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The Bizarre Case of Warthins Tumor in Submandibular Gland-Case Report

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Abstract:- The second most frequent salivary gland tumour after pleomorphic adenoma, Warthin's tumour is also known as adenolymphoma, cystadenolymphoma, and papillary cystadenoma lymphomatosum. It often develops from the parotid gland and makes up between 4% and 13% of all salivary gland tumours. With relative rates of 6.9% and8%, extraparotid Warthin tumours from the submandibular gland and cervical lymph nodes are extremely uncommon. Moreover, Warthins tumours that originate in the minor salivary gland are incredibly uncommon, with documented incidences ranging from 0.1% to 1.2%. Several cysts with numerous papillae are visible in Warthin's tumour, which is covered in bilayered columnar and basaloid oncocytic epithelium. They typically appear as slowly expanding, painless masses that emerge from the parotid gland's tail, although they can also infrequently appear as quickly expanding, painful lesions. We describe a rare instance of a 6-month-old asymptomatic Warthin's tumour in a 55-year-old male patient's left submandibular gland. Warthins tumour, extra parotid, and submandibular gland are important terms.

I. INTRODUCTION

Albrecht and Arzt published the first description of Warthin's tumour in 1910. Aldred Scott, a pathologist from USA. The first two case reports were published by Warthin in 1929 under the name papillary cystadenoma lymphomatosum, which was thought to be an auxiliary eustachian tube analage mucous membrane heterotopia. [1] Warthin tumour was later used to distinguish it from malignant lymphomas. This tumour, which makes up 15% of parotid gland epithelial tumours [2], affects people between the ages of 13 and 85, primarily in their sixth and seventh decades, with a preference for men (4:1 male to female ratio). [3] Only two theories of development finally stand out among the many others. First, there is the concept of heterotopia, which has already been addressed, and second, there is the theory that this tumour is an adenoma with concurrent lymphocytic infiltration. [2] This tumour can occasionally have a tendency to encompass both sides, and it can also arise in the lateral neck, palate, upper lip, nasopharynx, submandibular glands, sinuses, and lacrimal glands. [3] Less than 2% of cases return, and there is only a 1% chance that the tumour may turn malignant. Tracking the lesion after excision is crucial. [4]

II. CASE REPORT

A 55-year-old patient presented to the Bmaxfac clinic in October 2018 with a single large swelling in the left submandibular region. He had a painless tiny mass below 1cm in the submandibular region with a gradual progression during one year, patient has no relevant family and medical history, on examination the swelling was approximately 3.5*3 cms in size freely movable without any fixation to overlying structures, non-tender and doughy in consistency with no lymphadenopathy on palpation, intraorally no relevant dental findings were observed. There is no paraesthesia or discharge, associated fever, or change in the salivaryflow.

Ultrasonography revealed a well defined thin-walled homogenous hypoechoic lesion measuring 3.5*3 cms in the left submandibular region. Aspirationcytologyyieldedblood-tingedpusstudiedundermicroscoperevealed epithelial cells arranged in flat, monolayered sheets and the epithelial cells were with abundant and densely eosinophilic, granular cytoplasm with rounduniform bland nuclei; in the background, there is a mixed population of reactive lymphocytes, with abundant cellular debris along with single, Cytological diagnosis of Warthin's tumor was rendered.

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Correlating the above findings, a provisional diagnosis of warthins tumor of the left submandibular gland was made and surgical excision of the lump was advised.

The patient was informed about the surgical procedure of surgical excision under general anesthesia and was also informed about the chances of negligible post-operative scar in the left submandibular region postoperatively.

Thereafter under general anesthesia and aseptic conditions, a Risdon/ submandibular incision is made, layer by layer dissection was done, the encapsulated tumor was completely removed three-dimensionally taking care no remnants of the lesion left behind at operated site by using blunt dissection and external traction protecting anatomical structures. The wound is closed in layers from inside to outside with absorbable and non-absorbable sutures, respectively and the specimen was sent for histopathologicalexamination.

Post-operative recovery was uneventful and patient was discharged on 3rd day with oral antibiotics and necessary medications.

Microscopic examination shows lymphoid aggregates with the focal germinal center formation with overlying stratified epithelium and also a large areaofcysticdegenerationandcholesterolcleftformation,featuressuggestiveof lymphoepithelioma (warthins tumor).

The post-operative review of the thirdand sixthmonth until two years showed no signs of recurrence.

III. DISCUSSION

The term ‘Warthin’s tumor was first applied in 1944 by Martin and Ehrlich.

[5]HildebradfirstreportedWarthin’stumorin1895asavariant of a lateral cervical cyst.

AlbrechtandArizin1910firstproposedtheheterotropictheoryof an origin believed to be from neoplastic proliferation of salivary gland ducts present within intraparotid or paraparotid nodes. [6] This theory is widely accepted and even was supported by immunohistochemistry analysis (IHC). [7]Warthins described it as a very rare teratoid of the parotid gland region, representing a heterotopiaordystopiaofpharyngealendoderm,resemblinginthestructuremost closely to the mucosa of the cartilaginous portion of the Eustachian tube. [8]

The male to female ratio in this tumor is 10:1.[9]The association between smoking and Warthin’s tumor has been reported since the 1980s[10]. Salivary glands are relatively immune to tuberculosis because of thiocyanate ions and proteolytic enzymes like lysozymes, which impart antibacterial propertyWatanabe has reported two cases of tuberculosis with warthintumor one in parotid gland in a 75yr old female and other in a submandibular gland in a 78 yr old male.[11]

99.2% of WarthinsTumor were located in theparotid gland and only 0.8% in the submaxillary gland. [12] Warthin's tumor most commonly presentsasa slow-growing,asymptomaticmassinthetailoftheparotidgland. Only 10% of patients present with symptoms such as pain and pressure. As the lesion is asymptomatic, it takes several months to makeout diagnosis. Sialography, ultrasound, and radionuclide scanning have all been used with varying degrees of success. [13]On aspiration,The fluid from the tumor ranges from 3 to 4 ml and smears show clustered and dissociated oncocytes with lymphocytes.[14]Histologically they show lymphoid aggregate in focal germinal center formation with overlying stratified epitheliuma long with large areas of cysticde generation and cholesterol cleft formation. Treatment of warthins tumor remains controversial.It is difficult to diagnosis Warthin tumor from submandibular gland because of its rarity and may get confused with those from the anterior tail of the parotid or from periparotid lymph nodes.[15]The optimal treatment of Warthin’s tumor is surgical removal, which is easy due to the superficial location of the tumor. Recurrence of unilateral Warthin's tumors after excision has been noted.[16]Multifocal origin predisposes to "recurrence" if too conservative removal is attempted. A long period follow up is required to early detect the possibility of recurrence.Some malignant cases have been reported, in relation to both the epithelial and the lymphoid component. [17]

IV. CONCLUSION

Rarely occurring in the submandibular region, Warthins tumours can be diagnosed using techniques like FNAC and correlation with clinical, histological, and radiological markers.The optimal treatment option for wartin’s tumor is surgical excision.

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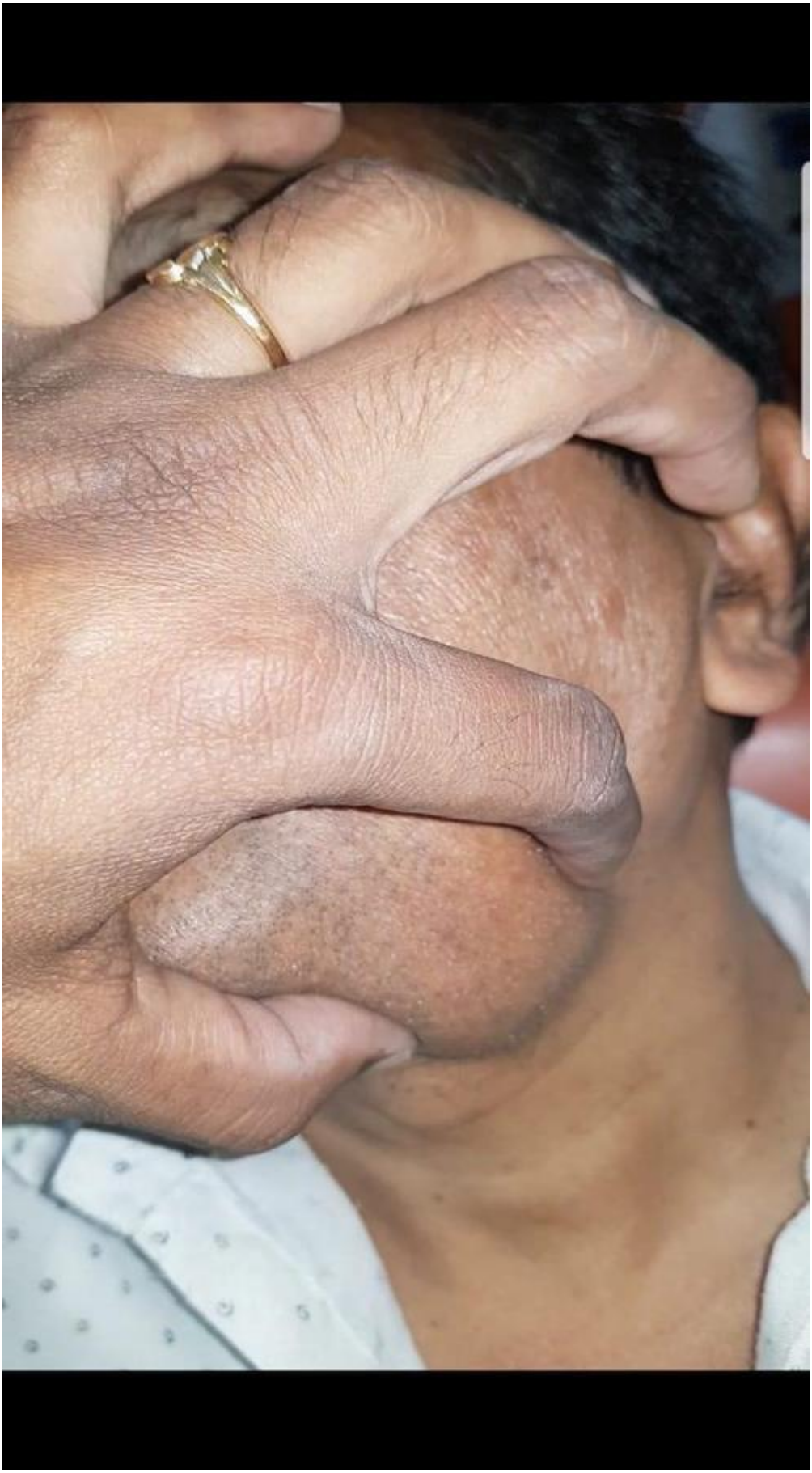

Picture 1 – Pre Operative Swelling

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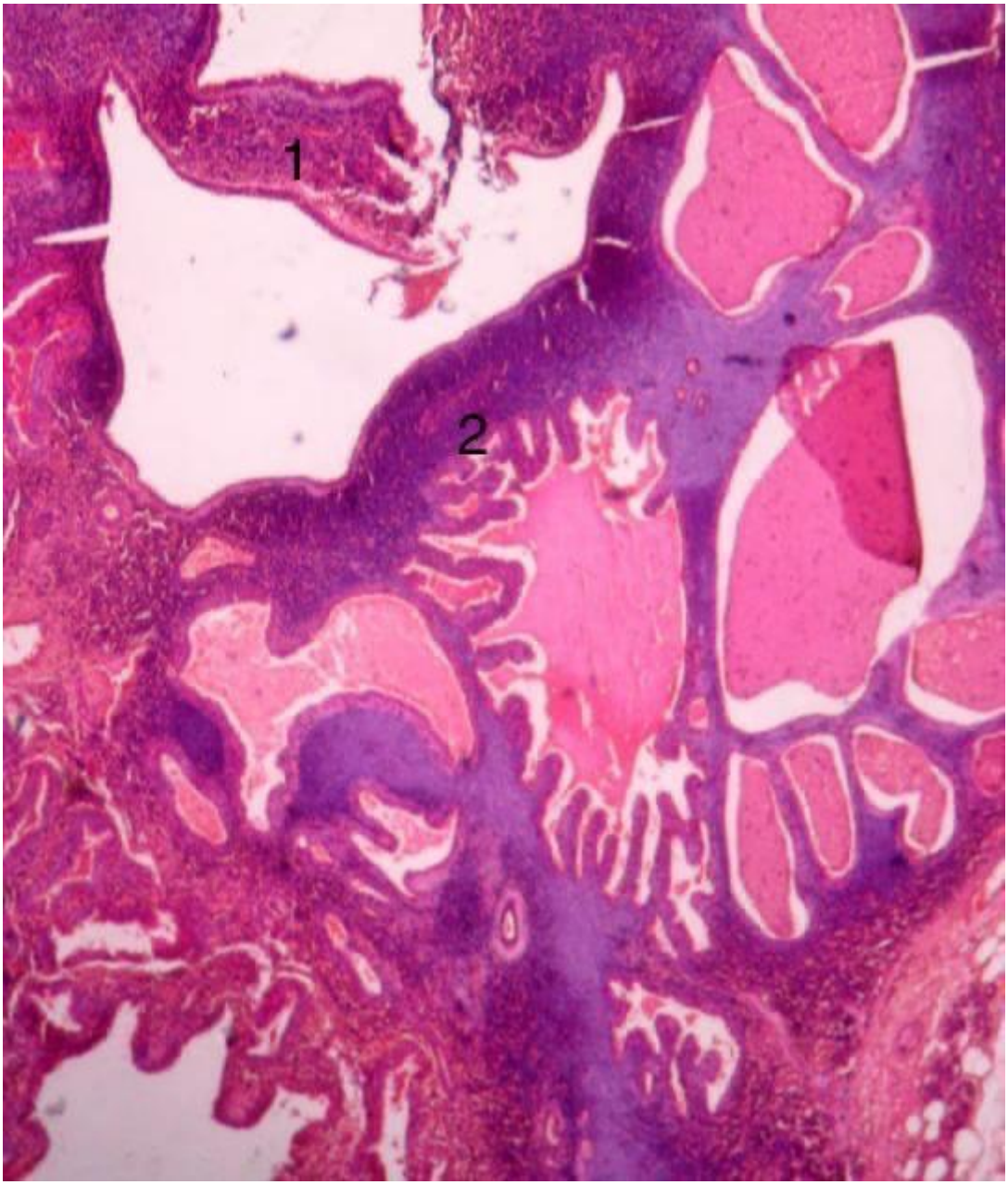

Picture 2-Histopathology of the swelling – Lymphocytes present with double layered lining epithelium inner columnar and outer cuboidal cells

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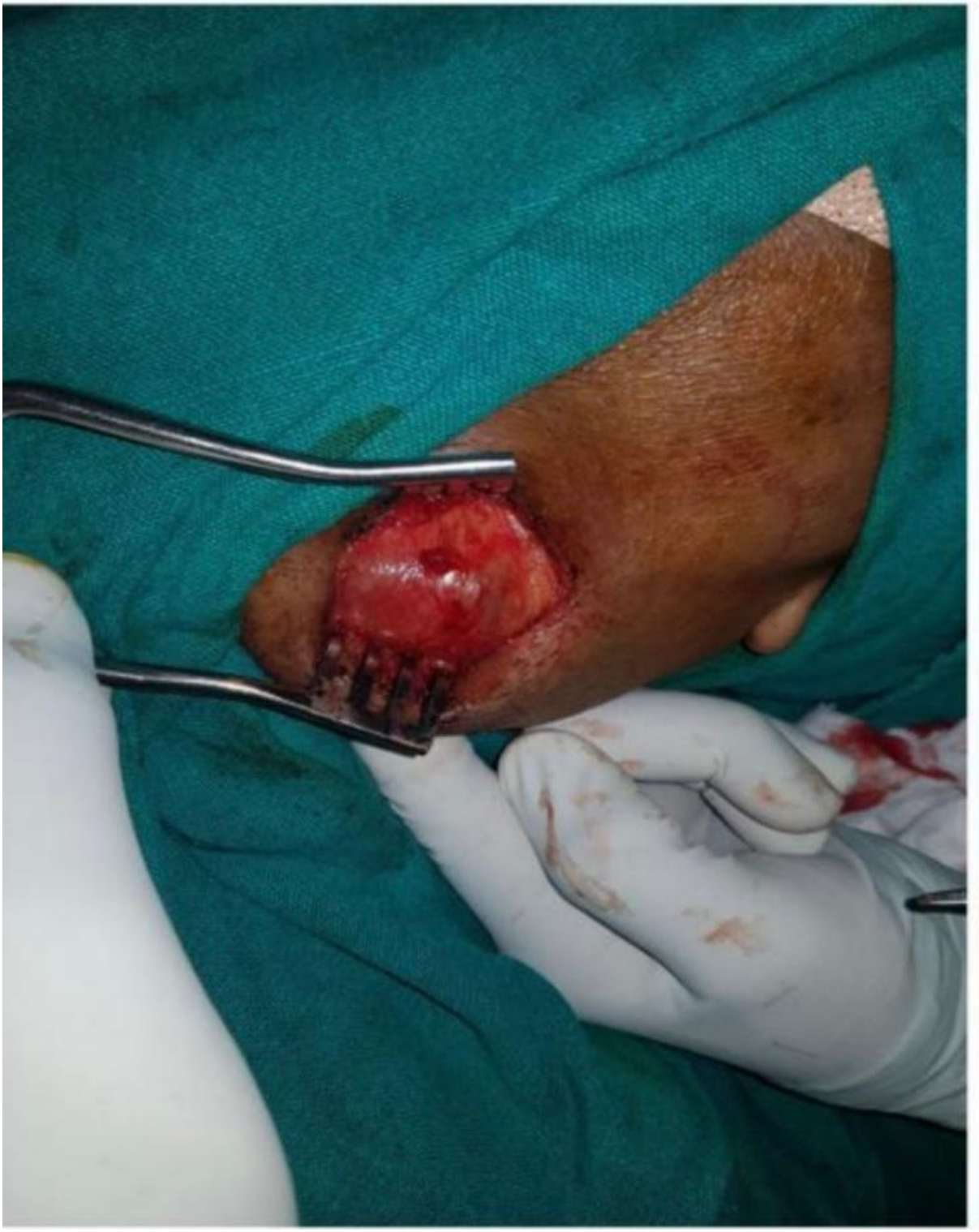

Picture 3- Intraoperatice Swelling Exposure

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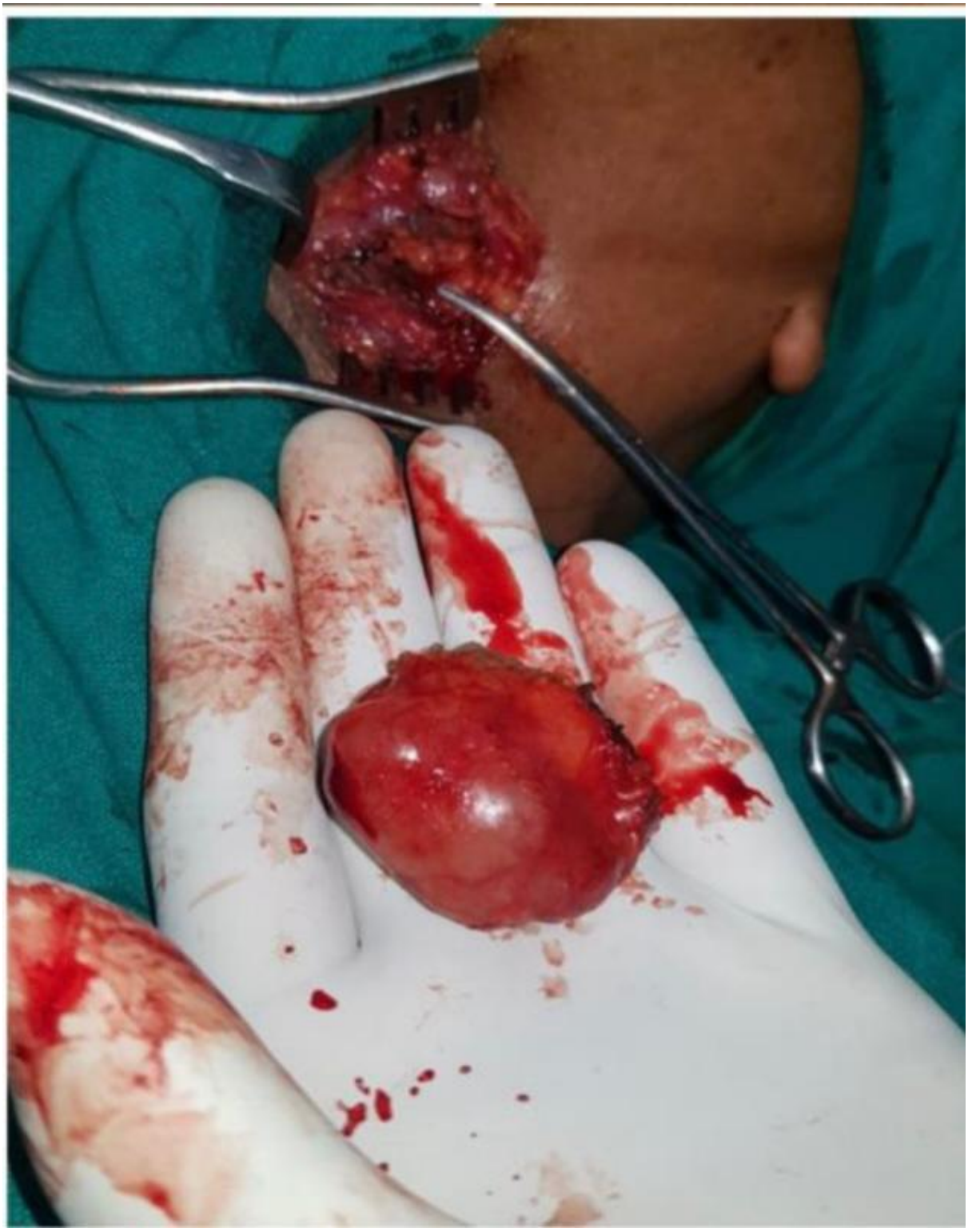

Picture 4- Intraoperative Excised Tumor

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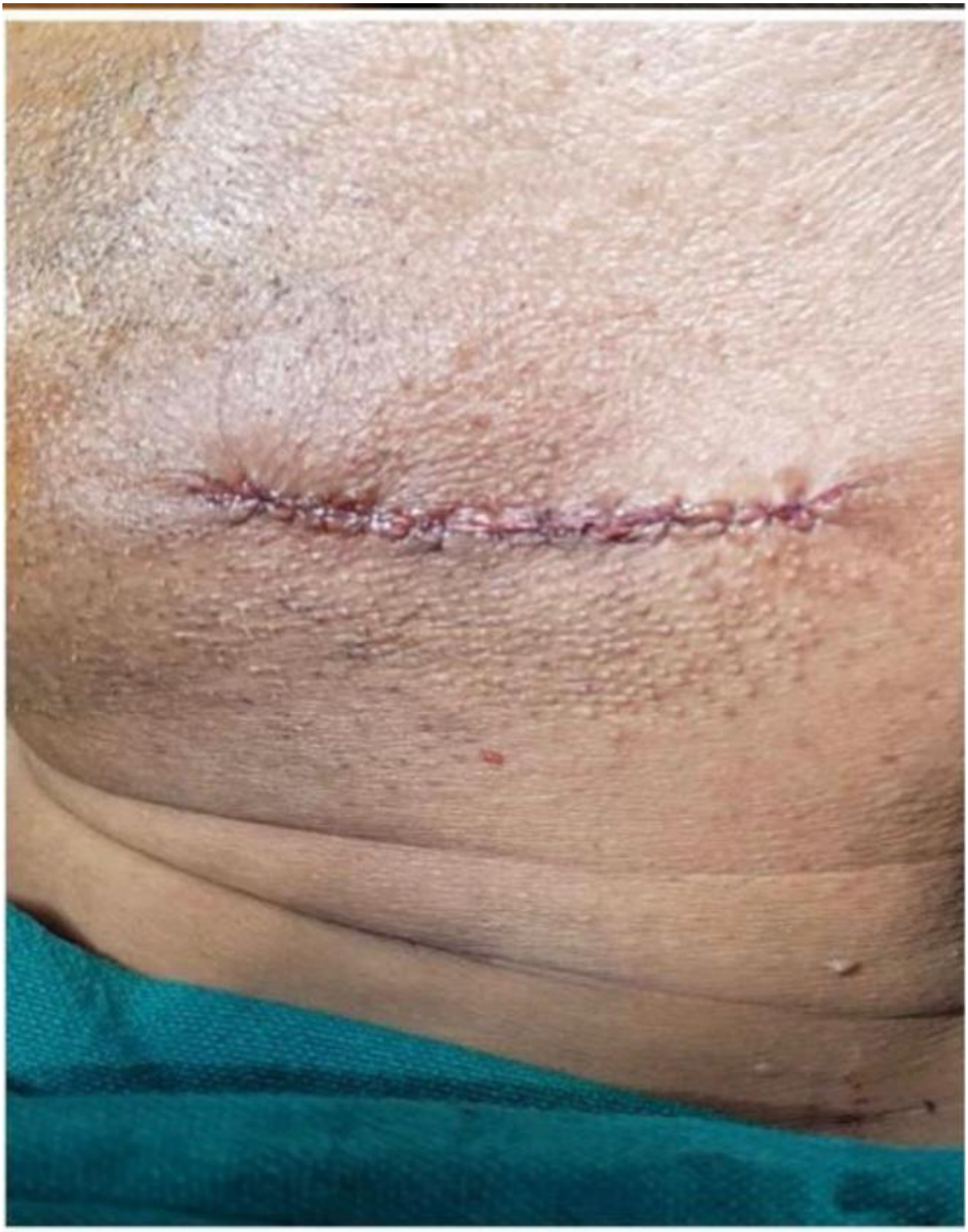

Picture 5- Wound Closure

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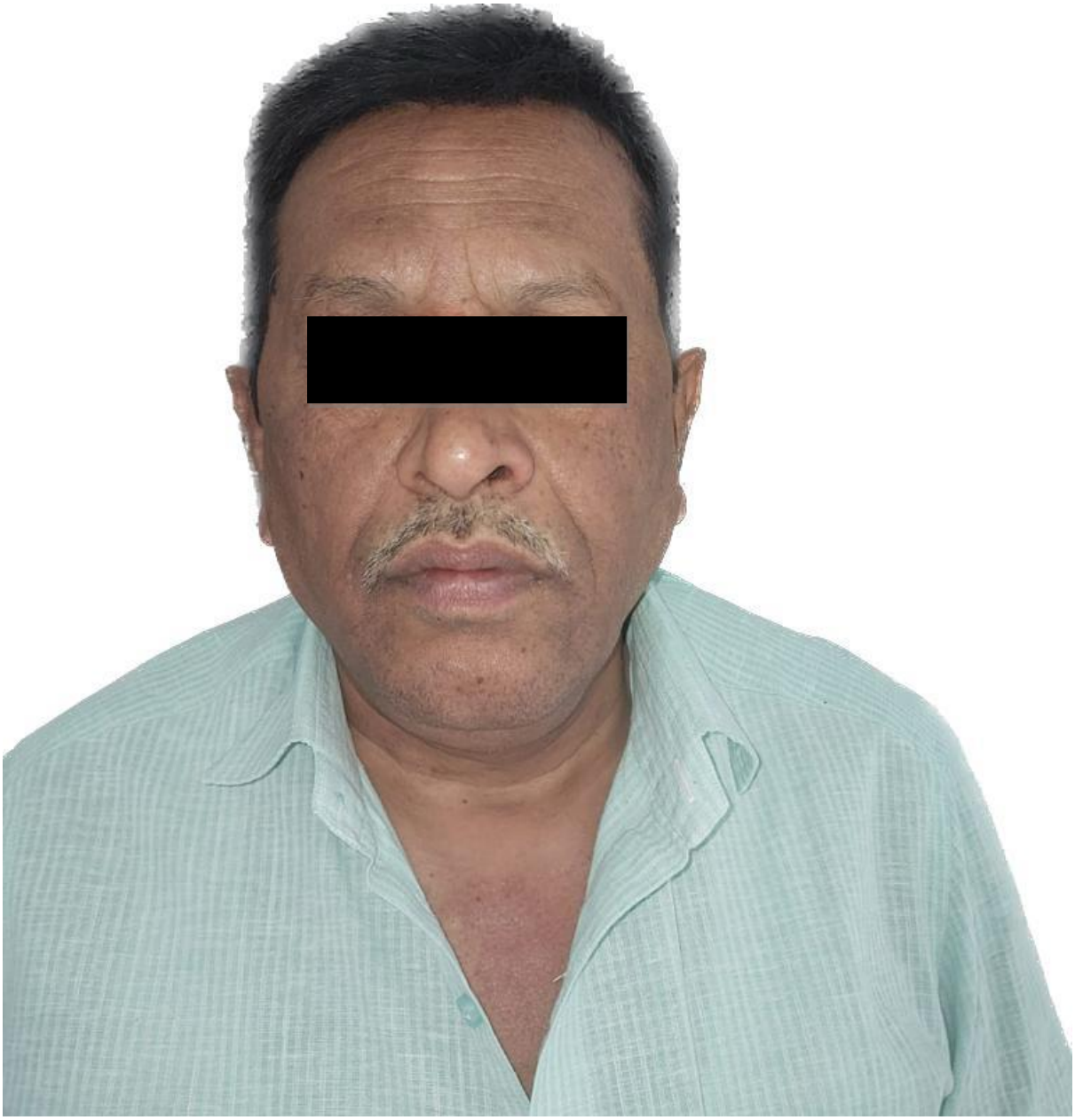

Picture 6-Post Operative Follow Up After 6 Years