


Misdiagnosis of Adenoid Cystic Carcinoma of Palate: A Case Report

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Introduction: Neoplasms of the salivary glands are uncommon, accounting for approximately 3 to 5 percent of all head and neck tumors. Adenoid cystic carcinoma (ACC) is a rare salivary gland malignancy with a poor survival rate because of a high rate of late local recurrence and distant metastasis. **Materials and Methods:** This paper reports a misdiagnosis of palatal ACC via fine needle aspiration cytology (FNAC). A 27-year-old female with painful swelling in the right side of the palate underwent FNAC and was misdiagnosed with pleomorphic adenoma. During surgery, hemi-maxillectomy was performed due to the fragile consistency of the tissues. **Result:** The tumor was found to be ACC after histopathologic evaluations. Hence, the patient underwent radiotherapy and chemotherapy postoperatively. **Conclusion:** Given specific limitations and diagnostic pitfalls, it seems that pre neural and pre vascular invasion of tumors as a sign of malignancy should be noticed.

Keywords: Adenoid cystic carcinoma; Fine needle aspiration cytology; Neoplasms

Introduction

Neoplasms of the Salivary gland are a number of tumors with variable clinical, histopathological, and immuno-histochemical features. These tumors are extremely rare, accounting for only 3 to 5% of all neoplastic changes, occurring in the jaws. Most of these neoplasms are benign. Neoplasms of the Minor salivary glands, such as those occurring in the cheek mucosa, lip and tongue, are even rarer (1). Adenoid Cystic Carcinomas (ACC) are slow growing malignant neoplasms with high potential for local recurrence (2). With a reported yearly incidence of 3–4.5 cases per million, Adenoid Cystic Carcinoma (ACC) is considered a rare tumor, accounting for about 1% of all head and neck malignancies and about 10% of all tumors of the salivary glands. It is the most commonly reported malignant tumor of the minor salivary glands (MSGs) and it is also one of the most common cancers of the major salivary glands (3). ACC is characterized by slow local growth, high tendency for peri-neural invasion (PNI), infrequent regional metastases and frequent

development of local recurrences and most of all slow progression and relatively frequent distant metastases. ACC occurs in all ages and is not related to any known risk factors (4). As is the case for all salivary gland neoplasms, preoperative diagnostic imaging of ACC includes computed tomography (CT) and/or magnetic resonance imaging (MRI). This allows the surgeon to estimate the anatomical extent of the disease, which is important for precise surgical planning. It is well accepted that CT-Scan is better at delineating bone invasion, while MRI is superior for assessing soft tissue extension (3). Fine needle aspiration cytology (FNAC) has been widely employed in pre-operative diagnosis of salivary gland lesions for many years. Different studies in the literature have shown a wide range of sensitivity and diagnostic accuracy for FNAC (5). Sometimes FNAC may have false-negative and false-positive results, so in each case, we should interpret Fine Needle Aspiration Cytology report with the entire clinical signs and symptoms of the patient (6).

Case Report

A 27-year-old female was referred to oral & maxillofacial surgery department of Shiraz University of Medical Sciences with the chief complaint of painful swelling in the right side of palate. The history revealed that the swelling was noticed by patient approximately 1 and a half year ago, which due to pregnancy and lack of pain, the patient didn't seek treatment. Therefore after 1 and half year, because of increased swelling and the incidence of pain, the patient was referred to medical clinic and underwent FNAC (Fine needle aspiration cytology) and the lesion was diagnosed to be pleomorphic adenoma. Then for surgical treatment the patient was referred to oral & maxillofacial surgery department of Shiraz University of Medical Sciences. On extra-oral examination there was no evidence of lymphadenopathy or any abnormal findings and the patient didn't report any numbness or paresthesia in the region. There was no evidence of pus or soreness. On Intra-oral examination there was a sessile exophytic growth on the right palate, extending from anterior region to tuberosity and across the mid-palatine raphe. Palpation revealed tenderness on the site of the lesion with firm consistency and smooth texture and with intact and normal overlying mucosa (Figure 1). Medical history was noncontributory.

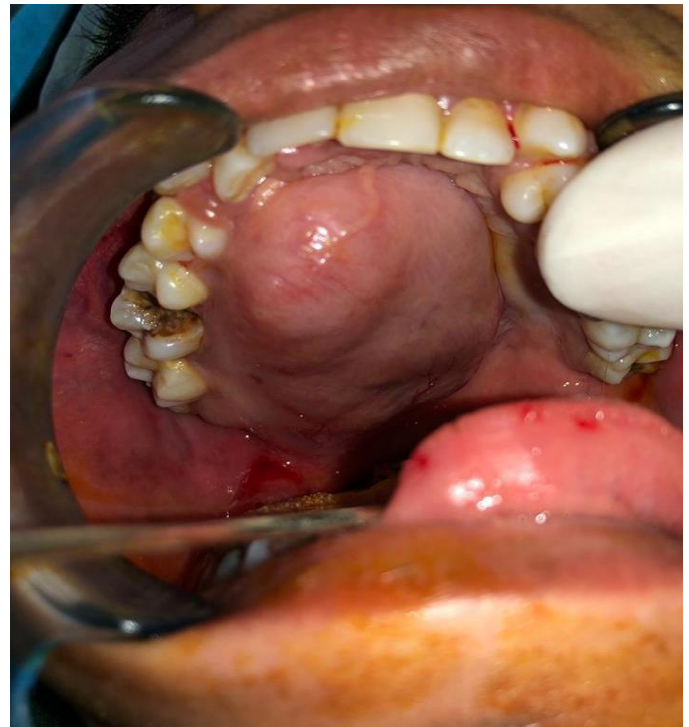


Figure 1. Smooth texture with intact and normal overlying mucosa

Radiographic Description

CT scan was performed to examine the lesion in detail. The Axial section demonstrated a large lesion measuring approximately 52/03 mm anterior-posteriorly and 47/05 mm mediolaterally (Figure 2). A destructive lesion with soft tissue density and invasive ill-defined periphery was depicted in the right side of maxilla. The lesion expanded posteriorly from the region of central incisors to maxillary tuberosity and lateral pterygoid plate of the left side (Figure 2). Destruction of medial and lateral pterygoid plates and invasion to pterygomaxillary space and alveolar process was clearly detected. Also, the pharyngeal space was involved, but not obstructed (Figure 3). The invasion to the facial nerve and carotid artery was also demonstrated. Destruction of medial, anterior and posterior walls of right maxillary sinus along with invasion to right and left nasal cavity due to destruction of nasal septum was seen (The lesion crossed the midline). The buccal cortex had a moth-eaten pattern (Figure 3).

Coronal section demonstrated extension of the lesion with the largest dimension of 50.01 mm superior-inferiorly and 48.26 mm mediolaterally (Figure 4). There was no evidence of soft tissue enlargement, striation of fatty tissue and destruction of borders of the facial muscles, thus we could assume that the lesion was not of inflammatory origin.

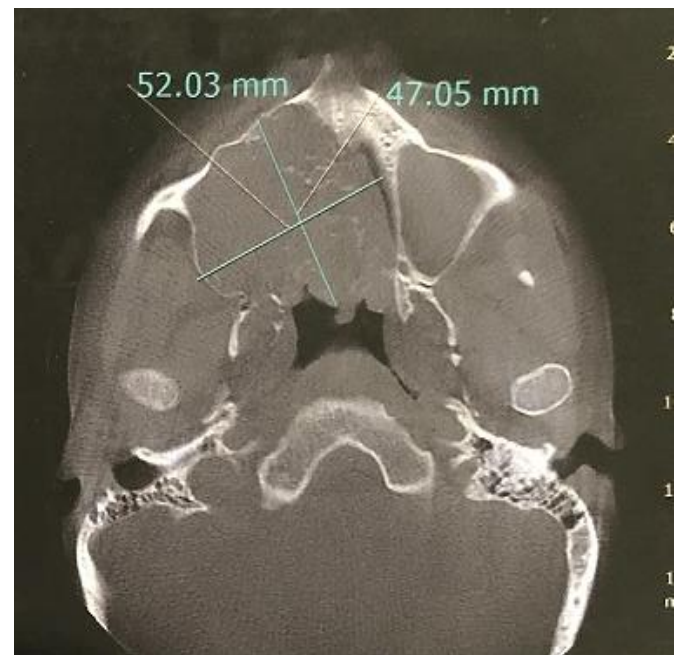


Figure 2. An axial section demonstrating a lesion measuring 52.03 × 47.05 mm



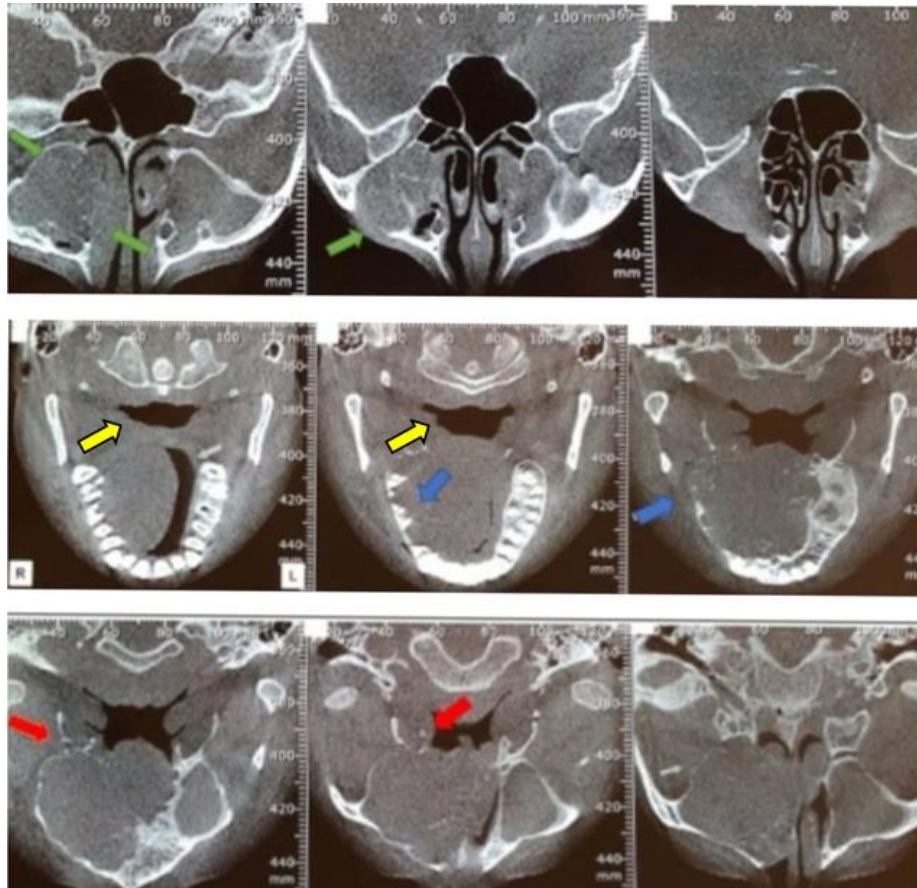


Figure 3. Axial section. Destruction of medial and lateral pterygoid plates (red arrows). Invasion to pharyngeal space (yellow arrows), alveolar process (blue arrows). Destruction of medial, anterior, and posterior walls of right maxillary sinus along with invasion to right and left nasal cavities (green arrows)

The vertical boundaries of the lesion were floor of the orbit and floor of the superior ethmoid sinuses and palatal bone. Thinning of inferior orbital rim (moth-eaten pattern) and involvement of infra-orbital foramen and middle and inferior ethmoid air cells on both right and left side along with the inferior concha on both sides was well recognized. The left maxillary sinus and alveolar process was still intact. Thinning of buccal cortex and the perforation of palatal cortical plate was marked and fragments of residual bone was detectable inside the lesion. There was also thinning and perforation in lateral wall of maxillary sinus. Invasion to adjacent soft tissue clearly depicted (Figure 5).

Sagittal sections demonstrated invasion to adjacent soft tissue, destruction of ethmoid air cells and thinning of buccal cortex (Figure 6).

After obtaining radiographic examinations the patient was scheduled for resection surgery but due to fragile consistency and destructive pattern of gross, wide surgical excision with clear margins (1 cm) and Hemi Maxillectomy of right maxillary region

was performed.



Figure 4. The coronal section demonstrated a lesion measuring 50.01 × 48.26 mm



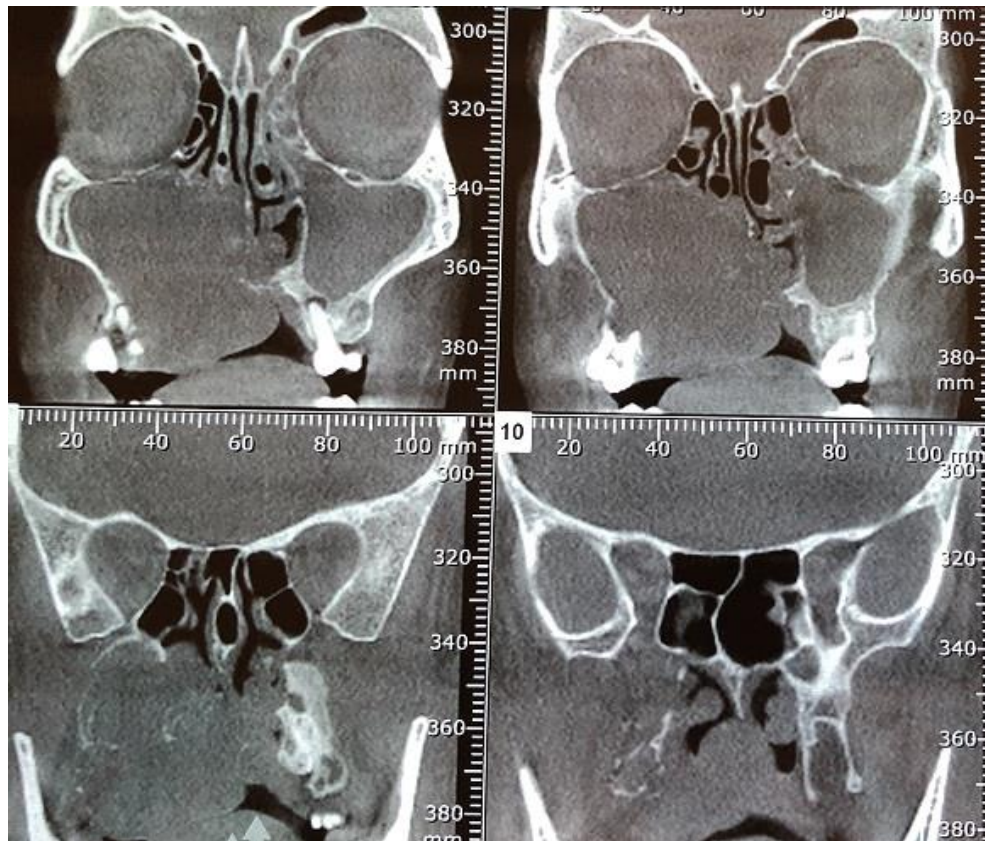


Figure 5. Coronal section. Thinning of inferior orbital rim and involvement of infra-orbital foramen and middle and inferior ethmoid air cells on both right and left side along with the inferior concha on both sides. Thinning of buccal cortex and perforation in palatal cortical plate and thinning and perforation in lateral wall of maxillary sinus also is seen.

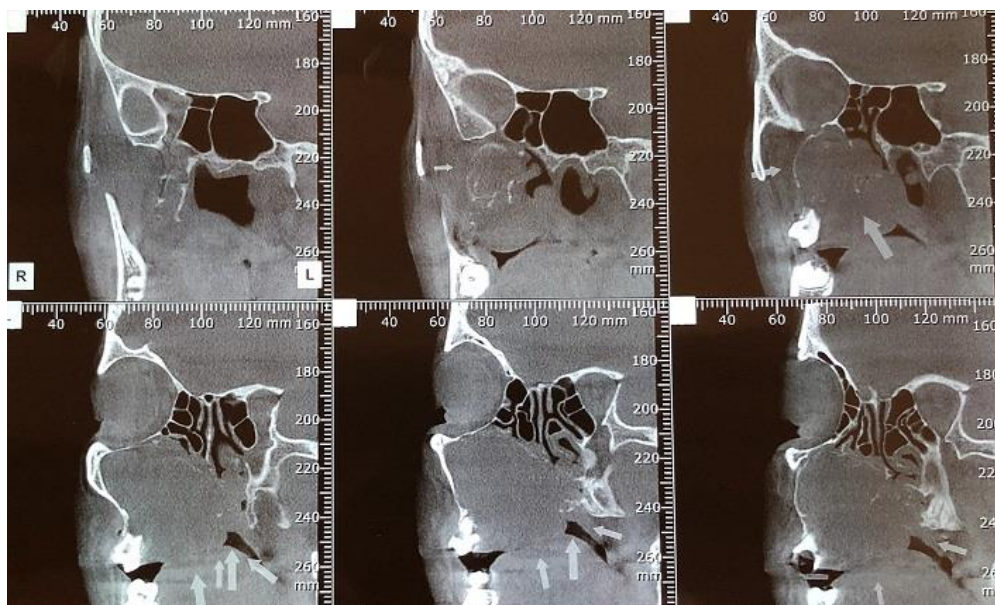


Figure 6. Sagittal sections



After prep & drape under general anesthesia in supine position with hypotension control, through right Weber Ferguson flap, the lesion was exposed and after osteotomy with bur and chisel the lesion was excised (Hemi Maxillectomy) with safe margins confirmed free margins with frozen sectioning evaluation (Figure 7). Right malar area was reconstructed with thick titanium mesh fixed to right zygomatic body and infraorbital rim with mini screws to support the flap and for esthetic reasons. Then the flap was reflected and sutured in 3 layers with vicryl 3-0 and nylon 5-0. The specimen was then sent to pathology

department.

Histopathological examination revealed a cellular neoplasm which was composed of hyperchromatic cells with cribriform and solid architecture with perineural and lymphovascular invasion (Figure 8). Also, right maxillary sinus was reported to be completely invaded by the tumor, while the left maxillary sinus was tumor-free. Histopathological examination supports the diagnosis of Adenoid cystic carcinoma and the patient was administered to postoperative radiotherapy and chemotherapy.



Figure 7. Hemi Maxillectomy of right maxillary region

Discussion

Salivary gland tumors are group of neoplasms that are characterized by distinctive different clinical and histological features. Pleomorphic adenomas (PA) is the most common benign salivary gland tumor, whereas Adenoid cystic carcinoma (ACC) is one of the most frequent salivary gland malignancies of

the head and neck. It appears in 10– 15% of all salivary neoplasms (3-7). Fine needle aspiration cytology (FNAC) is widely accepted as a useful, reliable and minimally invasive method for the diagnosis and treatment of salivary gland lesion (8-10). Numerous studies investigated diagnostic accuracy of FNAC.



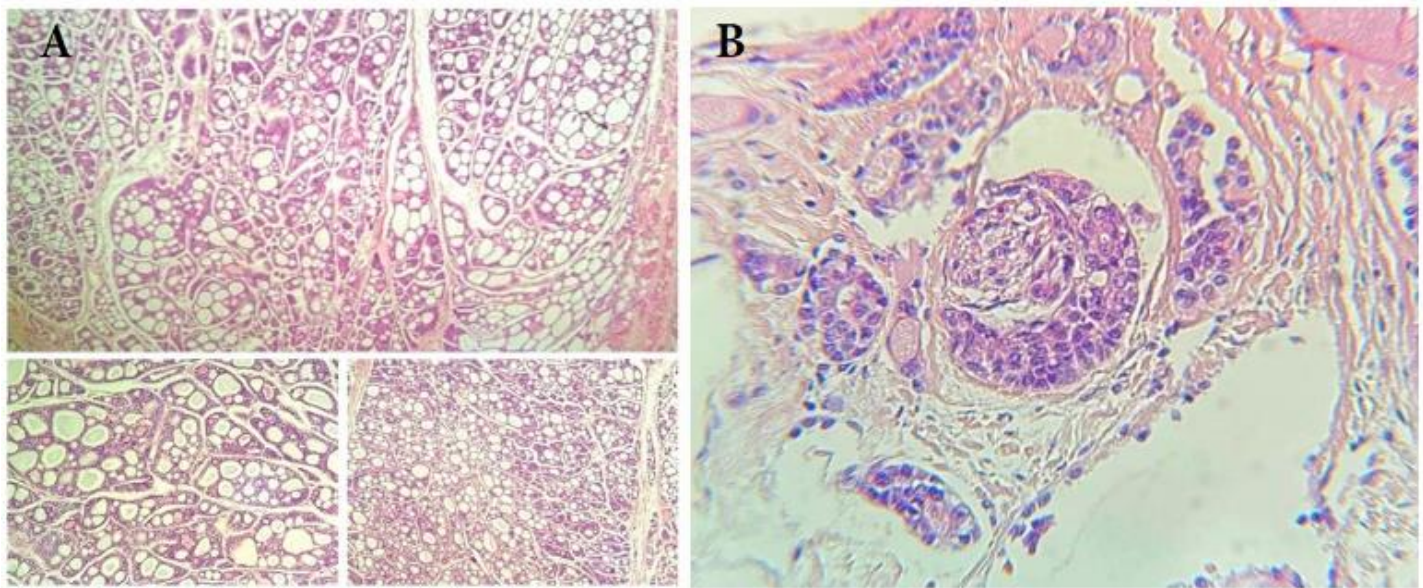


Figure 8. Hyperchromatic cells with cribriform and solid pattern(A); Perineural and lymphovascular invasion of tumor(B)

Samreen Naz and *et al.*, studied 187 cases who presented with parotid and submandibular swellings and the results of FNAC and histology were compared. They reported two cases of false negative diagnosis which were one each of mucoepidermoid carcinoma and acinic cell carcinoma which were initially diagnosed as pleomorphic adenoma on FNAC and 3 cases of false positive diagnosis. In one of that case there were extensive squamous elements without any other component and therefore a diagnosis of neoplastic lesion, suspicious for malignancy was given with a possibility of metastatic squamous cell carcinoma and mucoepidermoid carcinoma.

The final histology revealed the diagnosis of pleomorphic adenoma. The other two cases with false negative diagnosis were that of mucoepidermoid and adenoid cystic carcinoma which were inaccurately diagnosed on FNAC as pleomorphic adenoma, they revealed an overall diagnostic accuracy of FNAC to be 83.8% (11). Magdalena Chrabańska and *et al.*, studied 247 cases who had salivary gland lesions and underwent FNAC. 101 cases had been confirmed by histopathology examination. The cases with discrepancies between the FNAC and histopathologic results were reviewed to establish possible reasons for discordance. as a result, 135 cases were diagnosed as benign neoplasms, 15 as malignant neoplasms, and 97 as non-neoplastic lesions. Out of the 101 cases with histopathologic confirmation, discordant results between cytologic and histopathologic diagnosis were observed in 15 cases. They revealed no false positive and 4 false negative results for cancer. Cystic presentation of a lesion was considered as a reason for diagnostic pitfall. FNAC sensitivity was reported in the range Of 75%-

100%, specificity 81-100%, diagnostic accuracy 85-96%, PPV 31-100% and NPV 60-96% which emphasized that FNAC is a highly sensitive and specific method for diagnosis of most salivary gland lesions (12). one of the common and clinically important differential diagnoses that is challenging for cyto-pathologist is between pleomorphic adenoma and adenoid cystic carcinoma (13) because both PA and ACC can grow in a cylindromatous or cribriform pattern, and form hyaline globule (14, 15). The clinical prognosis and treatment of the two lesions are very different, For a benign tumor such as palatal PA, the surgery of choice is wide local excision of the lesion and surrounding capsule along with clear margins, involving the periosteum and associated mucosa, followed by curettage of the underlying bone to prevent recurrence (16), but For malignant salivary gland neoplasms such as Adenoid cystic carcinoma with poor prognosis, Surgical resection followed by post-surgery radiotherapy of the region is usually the treatment of choice (17), therefore it is important to distinguish between these two lesions. The neoplasms that are most frequently confused cytologically with Adenoid cystic carcinoma are pleomorphic adenoma and basal cell adenoma because ACC can show basement membrane like material which may be misinterpreted as stromal component. Also, the epithelial component of adenoid cystic carcinoma is usually very bland leading to inaccurate diagnosis of benignancy (11), similar to the case discussed. Patient survival is directly related to complete resection of the lesion. Considering the high rate of perineural invasion of ACC and sometimes difficulty of achieving the region of resection (18), failure to early diagnose can affect the patient survival rate. Computed



tomography (CT) and magnetic resonance imaging (MRI) can be cited as reliable and useful methods for demonstrating the extension of the ACC and predicting the prognosis (18-20). Yang Li and *et al.*, studied twenty cases of ACC in the maxillary sinus which were previously diagnosed histopathologically and investigated computed tomography (CT) and magnetic resonance imaging (MRI) findings and their correlation with all three histopathologic types of ACC included the tubular, cribriform, and solid. They reported significant differences in range, size, shape, margins, type of bone destruction and time intensity curve (TIC) among the three histopathologic types on CT scans. All tumors indicated heterogeneous intensity signal on T1- and T2-weighted images (T1WI and T2WI) and demonstrated as hypo- or isointense small cystic lesions on T1WI and hyperintense on T2WI ($n = 6$). marked heterogeneous enhancement for all lesions has been depicted on Postcontrast MRI. as a result they revealed that both CT and MRI modalities are advantageous methods in the diagnosis of ACC in the maxillary sinus and correlation between their findings and histopathologic features can help with diagnosis and treatment plan (20). Ying Liu and *et al.*, did the meta-analytic research based on literature published between 1982 and 2013 to compare ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI) for clinical differential diagnosis in patients with salivary gland tumor (SGT). Considering inclusion and exclusion criteria, 784 patients with 792 SGTs between 42 to 63 years were studied. The data from the literature confirmed computed tomography was recommended as an effective diagnostic method in patients with primary SGT and there was no difference between CT and MRI statistically. All three modalities were considered as reliable imaging techniques for differential diagnosis of SGTs (19).

Conclusion:

Despite high sensitivity of FNAC in diagnosing benign from malignant lesions, due to some overlapping features and subsequent diagnostic pitfalls it seems that pre neural and pre vascular invasion of tumors in graphies as a sign of malignancy should be noticed.

Conflict of Interest: 'None declared'.

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