

Case Report

An unusual Warthin's tumor arising from minor salivary glands in the floor of mouth

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Abstract: Background: Warthin tumor (papillary cystadenoma lymphomatosum) is a benign salivary gland tumor occurring almost exclusively in the parotid gland. Warthin tumor arising from the minor salivary glands is extremely rare, with a reported incidence of 0.1% to 1.2%. Case presentation: We report here a case of recurrent Warthin tumor arising from the minor salivary glands in the floor of the mouth in adult woman. The Computed tomography of the first presentation of the lesion (18 months ago) shows swelling on the floor of the mouth with cystic nature with 4.6 cm×5.2 cm size. The recurrent lesion was presented as a swelling of 2.6 cm×1.6 cm size (six months ago). Surgical local excision with wide tumor free margins was performed with a biopsy specimen sent to histopathology, which was diagnosed as papillary cystadenoma lymphomatosum. The patient has been followed up for six months without any signs of recurrence of the lesion. Conclusion: Warthin's tumor can arise "although rare" in the minor salivary glands of the floor of the mouth.

Keywords: Warthin's tumor, minor salivary glands, papillary cystadenoma lymphomatosum

Introduction

Warthin's tumor, also known as papillary cystadenoma lymphomatosum, or adenolymphoma, is a benign cystic tumor of the salivary glands, which is detected almost exclusively in the parotid gland, also, is the second most frequent benign salivary gland tumor following pleomorphic adenoma, Warthin's tumor accounts for about 15% of all epithelial tumors of the parotid gland [1]. In the WHO classification, the disease was named Warthin's tumor after the pathologist who published the first two case reports in the American literature in 1929 [1].

Extraparotid Warthin's tumor, may be arising from the submandibular gland, minor salivary glands and cervical lymph nodes but is very infrequent and extremely rare [2]. In this article, we present the first case report of Warthin tumor arising from the minor salivary glands in the floor of the mouth.

Case report

18 months ago, a 21-year-old Chinese non-smoker female was referred to the Department of Head and Neck Oncology, West China School of Stomatology, Sichuan University. The Patient reported that she noticed swelling on submental area 10 years ago with gradual and very slow growing with no other symptoms. The lesion appeared clinically as bilateral submental diffuse swelling, size of swelling was approximately 4 cm×4 cm, and the border was unclear, soft, palpable and diffuse, on floor of mouth. Computed tomography (CT) scan revealed a diffuse swelling on floor of mouth with cystic nature and sizing 4.6 cm×5.2 cm (**Figure 1**).

The clinical differential diagnosis was either mucocoele or dermoid cyst; the lesion was surgically excised under general anesthesia. Six months ago, the patient returned to the hospital with a swelling of the floor of the mouth with 2.6 cm×1.6 cm dimensions. The lesion was sur-

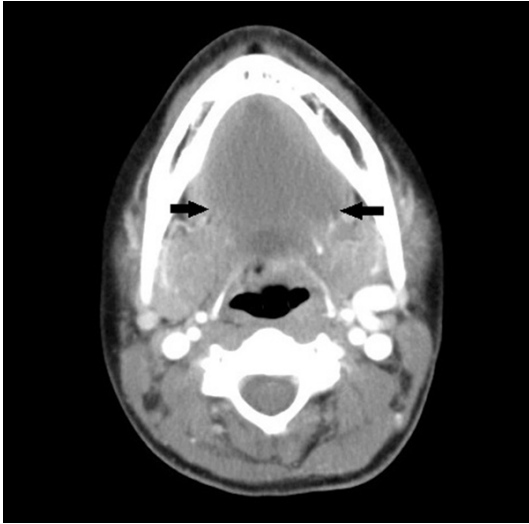


Figure 1. CT scan of the first presentation of Warthin's tumor, axial view. Pointed by an arrow.

gically excised with wide free margins. Histological examination with hematoxylin and eosin stain of the tumors revealed that the sections were grayish red, different sizes of cysts were seen. The tumor consisted of epithelial component and lymphoid stroma. The epithelial cells, the oncocytes, were disposed on two layers, a luminal layer of oncocytic columnar cells, supported by a discontinuous layer of oncocytic basal cells. The cystic spaces had epithelium referred to as papillary in foldings that protruded into them. In the surrounding of cysts, we saw varying degrees of lymphoid stroma which consists of small lymphocytes and some plasma cells, histiocytes and mast cells.

(The nuclei of the luminal cells appear uniform and display palisading towards the free surface, the basal cells possess round to oval nuclei, centrally located, small, with conspicuous nucleoli) (**Figure 2**). The histopathological diagnosis established was Warthin's tumor arising from minor salivary glands on the floor of mouth (**Figure 3**). The patient was followed up for six months without any signs of recurrence of the lesion.

Discussion

Warthin's tumor is a benign neoplasm of salivary glands, first described by Aldred Warthin in 1929, which occurs predominantly in the parotid gland and represents approximately 15% of parotid tumors, 8% of cases were reported

extra parotid and most often detected in lymph nodes of the cervical region [3]. Some cases reported in the submandibular glands, pharynx and larynx [4, 5]. The occurrence of Warthin's tumor in minor salivary glands is extremely rare and the incidence was reported between 0.1% to 1.2% [6]. The Incidence of Warthin's tumor commonly appears between ages of 58 and 70 years and can arise in adults; Warthin's tumor is more common in men than in women [7]. The precise etiology of Warthin's tumor is still unknown but smokers have 8 times risk of developing Warthin's tumor, the correlation between cigarette smoking and Warthin's tumor is hypothesized [8].

Due to the unusual occurrence of Warthin's tumor in minor salivary glands, Iwai et al [2] published case report and literature review based on the few articles published in the literature, according to the literature, they mentioned only 22 cases of Warthin's tumor arising from the minor salivary glands between 1960 and 2011, they were distributed with different locations in the oral cavity, 8 cases were originated in buccal mucosa (2 cases in cheek, 6 cases in buccal fold), 6 cases in the lip (4 cases in upper lip and 2 cases in lower lip), 7 cases in the palate and only one case was in oropharynxes. To our knowledge, our case is the 23th case of Warthin's tumor arising from the minor salivary glands and the first case in the floor of mouth occurring in non-smoker female adult. Because Warthin's tumor of minor salivary glands is extremely rare, the clinical diagnosis is very difficult, hence, all cases reported were misdiagnosed like (mucocele, minor salivary gland tumor, dentoalveolar abscess and Pleomorphic adenoma [9, 10]. Macroscopically, Warthin's tumor presents as an ovoid mass, with a dense fibrous capsule and displaying multiple cystic compartments filled with a viscous yellow material. Eveson and Cawson [9] found 77% of cases with an incomplete capsule, a full capsule in 8% and 16% of tumors in which there was no evidence of capsule. Microscopically, it is a well-circumscribed tumor, typically cystic and papillary, with an adenoid columnar eosinophilic oncocytic epithelial lining arranged in two layers resting on a background of lymphoid stroma. Akin et al [11] and Parnes [12] eventually diagnosed lesions "suspected to be Warthin's tumor" as a papillary cystadenoma because there was no evidence

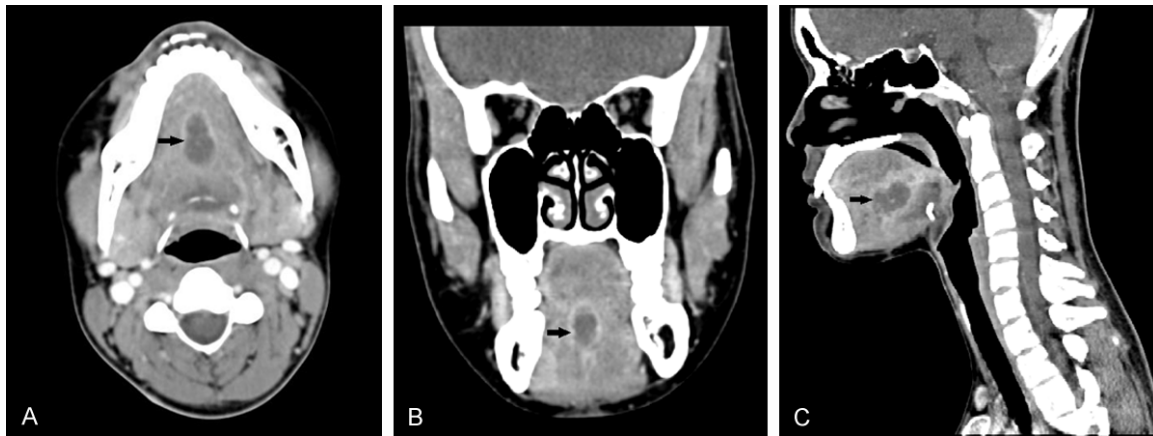


Figure 2. CT scan of the recurrent lesion. A. Axial view. B. Coronal view. C. Sagittal view. Pointed by arrows.

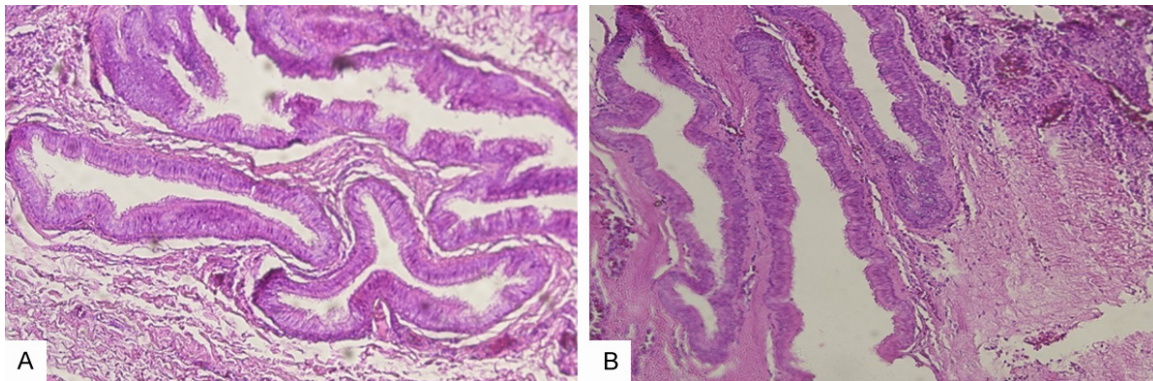


Figure 3. Histopathology, H&E staining showing oncocytic epithelium surrounded by lymphoid stroma at varying degrees (magnification 100×). A. First presentation, B. Recurrent lesion.

of a germinal center and lack of well-defined lymph nodules. The treatment of choice is complete local excision with a wide tumor free margins to prevent recurrence, a long period follow up is required to early detect the possibility of recurrence [13].

Conclusion

This case illustrates an unusual presentation of Warthin's tumor which arises from the minor salivary glands; the current case represents, to the best of our knowledge, the first documented instance of Warthin tumor of the floor of mouth.

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Informed consent for publication is signed by the patient to be provided upon request.

Disclosure of conflict of interest

None.

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