with corresponding bone support but at non-tooth-bearing areas.[54]

Case of osseous labial choristoma was described by Bastian et al.[55] While presenting another case of osseous choristoma specifically at the upper lip, Veni and coauthors also stated that such lesion localization is firstly described, and previously 72 cases of osseous choristomas were found in tongue, 15 in mucous membrane of cheek and 1 in lower lip.[56]

Osseous choristoma of the periodontium at the projection of second mandibular premolar have been firstly described in 2012, while itself being asymptomatic but causing discomfort during mastification.[57] During such localization authors recommended to differentiate such mass with heterotopic ossification, peripheral ossifying fibroma and exostosis. Later in 2017 Karci and colleagues also reported periodontal osseous choristomas in the region of teeth 31-41, which potentially could be caused by excessive orthodontic forces provoking traumatization of periodontium.[58] Cartilaginous choristoma of the gingiva was also reported in the literature, which was clinically accompanied with the sinus tract.[59]

Several times osseous choristomas also were found within the soft tissues of head and neck region. Arens et al. systematized only 6 available previous cases of osseous choristomas within cervical soft tissues predominantly at submandibular region, while also describing case with choristoma localization at lateral cervical triangle.[60]

Ozaki and colleagues described first case of multiple osseous choristomas on the medial side of the lateral pterygoid muscle, which caused temporo-mandibular joint disorders.[61]

IDENTIFICATION, EXAMINATION AND TREATMENT MODALITIES OF ORAL CAVITY CHORISTOMAS

Identification and examination of oral cavity choristomas may include periapical roentgenological methods, computed tomography, cone beam computed tomography, MRI, ultrasonography. Yoshimura et al. described possibility of using dermoscopy method for lingual choristoma evaluation.[25] Dermocsopic characteristics of the excised lesion sample included hypovascular and homogeneous pattern accompanied with round extruded white substance.[25] Markedly low signal-intensity on both T1and T2-weighted MRI scans in conjunction with clinical signs and anamnesis data could help to presume possible diagnosis of lingual osteoma.[62] Specifics of osseous choristomas obtained during the analysis of CBCT findings was described by Sinha et al., who have highlighted their corticated margins, mixed density content, no contact with adjacent bony structures.[63] Pereira et al. also highlighted potential role of immunohistochemical analysis for differential diagnosing of cartilaginous choristoma especially under condition without usage of imaging methods.[27]

Choristomas should be differentiated from hamartomas, different kind of metaplasia, chondromas, papilloma, myxoid tumors, pleomorphic adenomas, granular cell tumors, malignant lesions of oral soft tissues. Osseous buccal choristomas should be differentiated from peripheral ossifying fibroma, myositis ossificans, heterotopic ossification, calcifications related with dystrophy process, extraskeletal osteochondroma.[64]

Usual treatment modalities of oral cavity choristomas include excision with laser or scalpel. But Hemmi et al. also described a case, where osseous choristoma of the tongue was spontaneously removed during patients coughing.[29] Literature does not provide enough evidences about possible recurrence of choristomas after their full excision. Due to the data collected within systematic review osseous lingual choristomas had not demonstrated any tendency for recurrence, while osseous choristomas of other localization within oral cavity in some reported cases were recurrent.[6] Cartilaginous choristomas of oral cavity may demonstrate tendency of size increase, which was documented by the histological sings of interstitial and/or appositional growth.[19] In the publication of Sotorra-Figuerola et al. authors mentioned that because of immature choristomas' nature, risk of their possible malignization is theoretically possible.[42]

CONCLUSION

Choristomas of oral cavity and such of oral cavity origin, but located within other organs and systems, characterized with low level prevalence, nevertheless, risk of such lesions development should always be considered during the process of differential diagnosis. Deficiency of systematized data regarding oral cavity choristomas limiting possibilities for prognosis regarding their recurrence or potential malignization, even though only individual clinical reports demonstrated choristomas relapses after full excision and their potential for getting infected or irritated. Considering that etiology of choristomas remains under on-going investigation, descriptive representation of such pathology's diagnosis with mentioning the specifics of covering tissue, presence of some structures within main cells aggregation and characteristics of content and localization of such, would be supportive for further evidence-base expansion regarding above-mentioned tumor-like masses.

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