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SINE QUA NON CLINICOPATHOLOGIC CORRELAT

Oral Adult Rhabdomyoma

Augusto César Leal da Silva Leonel[1] · Stefanny Torres dos Santos[1] · Elaine Judite de Amorim Carvalho[1] · Jurema Freire Lisboa de Castro[1] · Oslei Paes de Almeida[2] · Danyel Elias da Cruz Perez[1,2,3]

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Abstract
This report describes a case of an adult rhabdomyoma (ARM) occurring in the oral cavity. A 47-year-old man was referred for the diagnosis of a painless, well-circumscribed, submucous nodule located on the floor of the mouth, measuring approximately 6.0 cm in length. Computed tomography revealed a well-defined, solid, and hypodense mass. A benign salivary gland or mesenchymal tumor were the main diagnostic hypotheses. Under local anesthesia, the patient underwent surgical excision. Microscopically, the tumor comprised large polygonal well-defined cells with abundant, eosinophilic granular cytoplasm with cross striations. No atypia or mitosis was observed. The cells were positive for muscle-specific actin, desmin, and sarcomeric alpha-actin. Based on these features, a diagnosis of ARM was established. No recurrence was observed after 48 months. Although rare, ARM should be considered in the differential diagnosis of oral submucosal nodules, especially those located on the floor of the mouth.

Keywords Benign neoplasms · Diagnosis · Differential · Immunohistochemistry · Mouth · Rhabdomyoma · Therapeutics

History and Clinical Findings

A 47-year-old man was referred for the evaluation of an oral, painless, slow-growing mass with approximately 2 years of evolution. The patient complained of dysphagia and discomfort during speech. Intraoral examination revealed a large, painless, well-circumscribed, and non-tender mass located on the right floor of the mouth, measuring approximately 6.0 cm in length (Fig. 1). The lesion was recovered by smooth, intact, and normal-colored mucosa. The tongue could not protrude normally due to the superior and posterior displacement of the lesion. Axial section computerized tomography images showed a well-delimited, solid, hypodense mass located on the floor of the mouth/submandibular region. The main hypotheses for diagnosis were a benign salivary gland tumor as a pleomorphic adenoma and a benign mesenchymal tumor. As the lesion was well delimited, it was surgically excised under local anesthesia.

Diagnosis and Treatment

Gross examination revealed a bilobulated nodule with an irregular surface, brown coloring, and fibrous consistency, measuring 6.9 × 3.2 × 2.0 cm. Microscopically, at low magnification, the lesion appeared as a well-circumscribed and partially encapsulated nodule with irregular margins (Fig. 2A). The tumor comprised large polygonal cells with well-defined borders, with central or eccentric nuclei and prominent nucleoli (Fig. 2B–D). The cells exhibited abundant, eosinophilic, and granular cytoplasm, which often presented complete or partial vacuolization; so-called "spider cells" (Fig. 2B–D). Cross-striations in the cytoplasm were also observed, and there were no atypia or mitoses (Fig. 2D). In addition, hypocellular areas formed by fibrous connective tissue were also observed. Immunohistochemically, the tumor cells were positive for muscle-specific actin (HHF-35,

✉ Danyel Elias da Cruz Perez
danyel.perez@ufpe.br

[1] Department of Clinical and Preventive Dentistry, School of Dentistry, Universidade Federal de Pernambuco (UFPE), 4ª Travessa Professor Artur de Sá, s/n, Cidade Universitária, Recife, Pernambuco CEP 50740-521, Brazil

[2] Oral Diagnosis Department, Piracicaba Dental School, University of Campinas (UNICAMP), Av. Limeira, 901. Areão, Piracicaba, São Paulo CEP 13414-903, Brazil

[3] Departamento de Clínica e Odontologia Preventiva, Universidade Federal de Pernambuco, 4ª Travessa Professor Artur de Sá, s/n, Cidade Universitária, Recife, Pernambuco CEP 50740-521, Brazil

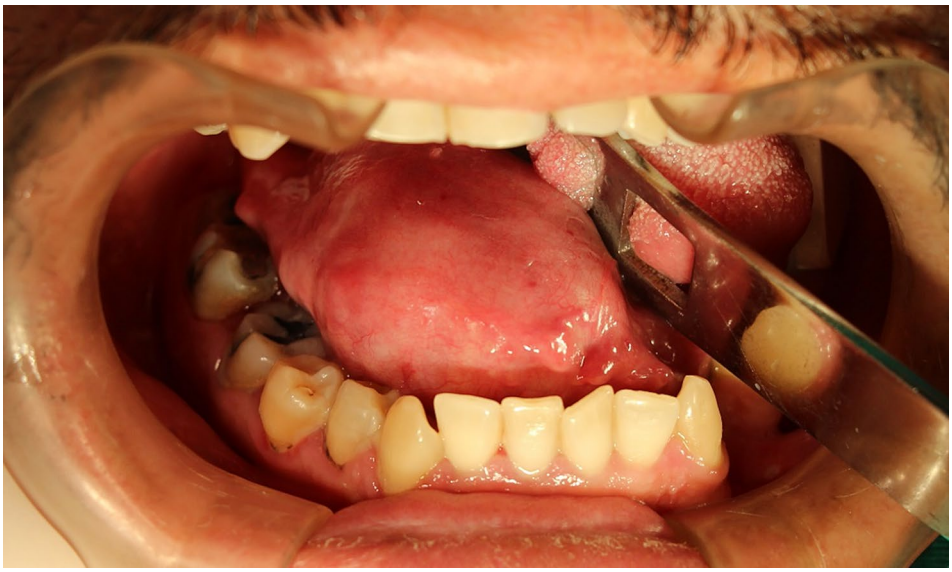

Fig. 1 Large, painless, well-circumscribed submucous nodule located at the right side of the floor of the mouth. The lesion was covered by smooth, intact, and normal-color mucosa

1:800; Dako, Glostrup, Denmark), desmin (D33, 1:800, Dako) (Fig. 3A), and sarcomeric alpha-actin (EP2529Y, 1:300, Abcam, Cambridge, England) (Fig. 3B). In contrast, the cells were negative for calponin (CALP, 1:600, Dako), H-caldesmon (h-CD, 1:400, Dako), and myogenin (F5D, 1:500, Dako). Thus, a diagnosis of ARM was established.

The patient received clinical and imaging follow-up, with no signs of recurrence 48 months after treatment.

Discussion

Rhabdomyoma (RM) is an uncommon benign mesenchymal tumor that accounts for approximately 2% of all striated muscle tumors. It is usually classified into cardiac and extracardiac forms [1, 2]. Cardiac RM occurs more frequently in young children and is often associated with tuberous sclerosis, neurofibromatosis, and sebaceous adenomas [1, 3, 4]. Extracardiac RMs are rare and classified into adult and fetal types, depending on the degree of differentiation. RM also occurs as a genital type located in the vagina and vulva [1, 5].

Adult rhabdomyoma (ARM) is a rare benign neoplasm with differentiation of mature skeletal muscle. ARM is often solitary (70%) but may be multinodular (26%), and rarely multicentric (4%) [1, 3, 6, 7]. ARM occurs most commonly in men over 50 years of age, and shows a predilection for the head and neck region, followed by the extremities, esophagus, stomach, mediastinum, orbit, prostate, and intracranial regions. ARM is rare in the oral cavity and, to date, about 80 cases, including the present one, have been reported in the English-language literature (Table 1) [1–61]. The fetal type also typically involves the head and neck region, tends to occur at younger ages, and rarely affects adults, whereas the genital type is found in the vagina and vulva of young and middle-aged women [6, 8]. The present case fulfilled the ARM criteria.

According to previously published cases, oral ARM more commonly affects men (75%), with a male:female ratio of 3:1, and a mean age of 57 years, with a peak incidence between the sixth and seventh decades of life [1–9]. Most cases occur on the floor of the mouth/submandibular region (49–61.2%), followed by the tongue base (16–20%), soft palate (9–11.2%), buccal mucosa (3–3.8%), and lip (3–3.8%) [1–61]. Some patients (14 cases, 17.5%) present with multifocal lesions [6, 10, 22, 24, 25, 30, 36, 37, 47, 49, 51, 52, 57, 60]. The lesions vary in size from a few millimeters to 15 cm [1–61]. Similar features were observed in the present case.

Clinically, oral ARM usually presents as a painless, slow-growing, well-circumscribed, homogeneous, non-tender, and mobile submucosal nodule. However, depending on the size and site of occurrence, such as the floor of the mouth, displacement of the tongue, dysphagia, dyspnea and/or apnea, and facial asymmetry may occur [1–61]. In our case, the patient presented with dysphagia and difficulty during speech. The mean time of evolution is 16 months, ranging from 2 to 60 months [1–61]. The clinical differential diagnosis depends on the location and may include benign and malignant salivary gland tumors and benign mesenchymal tumors, as considered in the present case.

Histologically, ARM is characterized by polygonal cells of variable size with abundant eosinophilic and granular cytoplasm. Cross-striations are typically readily identifiable. Mitoses and necrosis are absent. Immunohistochemical

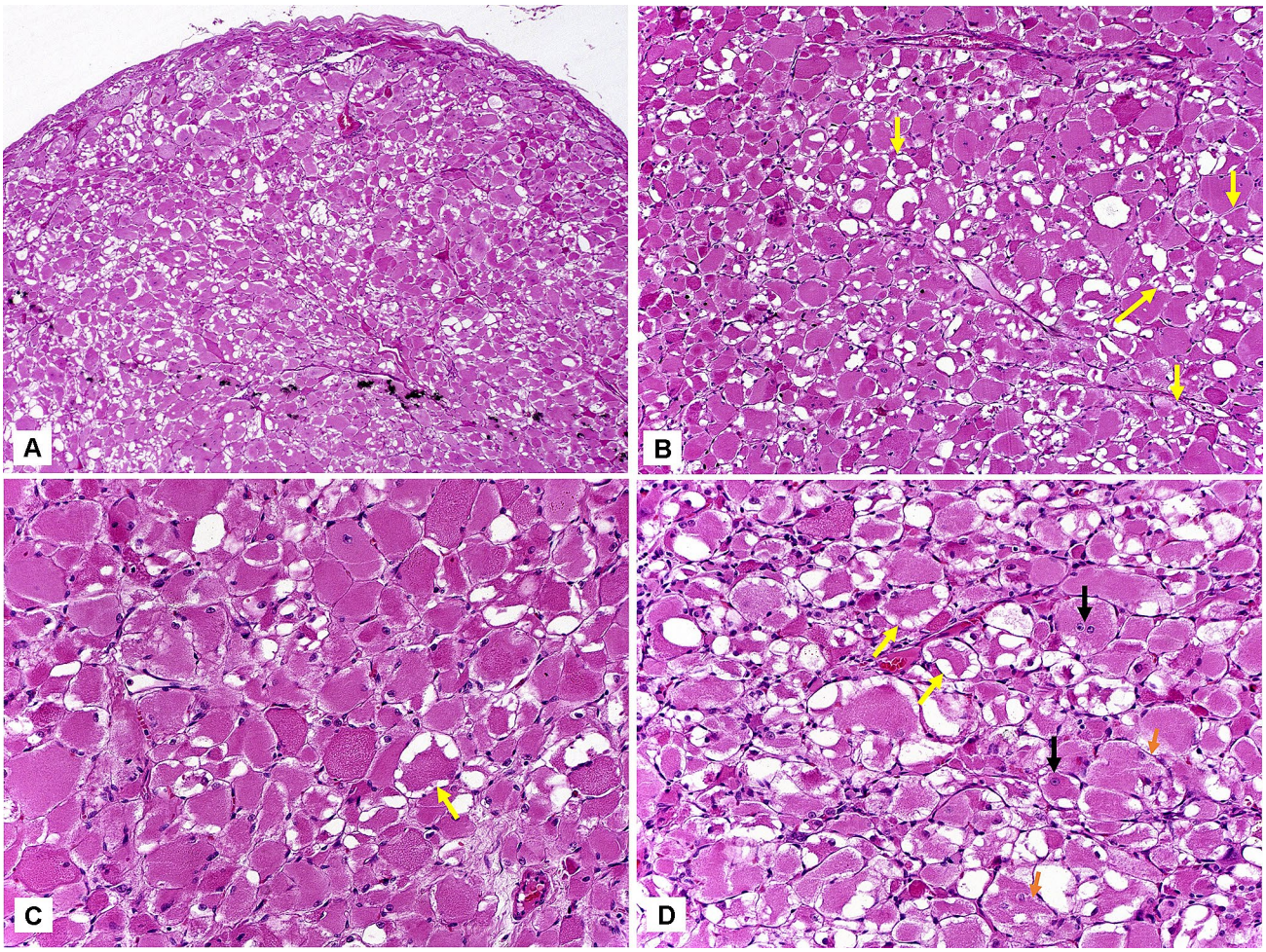

Fig. 2 **A** Microscopically, at low magnification, the lesion appeared as a well-circumscribed and partially encapsulated nodule, with irregular margins (hematoxylin-eosin, 50x). **B** Large polygonal cells exhibited abundant, eosinophilic, and granular cytoplasm, which often presented complete or partial vacuolization; so-called "spider cells" (yellow arrows) (hematoxylin-eosin, 100x). **C** Large tumor cells with well-defined borders, and central or eccentric nuclei. Cytoplasmic vacuolization is also observed ("spider cells"—yellow arrows) (hematoxylin-eosin, 200x). **D** Most cells exhibiting granular cytoplasm, with complete or partial vacuolization ("spider cells"—yellow arrows). Prominent nucleoli (black arrows) and cross-striations (orange arrows) were also observed (hematoxylin-eosin, 200x)

reactions confirm skeletal muscle differentiation [3, 10]. Histopathological differential diagnoses include lesions consisting of cells with abundant eosinophilic cytoplasm, such as a granular cell tumor, hibernoma, and oncocytoma [11]. However, none of these tumors present with cross-striations or glycogen. In addition, paraganglioma and crystal storage histiocytosis should also be considered [1, 14].

Granular cell tumors are characterized by cells showing large and eosinophilic cells, with finely granular cytoplasm devoid of cross-striations and defined borders characteristics of ARM cells. Tumor cells are strongly positive for S-100 protein, and skeletal muscle markers are typically absent [1, 6]. Hibernomas consist of cells containing diverse intracytoplasmic lipid droplets tending to show centrally placed nuclei, while ARM cells have predominantly peripherally located nuclei. Immunohistochemically, cells display strong cytoplasmic positivity for S-100 [14]. Oncocytoma is a benign salivary gland neoplasm comprising polyhedral cells rich in mitochondria that show finely granular and eosinophilic cytoplasm. Tumor cells are positive for epithelial markers, whereas muscular

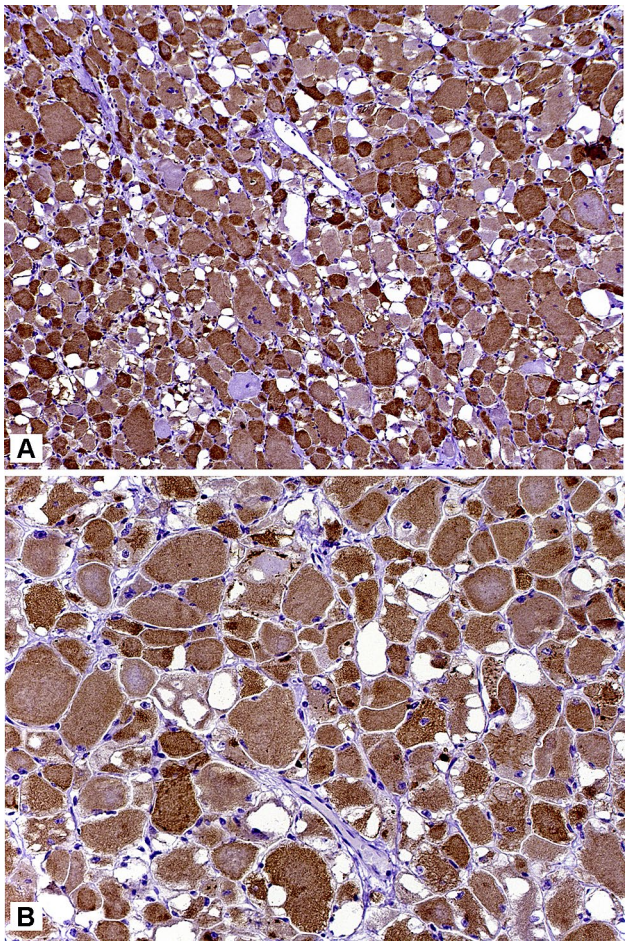

Fig. 3 **A** Tumor cells presenting strong positivity for desmin (streptavidin-biotin-peroxidase method, 100x). **B** Tumor cells exhibiting strong positivity for sarcomeric alpha actin. Cross-striations are evident (streptavidin-biotin-peroxidase method, 400x)

markers are negative [2, 3]. Rhabdomyosarcomas may also be considered but are composed of spindle-shaped or rounded cells with atypical and pleomorphic nuclei, occasionally presenting cross-striations. Usually, obvious nuclear atypia and pleomorphism allow the distinction between rhabdomyosarcoma and rhabdomyoma [1, 12]. Moreover, rhabdomyosarcomas are ill-circumscribed and infiltrative tumors [12].

Cardiac RM is often associated with tuberous sclerosis complex, whereas extracardiac forms are usually not. Cases of fetal RM have been described in patients with nevoid basal cell carcinoma syndrome with homozygous inactivating mutations in the PTCH gene [62]. In addition, transcriptional activation of PTCH has been observed in all sporadic adult and fetal RMs. This finding suggests that dysregulation of Hedgehog signaling may play an important role in the pathogenesis of syndromic and sporadic RMs [62].

The treatment of ARM involves surgical excision with preservation of the surrounding tissues [6, 10]. Recurrence is uncommon in oral ARM but may occur, especially in cases with incomplete surgical excision or in which the tumor appears as a multinodular lesion [4, 10]. Only 32 (40%) cases reported clinical follow-up, 5 (15.6%) of which experienced relapse after treatment (ranging from 2 to 132 months) [1–61]. As recurrence may be observed after several years, periodic follow-up is essential [6].

In summary, although rare, ARM has a predilection for the head and neck region. This lesion should be considered in the differential diagnosis of submucosal nodules mainly affecting the floor of the mouth.

Table 1 Oral adult rhabdomyoma published in the English-language literature

Authors	Year	Age (Years)	Sex	Site	Duration (months)	Follow-up
Beyer and Blair	1948	52	M	Floor of the mouth	24	No recurrence at 7 months
Mish	1958	21	F	Tongue	36	No recurrence at 48 months
Horn	1960	65	F	Floor of the mouth	48	NA
Sirsat and Vakil	1962	42	F	Soft Palate	2	NA
Tsukata and Pickren	1965	81	M	Floor of the mouth	3	No recurrence at 72 months
Kay et al.	1969	64	F	Floor of the mouth	2	NA
Assor and Thomas	1969	59	M	Submandibular region (multifocal)	6	NA
Wyatt et al.	1970	54	M	Tongue	4	No recurrence at 60 months
Tandler et al.	1970	69	M	Lip	NA	NA
Olofsson	1972	36	M	Floor of the mouth	18	No recurrence at 24 months
Albrechtensen et al.	1974	62	M	Floor of the mouth	NA	NA
Albrechtensen et al.	1974	29	–	Floor of the mouth	NA	NA
Ferracini et al.	1977	40	M	Floor of the mouth	NA	NA
Jones and Buntine	1977	49	M	Floor of the mouth	NA	NA
Caracta et al.	1978	55	M	Submandibular region	NA	NA
Everson and Merchant	1978	59	M	Floor of the mouth	36	NA
Heiden et al.	1978	50	F	Soft palate	NA	NA
Weitzner et al.	1979	60	M	Soft palate	2	No recurrence at 24 months
Solomon and Tolete-Velcek	1979	11	M	Tongue	NA	NA
Neville and McConnel	1981	58	M	Floor of the mouth (multifocal)	NA	NA
Warner et al.	1981	66	F	Submandibular region	NA	NA
Gardner and Corio	1983	60	M	Submandibular region (multifocal)	NA	NA
Schlossnagle et al.	1983	65	F	Submandibular region (multifocal)	NA	NA
Reid and Smith	1985	39	M	Floor of the mouth	NA	NA
Bock and Bock	1987	72	M	Soft palate	NA	No recurrence at 72 months
Bertholf et al.	1988	65	M	Floor of the mouth	4	No recurrence at 9 months
Nam et al.	1990	–		Base of the tongue	NA	NA
Walker and Laszewski	1990	76	M	Tongue (multifocal)	NA	NA
Sangueza et al.	1990	84	M	Tongue	NA	NA
Garcia-Ruiz et al.	1991	74	M	Submandibular region	60	No recurrence at 12 months
Napier et al.	1991	59	M	Floor of the mouth	NA	No recurrence at 48 months
Gibas and Miettinen	1992	34	M	Soft palate	NA	NA
Horn et al.	1992	52	F	Floor of the mouth	NA	NA
Shemen et al.	1992	53	M	Floor of the mouth (multifocal)	NA	NA
Shemen et al.	1992	75	M	Floor of the mouth	NA	NA
Fortson et al.	1993	71	M	Submandibular region (multifocal)	NA	NA
Kapadia et al.	1993	46	M	Tongue	NA	Recurrence at 96 and 102 months
Kapadia et al.	1993	81	F	Floor of the mouth	NA	NA
Kapadia et al.	1993	75	F	Soft palate	NA	No recurrence at 221 months
Kapadia et al.	1993	36	M	Buccal mucosa	NA	Recurrence at 72 months
Kapadia et al.	1993	51	F	Soft palate	NA	No recurrence at 216 months
Kapadia et al.	1993	59	M	Base of the tongue	NA	Recurrence at 2 months
Kapadia et al.	1993	60	M	Submandibular region	NA	No recurrence 27 months
Kapadia et al.	1993	55	M	Floor of the mouth	NA	Recurrence at 79 and 91 mo. No recurrence at 75 months
Kapadia et al.	1993	69	F	Base of tongue	NA	Recurrence at 132 mo. No recurrence at 156 months
Kapadia et al.	1993	49	F	Soft palate	NA	NA
Zachariades et al.	1994	49	M	Buccal mucosa	NA	NA

Table 1 (continued)

Authors	Year	Age (Years)	Sex	Site	Duration (months)	Follow-up
Zbaren et al.	1995	64	M	Submandibular region	NA	NA
Bastian and Brocker	1998	75	M	Lip	NA	NA
Ballester et al.	2000	–	–	Tongue	NA	NA
Moriniere et al.	2001	–	–	Base of tongue	NA	NA
Fukuda et al.	2003	51	M	Base of tongue	NA	NA
Favia et al.	2003	58	M	Floor of the mouth	NA	No recurrence at 144 months
Favia et al.gil	2003	63	F	Submandibular region	NA	No recurrence at 168 months
McGregor et al.	2003	78	M	Floor of the mouth	12	NA
Delides et al.	2005	59	M	Tongue (multifocal)	NA	NA
Hansen et al.	2005	82	F	Floor of the mouth	NA	NA
Liess et al.	2005	69	M	Submandbular region (multifocal)	NA	NA
Bellis et al.	2006	62	M	Submandibular region	24	No recurrence at 72 months
De Medts et al.	2007	65	M	Submandibular region (multifocal)	NA	NA
Bizon et al.	2008	65	M	Submandibular region (multifocal)	NA	NA
Catalfamo et al.	2010	52	M	Submandibular region	24	No recurrence at 48 months
Etit et al.	2010	67	M	Base of tongue	NA	NA
Gupta et al.	2010	43	M	Floor of the mouth	NA	NA
Gupta et al.	2010	37	M	Bilateral submandibular region	NA	No recurrence at 2 months
Parara et al.	2010	69	M	Floor of the mouth	3	No recurrence at 24 months
Maglio et al.	2012	72	M	Submandibular region	NA	–
Sirera and Sempere	2012	54	M	Floor of the mouth	12	No recurrence at 36 months
Zhang et al.	2012	78	M	Floor of mouth (multifocal)	2	No recurrence at 24 months
Vera-Sirera and Vera-Sempere	2012	54	M	Floor of the mouth	12	No recurrence at 36 months
de Tray	2013	55	M	Submandibular region (multifocal)	NA	NA
Schlittenbauer et al.	2013	38	M	Buccal mucosa	30	No recurrence at 2 months
Amelia Souza et al.	2013	40	F	Floor of the mouth	6	No recurrence at 24 months
Mengoli et al.	2016	48	M	Lip	NA	NA
Mistry	2017	60	M	Bilateral submandibular region	NA	NA
Andrade	2018	11	F	Tongue	4	NA
Dau et al.	2019	65	M	Soft palate (multifocal)	12	No recurrence at 36 months
Yadav et al.	2019	55	F	Floor of the mouth	2	NA
Hakim et al.	2020	56	M	Floor of the mouth	24	No recurrence at 12 months
Present case	2021	47	M	Floor of the mouth	24	No recurrence at 48 months

M male, *F* female, *NA* not available

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Declarations

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval This article does not contain any studies with human participants or animals performed by any of the authors.

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