Journal Homepage: - www.journalijar.com
INTERNATIONAL JOURNAL OF ADVANCED RESEARCH (IJAR)
Article DOI: 10.21474/IJAR01/6368
DOI URL: http://dx.doi.org/10.21474/IJAR01/6368

RESEARCH ARTICLE

RECURRENT WARTHIN TUMOR PRESENTING WITH SYNCHRONOUS, BILATERAL PAROTID AND MULTI-CENTRIC UPPER NECK INVOLVEMENT.

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Manuscript Info

Manuscript History

Received: 21 November 2017
Final Accepted: 23 December 2017
Published: January 2018

Abstract

Background: Warthin tumor is the second most common benign salivary tumor. This case highlights a patient who underwent local excision of a solitary Warthin tumor from the peri-parotid area. He subsequently presented with recurrent, multi-centric disease with bilateral parotid and cervical lymph node involvement. **Case Report:** A 45 year old, tobacco-using male was referred for a right head and neck mass. Exam revealed a mobile, painless mass near the angle of the jaw. Nasofibroscopic exam revealed a non-obstructive nasopharyngeal mass. CT confirmed an ovoid mass inferior to the right parotid gland. Fine-needle aspiration (FNA) was non-diagnostic. Deep biopsies and local excision of the right neck mass were performed. Nasopharyngeal biopsies were negative; the neck mass was consistent with Warthin tumor. The patient returned approximately 4 years later with a right pre-auricular mass and right upper neck lymphadenopathy. Diagnostic work-up, including ultrasound-guided FNA, revealed bilateral parotid gland Warthin tumor. The patient was symptomatic from the right-sided disease and not the left. Consequently, he elected right parotidectomy and excision of the right upper neck lymph nodes and clinical follow-up of the left mass. Upon pathologic review, there was Warthin tumor present in the parotidectomy and right lymph node specimens.

Introduction:-

In this case report, we describe a patient with recurrent Warthin tumor in both parotid glands and lymph nodes of the upper neck. Warthin tumor is the second most common benign salivary tumor [1]. Studies have demonstrated that the epithelial and lymphocytic components of Warthin tumor are polyclonal and hence this lesion is best described as non-neoplastic [2,3]. Multi-centric recurrence is not due to disease spread; rather, it is likely due to the heterotopia of salivary tissue and sub-clinical multi-centric disease at initial presentation.

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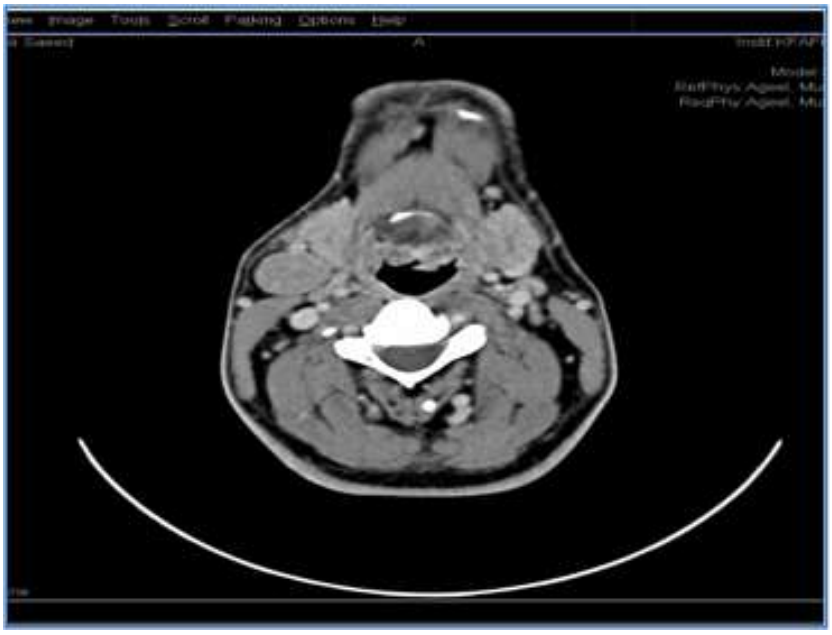
Case Report:-

A 45-year-old male was referred to our otolaryngology practice with a right neck mass with an underlying concern for metastatic head and neck cancer. He smoked one pack of cigarettes a day and had a history of insulin-dependent diabetes mellitus. A directed head and neck exam revealed a mobile, painless 2x2 cm right neck mass near the angle of the jaw with normal overlying skin. Nasofibroscopic examination revealed a smooth, non-obstructive nasopharyngeal mass. CT of the neck revealed normal parotid and submandibular glands. However, there was an ovoid, soft tissue neck mass inferior to the right parotid gland. After IV contrast administration, there was almost homogenous enhancement of a 2.6x2.0x4.1 cm peri-parotid mass which was clearly delineated from the surrounding soft tissue; there was no enhancement in the nasopharyngeal area. Note was also made of bilateral sub-centimeter cervical lymphadenopathy **Picture.1.**

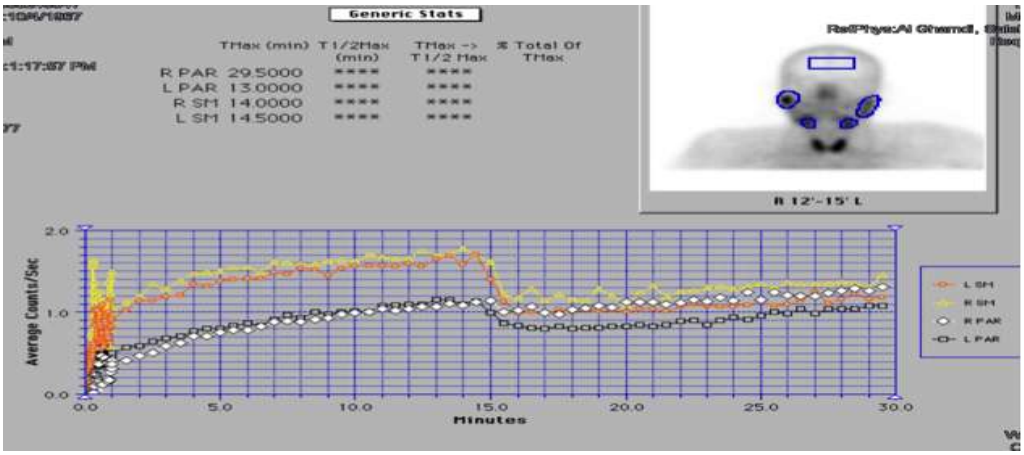
FNA under local anesthesia revealed no evidence of malignancy in the nasopharyngeal mass. However, in the palpable right neck mass, borderline malignancy could not be ruled out. Subsequently, under general anesthesia, the patient was subjected to local excision of the palpable right neck mass and deep biopsy of the nasopharyngeal mass. The histopathology for the nasopharyngeal mass was negative. However, the excisional biopsy of the palpable mass revealed a well-circumscribed lesion composed of a cystic area, necrotic inflammatory debris, and two layers of oncocytic cells within a lymph node consistent with Warthin tumor involvement.

After several months, the patient was lost to follow-up. However, approximately 4 years later, the patient presented to the clinic with a painless and enlarging right pre-auricular mass and right upper neck lymphadenopathy. An ultrasound was performed and revealed a hypo-echoic, smooth walled mass in the right pre-auricular region, enlarged, benign-appearing right and left cervical lymph nodes, and a heterogeneous 2.1 x 1.0 cm lesion with smooth borders and vascularity in the left pre-auricular area. On nuclear scan, both parotid and mandibular glands demonstrated good uptake of tracer with spontaneous excretion after lemon juice. However, there was focal uptake seen in the upper portion of the right parotid gland **Picture.2.** Ultrasound-guided FNA of the right and left parotid masses confirmed Warthin tumor.

Because the patient was more symptomatic with respect to the right mass and lymph nodes and not the left mass, he opted for right parotidectomy and excision of the right upper neck lymph nodes and close clinical follow-up of the left mass. Upon pathologic review, Warthin tumor was found in the parotidectomy and right lymph node specimens.



Picture.1:- CT scan of the neck



Picture 2:- Nuclear scan, both parotid and mandibular glands.

Discussion:-

Warthin tumor is the second most common benign salivary neoplasm [1]. Also known as papillary cystadenoma lymphomatosum, this lesion is typically surrounded by a thin capsule and has lymphoid stroma, cystic changes, and intraluminal papillary projections lined by the characteristic bi-layered epithelium of oncocytic cells surrounded by smaller basal cells [1]. PCR analysis has demonstrated that the epithelial and lymphoid components of Warthin tumor are polyclonal [2,3] which corresponds to its benign, non-neoplastic behavior. Although the tumor is more commonly seen in older adults (6^{th} decade in women and 7^{th} decade in men), it can occur in younger patients as described in this case [4]. This patient's presentation with a painless mass and smoking history is characteristic; a positive smoking history has been found in over 88% and 89% of men and women, respectively [4].

This tumor tends to present in the parotid glands and peri-parotid lymph nodes [1]. However, in large registries, Warthin tumor outside of the parotid or peri-parotid tissue is noted in up to 8% of cases, most commonly in the cervical lymph nodes, submandibular glands, and larynx [5,6]. There are several theories to explain this localization pattern and histogenesis. The most favored theory postulates that the tumor results from the proliferation of salivary gland ductal cells that were trapped in intra- and peri-parotid lymph nodes during embryonal life [5]. The multi-centric and extra-parotid potential of this tumor is similarly attributed to this same embryonal heterotopia of salivary tissue [5]. The other favored theory suggests that these tumors are adenomas developing from the ductal epithelium of the parotid gland with subsequent lymphocytic infiltration [7].

On ultrasound, Warthin tumors appear as well demarcated, hypo-echoic, and hypervascularized oval lesions [8]. In a retrospective study of 34 Warthin tumors, 94% of these tumors had greater uptake than normal parotid gland tissue on Tc-99m scintigraphy after lemon juice stimulation [9]. On CT scans, Warthin tumors appear as well-delineated lesions commonly involving the inferior pole of the gland; the presence of bilateral or multiple lesions is suggestive, but not diagnostic of Warthin tumor [10]. A validation study showed that gadolinium-enhanced dynamic MRI has been found to have a 100% sensitivity and an 80% specificity in differentiating malignant salivary tumors from benign tumors [11]. Consequently, tissue sampling is regarded as necessary for a definitive diagnosis. In a review of 341 salivary gland FNA biopsies, cytologic sampling showed overall sensitivity, specificity, and accuracy of 92%, 100%, and 100%, respectively [12]. On FNA, typical cytomorphological features of Warthin tumor include monolayered sheets of oncocytic cells, lymphocytes, and amorphous, cystic debris [13].

When factoring in synchronous and metachronous clinical presentations, Warthin tumor is multi-centric in 12-20% of patients and bilateral in 5-14% [1]. In one study, when parotidectomy specimens were examined with whole-organ sectioning, 50% had multiple tumors [14]. Similarly, in a retrospective, cross-sectional study, only 19% of multi-centric tumors were identified by palpation and only 28% by imaging investigations [15]. Recurrence rates for solitary and multi-centric tumors were 0 and 10%, respectively [15]. This same study also revealed that intra-operative rupture of the tumor was not associated with recurrence however, positive margins and multi-centricity were [15]. The authors consequently question whether "recurrence" is related to sub-clinical or unrecognized multi-centric disease [15].

With respect to management, some advocate for conservative management once Warthin tumor has been confirmed by FNA cytology [16]. When surgery is elected, some researchers recommend local excision be limited to patients with solitary tumors and that multi-centric tumors be addressed with superficial parotidectomy [15]. Because these tumors have a less than a 1% chance of malignant transformation, once recurrent disease is confirmed with FNA cytology, surgery can be offered, however, conservative management with observation is not necessarily contraindicated [16].

Conclusion:-

At first blush, the terms "recurrent" and "multi-centric" might suggest that Warthin tumor is a malignant, neoplastic lesion; however, its true nature, as reflected by its behavior and the polyclonality of it cellular components [2,3], is best characterized as benign. A positive history of smoking is found in greater than 85% of cases [4]. Most patients present with a solitary, painless mass in the peri-parotid area. Radiologic evaluation, whether it be with ultrasound, CT scans, or MRI, reveal well-demarcated, heterogeneous lesions [8,10,11]. A definitive diagnosis is usually rendered when FNA reveals monolayered sheets of oncocytic cells, lymphocytes, and amorphous, cystic debris [13]. Management options for solitary lesions include conservative management with observation [16] or resection with either local excision or parotidectomy [15]. Multi-centric and bilateral disease is not uncommon but it is often sub-clinical and not easily detected by palpation or imaging [15]. With respect to histogenesis, it is believed that this tumor results from the proliferation of salivary gland ductal cells that were trapped in intra- and peri-parotid lymph nodes during embryonal life [5]. The multi-centric and extra-parotid potential of this tumor is similarly attributed to the embryonal heterotopia of salivary tissue [5]. Multi-centricity has been associated with increased recurrence [15]. Consequently, it appears that "recurrence" may be related to sub-clinical or unrecognized multi-centric disease

Conflict of Interests

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