Parotid mass, Metaplastic Whartin’s Tumor case report

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ABSTRACT

The Whartin Tumor is the second most frequent tumor of the parotid gland following pleomorphic adenoma. Among the Whartin tumors, Metaplastic Whartin Tumor (MWT) is rare. In case of a MWT, histopathological replacement of oncocytic cells by squamous cells, ruptured epidermoid and lymphoepithelial cystic areas, a large necrosis area, fibrosis and granuloma formation may be seen. MWT may display similar clinical characteristics as the malignant parotid tumor that causes sialadenitis, abscess formation, skin ulceration, and facial paralysis. Although the histopathogenesis of Metaplastic Whartin Tumors is not exactly known, it is postulated that the histopathological processes caused by trauma, infection, or radiotherapy have a role in the formation of these tumors. This case study presents a 48-year-old female patient with a complaint of panicula before her left ear persisting for 5 months. Following a superficial parotidectomy carried out 3 weeks after the fine-needle aspiration biopsy (FNAB), her pathological assessment was found to be in line with Metaplastic Whartin Tumor. Our case study presents the case of a Metaplastic Whartin Tumor case with a review of literature on the subject, accompanied by radiological and histological analyses.

Keywords: Head; Neck Cancer; Precancerous Conditions Oral; Maxillofacial Surgery

1. INTRODUCTION

The Whartin’s tumor is the second most frequent tumor of the parotid gland following pleomorphic adenoma [1]. Whartin’s tumors are classified into four histologic subgroups: the first three depending on the existence of changes in the epithelial-stromal components, and the fourth depending on the metaplastic-squamous changes [2]. The Metaplastic Whartin’s Tumor (MWT), which is the fourth type, is rarely seen with a frequency of 6.2% among the Whartin’s tumors [1]. With MWT cases, histopathological replacement of oncocytic cells by squamous cells, ruptured epidermoid and lymphoepithelial cystic areas, a large necrosis area, fibrosis and granuloma formation may be seen [1,3]. In some cases, however, following the fine-needle aspiration biopsy (FNAB) epidermoid granulomas, cholesterol clefts, and hemocidin deposits can be detected [4-6]. MWT may sometimes display similar clinical characteristics as the malignant parotid tumor that causes sialadenitis, abscess formation, purulent fistula, skin ulceration, and facial paralysis [7-12].

For the diagnosis of MWT a microscopic analysis with Hemotoxilin Eosin is generally sufficient. To differentiate between flat-cell and mucoepidermoid carcinoma is of utmost importance since these two might be confused histopathologically [13,14].

2. CASE REPORT

A 48-year-old female patient presented to our ear, nose and throat clinic (ENT) with a swelling of panicula that was gradually getting bigger before her left ear persisting for 5 months. Physical examination revealed a mass which was 15 × 20 × 15 mm in size, and was rigid, semi-mobile, and a little sensitive to palpation. The rest of her ENT examination results were normal.

Parotid ultrasonography revealed a hypoechoic solid lesion with smooth borders that was circular in shape, and of 17 × 20 × 20 mm in size having septations and ecogenic solid component in the left parotid superficial lobe was reported. The FNAB showed oncocytic cells with single and three dimensional strata on a hemorrhagic and partly lymphocytic ground (Figures 1 and 2). With this result the patient was pre-diagnosed with Whartin’s tumor.

Three weeks after the FNAB the patient underwent left superficial parotidectomy under general anesthesia. During the procedure it was seen that the mass had partial cystic formations and it was dissected from the tissues surrounding the parotitis with difficulty because of intense fibrosis.
Figure 1. During the fine needle aspiration biopsy oncosyte-like cells in small and partly three dimensional levels were seen (Giemza ×20).

Figure 2. Lymphocytic ground of the fine needle aspiration biopsy (Giemza ×60).

Pathological analysis of the specimen revealed three-dimensional solid structures formed by eosinophilic stolplasmal oncosytic structures. In some of the regions of these structures total infarct areas and papillary structures were detected. Further, characteristic squamous metaplasia and intense inflammation were seen in the surrounding tissues (Figures 3-6). As a result of this pathological evaluation the patient was diagnosed with Metaplastic Whartin’s Tumor. After surgical therapy, the patient was irradiated with 3 cGy to the parotid region. No complications or relapses occurred during the one year postoperative follow-up period.

3. DISCUSSION

FNAB offers a high diagnostic gain in salivary gland lesions and is also a safe, fast, inexpensive, physician-friendly, and an easily tolerable diagnostic method by patients. The most significant point regarding FNAB’s diagnostic importance in salivary glands is that it enables the pathologist to have a rate of 96% right diagnosis with its high sensitivity and specificity for all salivary gland tumors [15].

Malignant salivary gland tumors are histologically labile and structurally complex tumors. Epithelial, non-epithelial tumors, lymphomas, metastatic tumors, and non-neoplastic lesions are the malignant tumors of the salivary gland. This diversity makes it difficult to reach a diagnosis through cytological analysis [16]. FNAB has been shown to have 5.5% wrong positive
Figure 5. In a couple of areas intense lymphocytic cells in papillary structures and stroma within the infarcted regions were seen (HE ×20).

Figure 6. Intense lymphocytic cells in the stroma under the cystic cavity were detected (HE ×4).

and 2.7% wrong negative rates with parotid masses [17]. Although there are limited number of cases, one of the disadvantages of FNAB is the tumor cultures seen along the needle tract [15]. Because of these FNAB’s role in the diagnosis of malignant parotid masses is still questionabile.

Although the histopathogenesis of Metaplastic Whartin’s Tumors is not exactly known, it is argued that the histopathological processes caused by trauma, infection, or radiotherapy have a role in the formation of these tumors [18]. Most of the patients with Metaplastic Whartin’s Tumor have a history of trauma because of FNAB (specifically FNAB), [19,20] and a small portion of them, 20% - 40%, have a history of radiotherapy [2].

Especially the other tumors (hurtle cell adenoma of the thyroid) rich in oncysytic cells and mitochondria have also infarcts and squamous metaplasias of different levels following FNAB as is seen with this tumor [21-25].

Following the FNAB some changes may also take place in the inflammatory process of the tumor mass and the surrounding tissues [26]. As a result, inflammation and fibrosis may be seen around the facial nerve and xanthogranulomatous sialadenitis may develop [9,27].

All these changes may be seen in other organs like the lungs, lymph node, and the thyroid besides the salivary glands following the FNAB [22,24,25]. Experimental trials conducted with rats’ parotid tissues demonstrated that inflammatory processes as well as metaplastic changes start to take place in a couple of days when the arteries feeding the parotid are bound [28]. It has also been shown that the period for the development of squamous metaplasia is 6 to 101 days between the FNAB and the surgery [3,29].

Squamous metaplasia cannot be seen in every Warthin’s tumor where ischemia and necrosis occur. Moreover, metaplastic epithelium in metaplastic Warthin’s tumor does not have atypical cytological features and infiltrating growth pattern [14].

Most of the reported cases of malignant transformation in Warthin’s tumor did not show distant metastasis. However, local lymph node metastasis was reported [2].

Treatment of this tumor is primarily surgical, consisting of adequate excision of the primary site with or without neck dissection [30]. However, neither radiotherapy nor chemotherapy changed the prognosis, and these patients died earlier [31]. Surgeons usually use postoperative chemotherapy and radiotherapy if the histological type of the tumor is Adenocarcinoma [32].

The present case was diagnosed as having squamous metaplastic changes and intense inflammation, and underwent a superficial parotidectomy without neck dissection. After surgical therapy, the patient was irradiated with 3 cGy to the parotid region. The present case has a negative systemic screening for primary cancer, and no recurrence of the tumor for 1 year. The long-term prognosis of these patients is not clear.

Since there are no studies carried out with large case series regarding the frequency with which the FNAB causes metaplasia in patients with the Whartin’s tumor, it has not been possible to point out to the details of this relation in full extent. It should not be disregarded, however, that all Metaplastic Warthin’s Tumor patients do not have either a history of trauma or a history of FNAB [33]. But it should also be taken into consideration that with the patients diagnosed with the Whartin’s tumor following a FNAB, inflammation and tissue changes based on necrosis during the surgery may very well be seen.

In terms of a differential diagnosis, it is also important to direct the attention of the pathologist to the metaplastic changes during the postoperative period with those patients with a history of FNAB who had under-
gone surgery having been prediagnosed with the Warthin’s tumor and who had difficulties of this kind during the surgery.

REFERENCES


