International Journal of Medical Science and Research
www.medicaljournals.in
Online ISSN: 2664-8768, Print ISSN: 2664-875X
Received: 01-10-2022, Accepted: 17-10-2022, Published: 03-11-2022
Volume 4, Issue 2, 2022, Page No. 81-84

Warthin Tumor

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Abstract
Warthin tumor is the second most common benign tumor of salivary glands after pleomorphic adenoma. Warthin tumor is most commonly found in parotid gland, which can be unilateral or bilateral. This tumor is more common found in men, in the fifth or sixth decade of life. Smoking habits and radiation exposure are predisposing factors that play a role in the occurrence of Warthin tumor.
Here, we present a case of a 63-year-old man with Warthin tumor in his parotid gland with long smoking habit as a predisposing factor. The patient underwent superficial parotidectomy. In this case, no postoperative complications were found. This case is presented to provide information in the management of Warthin tumor.

Keywords: warthin tumor, smoking, radiation, parotidectomy, parotid

Introduction
Warthin tumor is the second most common benign tumor of salivary glands after pleomorphic adenoma. These tumors are more common found in men, in the fifth or sixth decade of life. Smoking habits and radiation exposure are predisposing factors that play a role in the occurrence of this tumor [1-5].
Although it can affect other salivary glands, Warthin tumor is most often affected the parotid gland (84%) [6] Other studies show that 5-14% of parotid gland neoplasia and 2-5% submandibular salivary gland neoplasia are Warthin tumors [1, 4, 7].
Fine Needle Aspiration Cytology or FNAC is a relatively simple, safe, accurate and inexpensive method for diagnosing salivary gland neoplasms [8, 9].With sensitivity ranging from 85.5-99% and specificity ranging from 96.3-100%, FNAC is quite reliable in distinguishing malignant and benign salivary gland tumors.
In general, the management of Warthin tumor is excision of the tumor by parotidectomy with facial nerve preservation or enucleation [2, 3, 6, 7]. Most of Warthin tumors are located in the superficial lobe of parotid gland, so superficial parotidectomy is usually sufficient [6].
We report a case of Warthin tumor in parotid gland which occurred in a 63-year-old man with a predisposing factor of a long smoking habit. The patient underwent superficial parotidectomy. In this case, no postoperative complications were found. This case is presented to provide more information about the management of Warthin tumor.

Case Report
A 63 years old male patient, who lived in Badung as a merchant, came to the ENT Polyclinic Sanglah Hospital on January 9, 2019 with chief complaint a lump under his left ear since 6 months ago. The lump was initially small but since the last 2 months it has been getting larger. Patient felt no fever and no pain. Lump in other body parts was denied, and patient had no history of toothache. The patient also said that there was no history of hearing loss, nose bleeding, double vision or headaches. There was no history of other diseases such as heart disease, asthma, high blood pressure, diabetes mellitus, malignancy and allergies. The patient had a smoking habit for more than 40 years, approximately 1 pack (12-16 cigarettes) per day but has stopped since approximately 3 years ago. History of exposure to radioactive radiation was denied.
On physical examination, general status of patient was good, did not appear to be seriously ill, patient was compos mentis with blood pressure 120/70 mmHg, pulse 70 beats per minute, respiration 16 times per minute, axillary temperature 36oC. Ears, nose and throat examination were found within normal limits. On endoscopic examination of the nasopharynx, no lumps, mucosal thickening or other abnormalities were found. There was a lump under the patient's left ear in size of 5x3x3 cm. The tumor was well-defined in palpation, flat surface, spongy consistency, and not fixed with the surrounding tissue. The patient was diagnosed with a left parotid tumor.
On January 10, 2019, a Fine Needle Aspiration Cytology (FNAC) examination was carried out, and we found hypercellular smears consisted of distributions and groups of neoplastic cells forming solid and papillary structures. The cell morphology was polygonal, relatively monotonous with broad cytoplasm, round-oval nucleus, regular nuclear membrane, and fine chromatin. There was a distribution of lymphocytes with various degrees of maturation ranging from blast to mature lymphocytes and macrophages.

The cytomorphological features suggest a Warthin tumor. Based on FNAC results, the patient was diagnosed with a Warthin tumor of left parotid. Furthermore, a computer tomography examination was carried out on January 18, 2019 and it was found that the left parotid gland was enlarged in size ± 2.7 x 2.2 cm. The lymph nodes in the right and left subcentimeter colli regions was also enlarged. Heart and lungs were within normal limit in chest X-ray. Complete blood count, blood chemistry and haemostasis function were also still within normal limits. Examination of the facial nerve showed a good facial nerve function. The patient was then planned to undergo left superficial parotidectomy surgery.

In order to preparation of surgery, the patient was consulted to Anaesthesiology and Cardiology divisions. The patient was in ASA II physical status with controlled hypertension, no contraindications for surgery and agreed to undergo the procedure under general anaesthesia. The next surgery is planned on March 14, 2019.

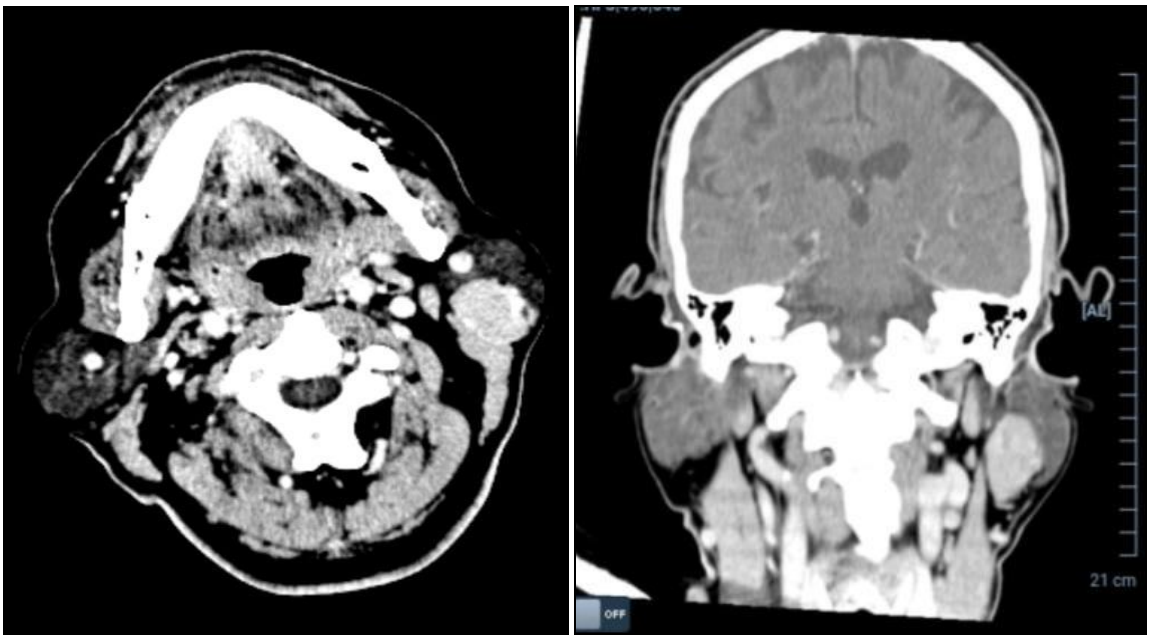

Fig 1: Computed tomography showed a mass on left parotid

On March 14, 2019, a left superficial parotidectomy was performed. The skin incision is made using a modified Blair's incision. After the skin flap had been removed, the main branch of the facial nerve was identified antegradely. The tumor was located in superficial part of facial nerve. The tumor is oval in shape, brown in colour, measuring 4x3x3cm with a spongy consistency. The tumor mass was then sent to the Anatomical Pathology Laboratory for histopathological examination. During the operation there was no significant bleeding (50 cc). Subsequently, a drain was applied and surgical wound was sutured. After procedure, facial nerve function was examined and found to be within normal limits. The patient was then given intravenous drip and morphine drip 10 micrograms/kgBW/hour from anaesthesiologist, and ceftriaxone antibiotic 1 gram every 12 hours intravenously.

On the first day after surgery (March 15, 2019), patient had no subjective complaint, physical examination was within normal limit, surgery wound was well-maintained, with minimal blood on his drain. On the second day after surgery, drain was removed and the patient was discharged. Patient was given 200 mg cefixime twice daily and paracetamol 500 mg thrice daily to bring home.

On March 19, 2019, patient came to ENT Polyclinic Sanglah Hospital for follow up. The surgical wound was well-maintained with no signs of secondary infection, then some surgical sutures were removed. Patient came again on March 22, 2019 and all surgical sutures were removed. The results of histopathological examination of tumor tissue showed that the tumor mass was well demarcated forming a papillary cystic structure with some of lymphoid stroma forming a germinal centre. The epithelial component consists of 2 layers of cells, namely luminal cells in the form of columnar, partially oncocytic, which are arranged in palisade with an ovoid round nucleus and basal cells which are flat and cuboidal in shape. The cystic space appears to contain eosinophilic amorphous material, inflammatory cells, lymphocytes and plasma cells. Histopathological features are consistent with Warthin tumor.

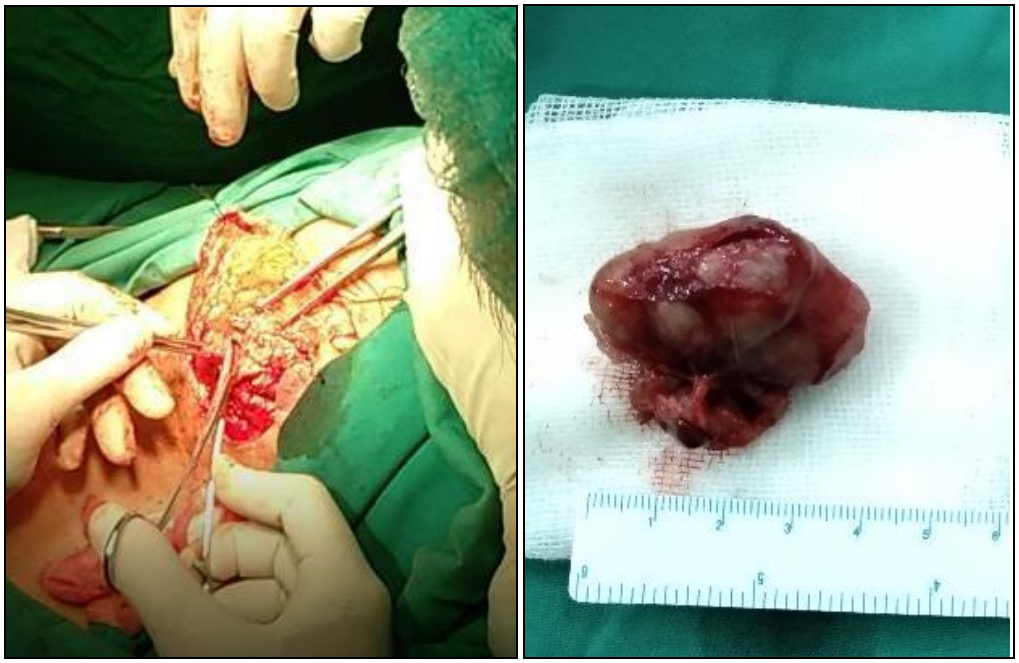

Fig 2: Tumor in left parotid superficial lobe and parotid tumor mass after removal

Discussion

Papillary cyst adenolymphoma or commonly known as Warthin tumor is the second most common benign cystic tumor of the salivary glands after pleomorphic adenoma [2, 4, 6, 10].

Batori said that 5-14% of parotid gland neoplasia and 2-5% submandibular salivary gland neoplasia are Warthin tumors [1, 2].

Warthin tumor occurred in the superficial lobe of parotid gland of our patient. It is consistent with Faur A *et al* who said that Warthin tumor almost always occur frequently in the parotid gland, especially in superficial lobe and very rarely in profunda lobe [4].

In this case, Warthin tumor was reported in a 63-year-old man who had a smoking habit for about 40 years. This is in accordance with a study by Kowall, Kuzenko DV, Lee DH, which stated that Warthin tumor often appears in the fifth or sixth decade of life and is found to occur more often in men than women with a ratio of 6:1 [1, 4, 6]. This is also consistent with the study of Klussmann JP in 203 cases of Warthin's tumor in which 89% were smokers and 69% of them were heavy smokers.[12] Besides, radiation is also said to be one of the determining factors in the occurrence of Warthin's tumor, as mentioned by Oh and Kuzenko [1, 11]. However, there was no history of radiation exposure in this case.

Kuzenko *et al* stated that the clinical symptom of Warthin tumor that most often complained by patient was the presence of a lump in the parotid region that grew slowly and painless [1]. Lee YH showed that most (63%) cases had unilateral lesions, and 65% of the tumors were in the superficial lobe [3]. In our case, the lump was unilateral, located under the left ear, which had been grown within 6 months and there was no pain.

FNAC is a relatively simple, safe, accurate and inexpensive way to diagnose salivary gland neoplasms. According to Oh YS and Cawson RA, sensitivity ranges from 85.5-99% and specificity ranges from 96.3-100%, FNAC is quite reliable in differentiating malignant and benign salivary gland tumors. However, the accuracy of FNAC is also highly dependent on the experience of the cytopathologist [8, 9]. Open biopsy of parotid gland tumors should be avoided because of the risk of injury to the facial nerve [8, 9]. In this case, to make a working diagnosis, FNAC examination was performed, which showed hypercellular smears consisting of distribution and groups of neoplastic cells forming a solid and papillary structures. The cell morphology is polygonal, relatively monotonous, with broad cytoplasm, round-oval nucleus, regular nuclear membrane, and fine chromatin. There was a distribution of lymphocytes with various degrees of maturation ranging from blast to mature lymphocytes and macrophages. The cytomorphological features suggest a Warthin tumor. This result was confirmed by postoperative biopsy of the tumor mass, which showed that the tumor mass was well demarcated forming a papillary cystic structure with the lymphoid stroma partially appearing to form a germinal centre. The epithelial component consists of 2 layers of cells, namely luminal cells in the form of columnar, partially oncocytic, which are arranged in palisade with an ovoid round nucleus and basal cells which are flat and cuboidal in shape. The cystic space appears to contain eosinophilic amorphous material, inflammatory cells, lymphocytes and plasma cells. Histopathological features are consistent with Warthin tumor.

Our patient underwent left superficial parotidectomy, considering that Warthin tumor in this case was located in superficial lobe. This is in accordance with what was mentioned by Lee DH, Oh, and Cawson [4, 8, 9]. The skin incision performed during surgery was the modified Blair's incision. It is the most often incision chosen in parotidectomy surgery, considering that the operating field obtained with this incision is wide enough to facilitate the operation [8].

In one study, Lee DH found that 10.1% of patients underwent parotidectomy had partial facial nerve dysfunction that improved within 3 months and the location of the tumor in the deep lobe increased the risk of these complications.

Conclusion

Warthin's tumor is the second most common benign tumor of salivary glands after pleomorphic adenoma and most commonly found in the parotid gland, which can be unilateral or bilateral. These tumors are more common in men, in the fifth or sixth decade of life. Smoking habits and radiation exposure are predisposing factors that play a role in the occurrence of this tumor.

The diagnosis of Warthin tumor is made through anamnesis and physical examination, the patient usually complains of a lump in the parotid area, bilateral or unilateral, slow growing, supple, solid, movable, painless, and there is a history of smoking or exposure to radiation. FNAC is a relatively simple, safe, accurate and inexpensive way to diagnose salivary gland neoplasms. However, its accuracy is also highly dependent on the experience of the cytopathologist. Open biopsy of parotid gland tumors should be avoided because of the risk of facial nerve injury.

Management for Warthin tumor is parotidectomy surgery with facial nerve preservation. The prognosis for Warthin tumor after surgery is generally good. Complications that can occur postoperatively are facial nerve paralysis and Frey's syndrome. The prognosis of this disease is commonly good if it is early detected and does not spread to the intracranial cavity.

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