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CASE REPORT

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A torus-like sublingual adenoid cystic carcinoma in a 35-year-old male: Review of literature and case report

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Key Clinical Message

The case report describes an adenoid cystic carcinoma occurring at a rare location and the diagnostic pathway. Swellings of the floor of the mouth, whether painful or without subjective symptoms and regardless of consistency, should be taken seriously.

Abstract

Adenoid cystic carcinoma (ADCC) of the sublingual salivary gland only accounts for 2% of all ADCCs. In this study, we report a rare case of ADCC with sublingual salivary gland origin in a 35-year-old man and a comprehensive review of articles published over the past 62 years.

KEYWORDS

adenoid Cystic Carcinoma, major Salivary Gland, oral, sublingual Saliva Gland

1 | INTRODUCTION

Malignancies of the sublingual glands are exceedingly uncommon and only account for 0.4%–0.6% of total salivary gland malignancies.[1,2] In general, among the major salivary glands, the smaller the gland itself, the more aggressive its tumor is likely to be and to have a worse prognosis.[3] Even though the most common tumors of the sublingual salivary gland are Adenoid Cystic Carcinoma (ADCC) and mucoepidermoid carcinoma (MEC), they only account for 2% and 1.6% of all the ADCCs and MECs, respectively.[1]

The cylindrical appearance of ADCC was first described by Robin, Lorain, and Laboulbene in two studies in 1853 and 1854. Afterward, Billroth was the first to choose the name "cylindroma" for ADCC lesions in 1856. This was due to the cribriform appearance formed by tumor cells with cylindrical pseudolumina or pseudospaces in the histological appearance of the lesion. ADCC was then used to define the tumor.[4,5]

ADCC is a quite rare epithelial tumor of major and minor salivary glands. ADCC accounts for only 1% of oral and maxillofacial malignancies.[6] Out of all ADCC cases, 15%–30% occur in the submandibular gland, 30% in the minor salivary glands, 2%–15% in the parotid gland, and 2% in the sublingual gland.[7] In terms of position, ADCC is located most frequently in the palate, followed by the tongue, buccal mucosa, lips, and floor of the mouth.[8] ADCC is more likely to occur in women and the fifth and sixth decades of life.[6,8]

Clinicopathologically, ADCC is a slow-growing swelling with perineural invasion, local recurrence, and distant metastasis.[9,10] Nerve involvement that leads to pain is a predominant feature of ADCC.[11] Most metastases occur through the lymph other than blood.[12] Distant metastasis occurs most commonly in the lungs, bones, liver, and brain, respectively.[13] The overall 5-year survival rate of patients with head and neck ADCC is 90.3%.[10]

Histopathologically, cribriform, solid, and tubular are the three patterns of ADCC. More than one pattern

is usually seen in the majority of tumors.[14] According to Szanto et al., the cribriform or tubular pattern is considered Grade I ADCC. If the solid pattern is less than 30%, it is considered Grade II, and if it is above 30%, it is regarded as Grade III.[15] Although the definitive diagnosis of ADCC is made based on histopathological assessments, radiographic modalities such as CT or MRI can help diagnose the extension and invasion of the lesion, along with its recurrence and metastasis.[16,17] The lesion is typically irregular in shape, containing bone fragments that can be seen by CT and MRI.[10] Among the treatment options available for ADCC, including radical surgery, radiotherapy, chemotherapy, and concurrent chemotherapy with radiotherapy, the most common modality is radical surgery in combination with adjuvant radiotherapy.[9,10,12,18] The effect of adjuvant radiotherapy on the survival rate of patients with ADCC is highly debated.[11] It is now clear that ADCC is still a very complex disease to treat. No single treatment can meet all the needs of ADCC management.[19]

In this study, we report a case of ADCC in a 35-year-old Afghan man occurring at floor of the mouth, the diagnostic pathway to it, and a comprehensive review of articles on sublingual ADCCs published over the past 62 years.

2 | CASE HISTORY/ EXAMINATION

A 35-year-old Afghan man with the chief complaint of swelling and stiffness in the floor of the mouth from a year ago, which had grown faster in the past 6 months and was recently felt while eating, was referred to the Department of Oral and Maxillofacial Medicine. No history of ulcer or pain was reported at the site. The patient did not have any systemic diseases or take any medications. He also had no familial history of cancer.

On extraoral examination, no significant findings indicating the disease were observed. On intraoral examination, an exophytic, nodular, sessile, and pink mass with superficial telangiectasia was observed. This lobular mass was located in the lingual aspect of the mandible and extended anteroposteriorly from 35 to 46 on the opposite side. Its surface was intact and the consistency was bony. (Figure 1).

3 | DIFFERENTIAL DIAGNOSIS, INVESTIGATIONS AND TREATMENT

Based on the location of the lesion, its consistency, and clinical appearance, the provisional diagnosis was torus mandibularis. However, according to the patient's confidence that the lesion had grown faster in the last 6 months and there had been no lesions a year earlier; we decided to investigate the lesion in detail. Therefore, a panoramic image and an occlusal radiograph were taken. The occlusal graph showed a small opacity that could not explain the large size of the torus mandibularis. (Figure 2).

Therefore, we decided to prepare a cone beam computed tomography (CBCT) image from the anterior of the mandible. (Figure 3) The CBCT cross-sectional and axial images with 2 and 3 mm slice intervals revealed a peripheral erosive lesion from the midline to the mesial region of the right first molar with an anteroposterior dimension of 28 mm. The lesion had a well-defined ragged border from the periapex area of the teeth to the inferior border of the

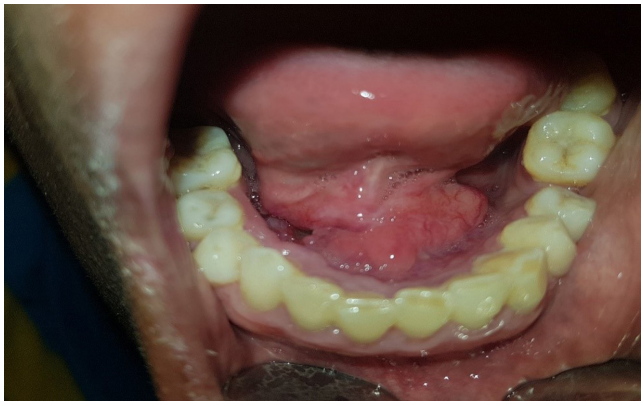


FIGURE 1 Intraoral photograph showing a lobular, sessile swelling in the floor of the mouth.

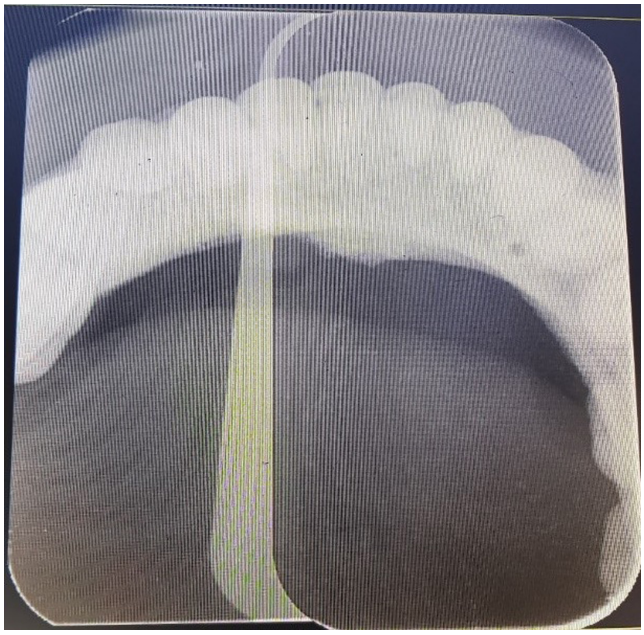


FIGURE 2 Mandibular occlusal radiograph shows only a small opacity that is not large enough to explain the torus mandibularis.

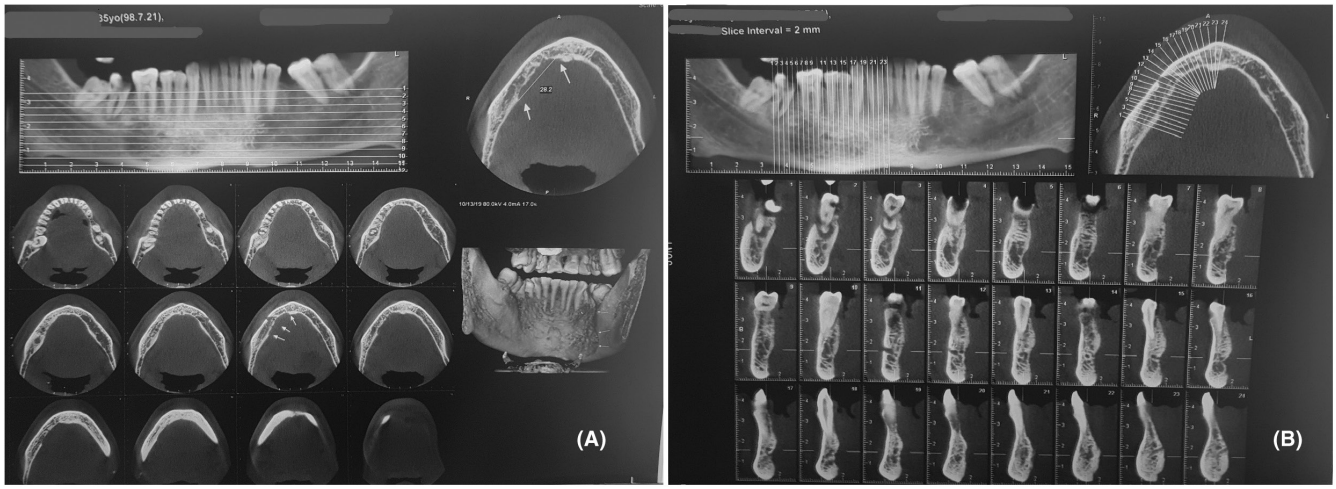


FIGURE 3 Cone-beam computed tomography images of the sublingual mass level. (A) Axial images with 2-3 mm slice intervals. (B) Cross-section images with 2 mm slice intervals. An erosive lesion with well-defined ragged border was seen.

mandible, and saucerization resorption was detected. No effect on IAC and teeth was seen.

Therefore, differential diagnoses of malignant tumors of the right sublingual glands and Rhabdomyosarcoma were made. Conclusively, the patient was referred to an oral and maxillofacial surgeon for an incisional biopsy.

Microscopic examinations showed a malignant neoplasm composed of nests of myoepithelial and ductal cells arranged in tubular patterns in most areas. Solid nests and also linear cords of single files were evident. The tumor cells mostly showed hyperchromatic basaloid nuclei, and the nuclei were pale-staining and vesicular in some foci. Numerous mitotic figures were also seen. The stroma was more mucoid in nature and also demonstrated hyalinization in some foci. The lesion was covered with parakeratinized atrophic stratified squamous epithelium. IHC study showed; P63: positive in peripheral cells of the islands and ducts, C-KIT: positive in most tumoral cells, and Ki67: positive in 30% of the tumoral cells in hot spots. (Figure 4).

The initial diagnosis was ADCC. Based on immunohistochemistry, the final diagnosis was ADCC, Grade II (tubular pattern with 5% solid component).

According to the final diagnosis, the surgeon requested a spiral computed tomography (CT) scan with IV contrast from the peripheral nervous system, neck, and chest.

At least 18 nodular opacities with a maximum size of 16 mm were seen in both lungs, which were in favor of pulmonary metastasis.

Based on these data, a CNB (core needle biopsy) with a CT guide was performed on the mass in the apex of the right lung, which showed adenoid cystic carcinoma.

Due to the spread of the disease and distant metastasis to the lung, the patient needed excisional surgery of the sublingual gland together with chemotherapy.

4 | OUTCOME AND FOLLOW-UP

The patient preferred to continue the treatment process in Pakistan and unfortunately, was lost to follow-up.

It should be noted that informed consent was obtained from the patient for publication.

5 | DISCUSSION

ADCC is a rare malignancy, and approximately 10% of ADCCs occur in the salivary glands.[20,21] In a study by Spiro et al., out of 2477 patients suffering from major salivary gland tumors or tumors of the mucous glands of the upper aerodigestive tract at the Memorial Cancer Center over 25 years, ADCC was diagnosed in only 242 of the patients, which accounted for only 10% of them. Among the 242 cases, 171 had minor salivary gland origin (71%). In the other 68 patients (28%), the malignancy involved parotid or submandibular glands, whereas in only three cases (1%) the origin of the tumor was the sublingual gland.[22]

Numerous case studies and review articles have been published on ADCC. In this study, we decided to review articles from 1960 to 2022. An electronic search was done on PubMed, Google Scholar, Scopus, Science Direct, and Web of Science databases using the keywords:

("Adenoid Cystic Carcinoma" OR "Intraoral ADCC" OR "Intraoral Adenoid Cystic Carcinoma" OR "oral ADCC") AND ("Major Salivary Gland" OR "Sublingual Salivary Gland")

Exclusion criteria included microscopic and molecular studies that did not provide enough information about each case (demographic factors of each case such as age and sex and detailed case presentation mentioning consistency, symptoms, etc.); or were not available. Inclusion

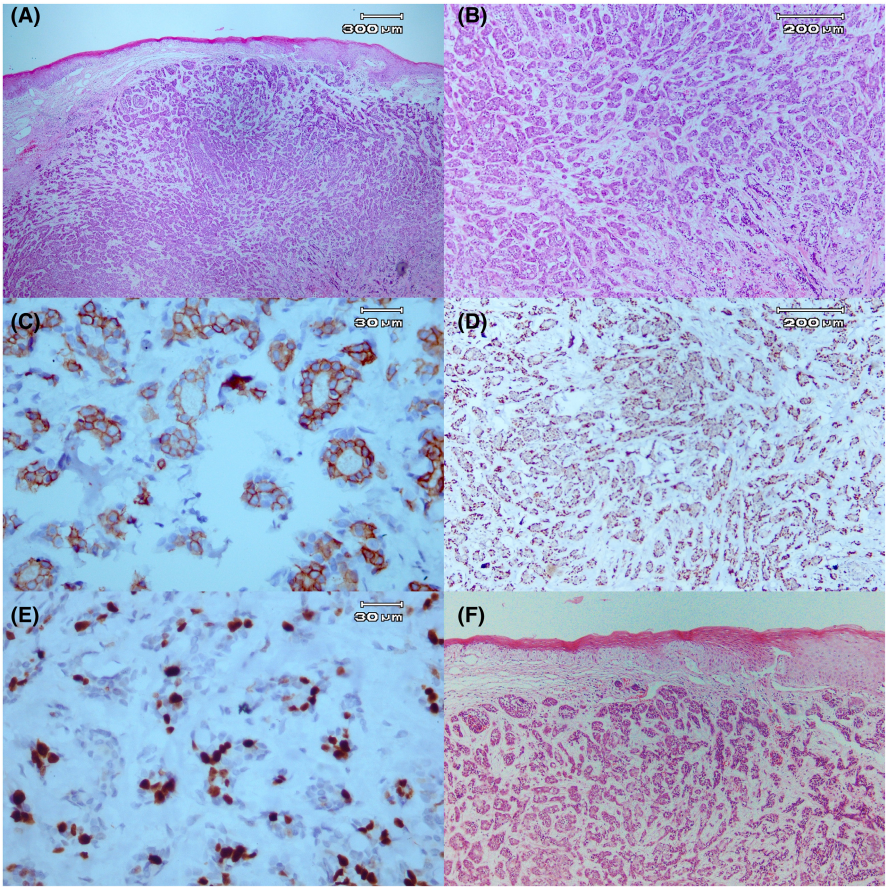


FIGURE 4 Histopathological findings of adenoid cystic carcinoma. (A, B) A biphasic tumor composed of ductal and myoepithelial cells arranged in tubular architecture; beneath the oral mucosa (H&E, ×40) (C) ductal structures highlighted by C-kit (CD117) immunostaining (×100) (D) myoepithelial cells highlighted by p63 immunostaining (×40) (E) strong reactivity for Ki67 in hotspot areas (×100) (F) multiple duct-to-tubule-like structures lined by several layers consisting of ductal or myoepithelial cells or both (H&E, ×40).

criteria were available English case reports, case series, research articles, and review articles published on sublingual salivary gland ADCCs from 1960 to 2022; that contained detailed demographic factors and case presentations case by case.

The results are as follows:

Identification: Altogether 97 articles out of 5 databases were collected. After duplicate records were excluded, 63 papers remained for screening. ($N=63$).

Screening: Based on the exclusion criteria, six articles were removed. One article was Chinese and five others were not available. ($N=57$).

Eligibility: The full text of these 57 articles was reviewed and one article was removed due to irrelevance because it was about a case with polymorphous low-grade adenocarcinomas (PLGA) and 29 others did not provide enough information about each case (mentioning detailed demographic factors and case presentation of each case). ($N=27$).

Included: Altogether only 27 of them were appropriate based on the inclusion and exclusion criteria (Figure 5).

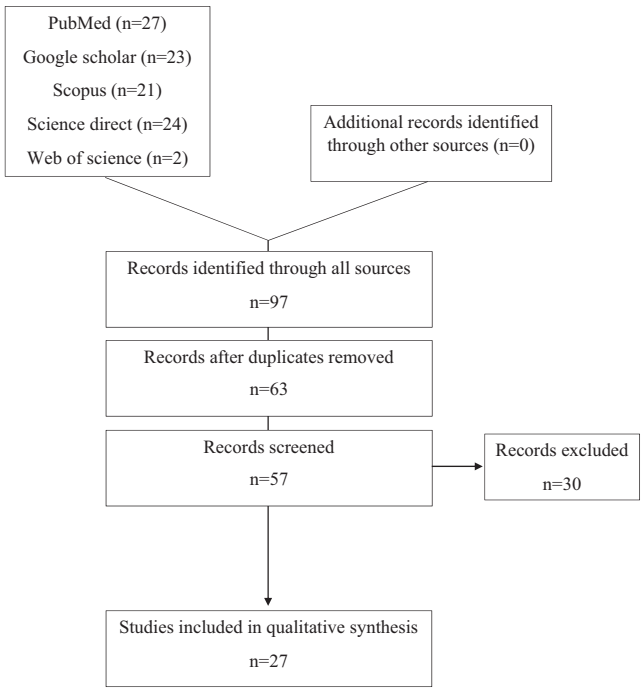


FIGURE 5 Total number of articles in terms of search engine (total $N=63$)

TABLE 1 Characteristic of patients with sublingual gland adenoid cystic carcinoma.

Author, Ref.	Sex	Age, years	Consistency	Symptoms	Metastasis
Ohta et al.[23]	M	≥40	Elastic hard	Pain	+ (Lung/skin/vertebrae)
Morita et al.[24]	M	≥40	NM	None	+ (Lung/eye)
Park et al.[25]	F	≥40	NM	None	–
	F	≥40	NM	None	–
	F	≥40	NM	None	–
	F	≥40	NM	Taste change	+ (NM)
	F	≥40	NM	None	–
	M	≥40	NM	None	–
	M	≥40	NM	None	–
	M	≥40	NM	Numbness	+ (NM)
	M	≥40	NM	None	+ (NM)
	F	≥40	NM	None	–
	F	≥40	NM	Pain	–
	M	≥40	NM	Numbness	–
	F	≥40	NM	None	–
	F	≥40	NM	None	+ (NM)
Gontarz et al.[26]	F	<40	NM	Pain	+ (Lung)
	M	≥40	NM	None	–
	F	≥40	NM	None	–
Li et al.[27]	F	≥40	NM	None	+ (Liver)
Kojima et al.[28]	F	≥40	NM	NM	+ (Lung)
	M	≥40	NM	NM	–
	F	≥40	NM	NM	+ (Lung)
	F	≥40	NM	NM	+ (Lung)
	M	≥40	NM	NM	+ (Lung)
	M	≥40	NM	NM	+ (Lung)
	F	≥40	NM	NM	+ (Lung/Bone)
	M	≥40	NM	NM	NM
Sepulveda et al.[29]	F	≥40	Solid	Pain	–
Fujita et al[30]	M	≥40	Firm	Tingling pain	+ (Bone)
Song et al.[31]	M	≥40	Fibrotic	Slight and dull pain	–
Huang 2016[32]	M	≥40	NM	NM	–
	F	≥40	NM	Pain/Numbness	+ (Lung)
	M	≥40	NM	Pain/Numbness	+ (Lung)
	F	≥40	NM	NM	–
	F	<40	NM	NM	–
	M	≥40	NM	Pain	+ (Lung/Bone)
Acharya et al.[33]	F	≥40	Firm	Throbbing pain–burning sensation	NM
Yoshioka et al.[34]	M	≥40	Elastic firm	None	+ (Lung)
	F	≥40	Firm	None	+ (Lung)
Kumar et al.[35]	M	≥40	Soft	Tenderness	NM

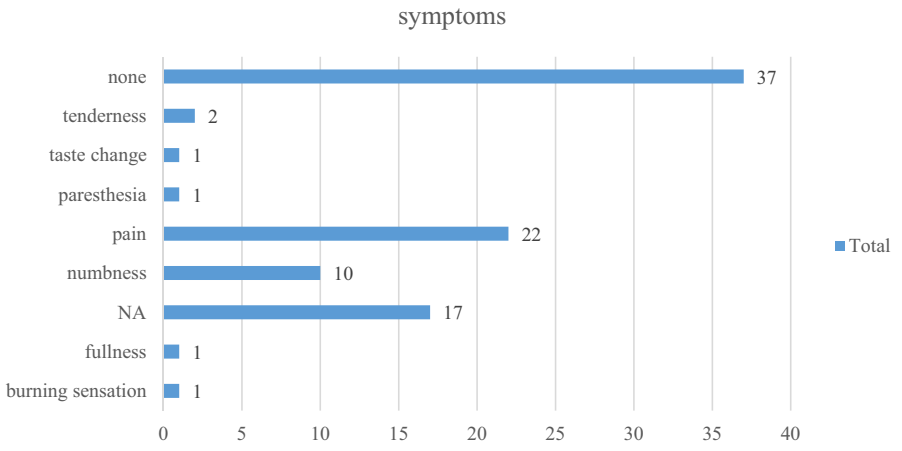
(Continues)

TABLE 1 (Continued)

Author, Ref.	Sex	Age, years	Consistency	Symptoms	Metastasis
Shimamoto et al.[36]	F	≥40	NM	Paresthesia	–
	F	≥40	NM	None	–
	F	≥40	NM	None	–
Adirajaiah et al.[37]	F	≥40	Firm	Pain	NM
Sun et al.[38]	F	≥40	NM	Pain/Numbness	+ (Lung)
	F	≥40	NM	Pain/Numbness	+ (Lung/brain)
	M	≥40	NM	None	–
	F	<40	NM	None	+ (Lung/chest wall)
	F	≥40	NM	None	–
	F	<40	NM	None	–
	F	≥40	NM	None	–
	F	≥40	NM	Pain/Numbness	–
	F	≥40	NM	None	–
	M	<40	NM	Pain/Numbness	–
	F	≥40	NM	Pain/Numbness	+ (vertebrae)
	M	≥40	NM	None	–
	M	≥40	NM	None	–
	F	<40	NM	None	+ (Lung)
	F	≥40	NM	Pain	–
	F	<40	NM	None	–
	M	<40	NM	Pain/Numbness	–
	M	≥40	NM	Pain	–
Papadogeorgakis et al.[39]	F	≥40	Firm	None	–
Saito et al.[2]	M	≥40	Elastic hard	None	NM
Perez et al.[1]	F	≥40	NM	None	NM
	M	≥40	NM	None	+ (Lung)
	F	≥40	NM	None	–
	F	≥40	NM	Pain	+ (Lung)
Shigematsu et al.[40]	F	≥40	NM	None	–
Ogawa et al.[41]	M	≥40	Elastic	None	NM
Avery et al.[42]	NM	NM	NM	NM	–
	NM	NM	NM	NM	–
	NM	NM	NM	NM	+ (Lung)
Sakashita et al.[43]	M	≥40	Elastic hard	Pain	–
McFall et al.[44]	F	<40	Firm	Fullness & tenderness	–
Califano et al.[45]	M	≥40	Elastic	None	NM
Whear & Addy[46]	F	≥40	Hard	Pain	–
Rankow & Mignogna[47]	F	≥40	NM	NM	NM
	M	≥40	Firm	None	+ (Lymph)
	M	≥40	Bony-hard	None	–
	M	≥40	NM	NM	NM
	F	≥40	Firm	NM	–

Abbreviations: NM, not mentioned; +, presenting metastasis; –, not presenting metastasis.

GRAPH 1 The prevalence of symptoms in ADCC patients.



The results of the review are summarized in Table 1 and are as follows:

Patients were divided into two age groups: 40 years and older (≥40) and under 40 years (<40). Only 9 out of 83 were under 40 years old (seven cases were female and two cases were male). In three cases, the sex of the patient was not determined, and from the rest, 32 cases were male and 48 cases were female. These are consistent with the findings of Sepulveda I et al.'s study, illustrating that ADCC often occurs between the fifth and sixth decades of life and mainly in women.[10] Despite that, in our reported case, the patient was male and under 40 years of age (35 years old), which is uncommon in terms of both gender and age according to the review.

In clinical examinations of 83 patients, bony-hard consistency was reported in only one case; however, our case also had a bony consistency that led to a misdiagnosis of exostosis (torus mandibularis). Furthermore, our patient was the first to have a lesion large enough to cross the midline. That is why at first the diagnosis was torus mandibularis.

In 29 cases, metastases were reported to be prevalent in the lung (21 cases), bone (three cases), lymph nodes and vertebrae (each in 2 cases), and liver, skin, brain, eye, and chest wall (each in 1 case). Out of the 29 patients with metastases in this review, only three were under 40 years old and 12 were male. Our case also involved a 35-year-old man with lung metastases.

Out of 83 patients, 28 had symptoms such as dull pain, throbbing and tingling, tenderness, burning sensation, fullness, paraesthesia, taste change, and numbness. Nevertheless, our patient did not have any of these symptoms. Accordingly, most patients did not have subjective symptoms. It may be concluded that ADCCs of the sublingual gland, unlike ADCCs of the other major salivary glands, are often asymptomatic. Hence, this feature may lead to late referrals, resulting in cases with a worse prognosis of this tumor in the smaller glands such as the sublingual gland. This result is consistent with the findings of Zdanowski et al. They indicated that ADCC of sublingual glands, was frequently diagnosed in advanced clinical stages as a consequence of lack of symptoms. In other words, most patients are diagnosed (about 83%) at stage III and stage IV.[48] (Graph 1).

In this study, we reported a case of an ADCC tumor in a 35-year-old Afghan man. This case has all the rare features simultaneously, including the incidence of the malignancy in a sublingual salivary gland of a young male patient, having bony-hard consistency with no subjective symptoms, being bilateral, and crossing the midline with asymptomatic pulmonary metastases. None of the cases reported so far had all these characteristics altogether, so it can be argued that this case is the first case reported in the world with the above features. Therefore, it can be concluded that all swellings of the floor of the mouth, whether painful or without subjective symptoms and regardless of consistency, should be taken seriously. The disease may have been so progressive that metastases have occurred.

AUTHOR CONTRIBUTIONS

Farzaneh Agha-Hosseini: Conceptualization; supervision; writing – review and editing. **Parnian Esmaili:** Formal analysis; project administration; writing – original draft. **Marzieh Yousefian:** Conceptualization; supervision; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

Authors declare that there is no conflict of interest.

DATA AVAILABILITY STATEMENT

All data generated or analyzed during this study are included in this published article and its supplementary information files.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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