

## Case Report

# Malignant appearance of trichilemmal cyst: a case report with review of the literature

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### ABSTRACT

69 years old male presented with one month history of non-tender, nodular, pinkish left neck swelling, measuring 10×20 mm. CECT revealed left neck dermatofibrosarcoma protuberans with submandibular vascular malformation. Surgical wide local excision of the lesion and split skin graft was done. Histopathology confirmed trichilemmal cyst (TC). TC is adnexal lesions, found in hair bearing areas. Pinkus pointed out that TC are keratin-filled cysts with a wall resembling the external root sheath of hair follicle. 2% can develop into proliferating trichilemmal tumours (PTTs) and malignant proliferating trichilemmal tumours (MPTTs). PTTs were originally described by Jones as benign but locally aggressive skin neoplasms. Female to male preponderance of 6% to 1%, with average age of 65 years at presentation. 90% are located on the scalp, while the residual 10% occur mainly on the back but they can develop any part of the body such as submandibular region. Proliferating TCs present as lobulated masses, soft in consistency. The masses may become exophytic, ulceration and may show malignant transformation. On imaging, these lesions can be either cystic or solid mass. Histopathological, it is characterised by absence of intercellular bridges between epithelial cells lining the cyst wall. Cyst cavity contains amorphous eosinophilic keratin. Satyaprakash et al report, complete surgical excision is warranted for TCs as if there is proliferative or malignant potential, it would affect the postoperative management and overall prognosis. TC, locally aggressive skin neoplasms and rarely, occurrence at submandibular region or become malignant and appropriately managed to prevent recurrence.

**Keywords:** Submandibular cysts, Trichilemmal cysts, Adnexal tumours, Malignant

### INTRODUCTION

TC are benign adnexal tumours usually occurring in hair bearing areas such as scalp, groin, thigh especially in elderly women. They arise from the outer root sheath of the hair follicle. The cyst contains keratin and is outlined by stratified squamous epithelium. TCs may be inherited as an autosomal dominant trait, there can be familial predisposition with patients presenting at younger age and with multiple lesions. Occurrence of trichilemmal cyst at submandibular region has been rarely reported.<sup>3</sup>

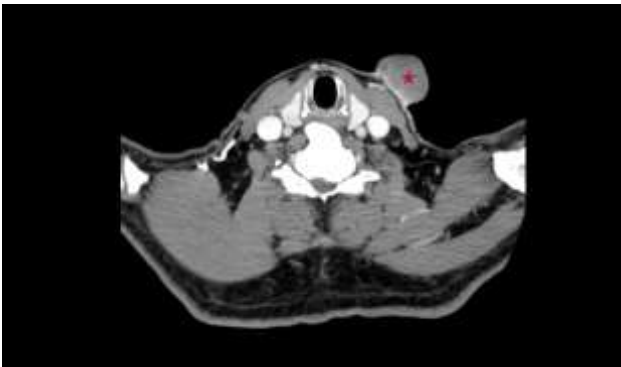
### CASE REPORT

A 69-year-old, male patient presented to the surgical outpatient department with a complaint of swelling over left neck for 1 month. He had underlying comorbid conditions such as hypertension, diabetes mellitus and atrial fibrillation on warfarin. In addition, he had a strong family history of cancer; mother known case of neck cancer and brother known case of nose cancer. The swelling was gradually progressive in size and bleeding. On examination, it was 1×2 cm in size, nodular in

appearance, pinkish in colour, non-tender involving the left neck. A diagnosis of left neck keratoacanthoma was made.



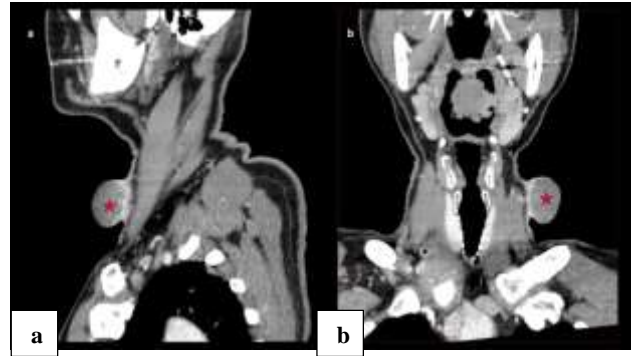
**Figure 1: Appearance of the lesion.**



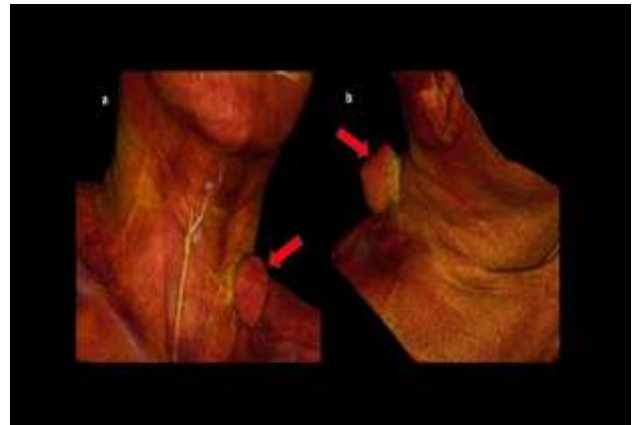
**Figure 2: Contrast enhanced CT in axial plane shows solitary well defined enhancing lesion over left anterior neck, arising from skin with associated skin thickening and subcutaneous fat stranding; no calcification or necrotic component within the lesion.**

We have proceeded with contrast enhanced computed tomography (CECT) of neck and thorax which showed well defined enhancing round lesion over left anterior neck, measuring 2.7×3.4×3.9 cm. Lesion was seen arising from the skin at C6-C7 level with associated skin thickening and subcutaneous fat stranding (Figure 1-3). No extension to overlying subcutaneous planes. No calcification or necrotic component within the lesion. Several enlarged level III nodes with loss of fatty hilum were also seen.

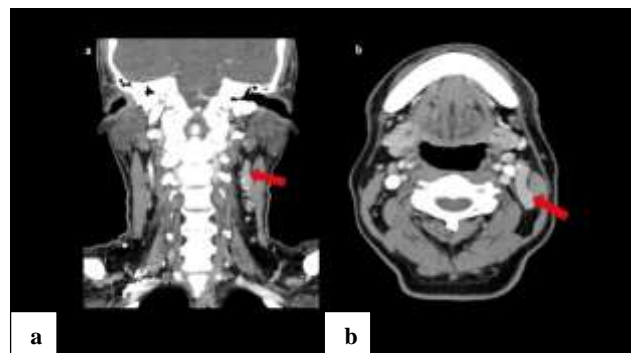
A tubular left submandibular enhancing lesion seen insinuating lateral to left carotid space and medial to left sternocleidomastoid muscle. No prominent artery seen adjacent to the lesion, however a prominent vein within the lesion seen draining into the left jugular vein (Figure 5). A diagnosis of left neck level III (DFSP) dermatofibrosarcoma protuberans with left submandibular vascular malformation was made following imaging.



**Figure 3 (a and b): Contrast enhanced CT in sagittal and coronal plane shows solitary well-defined enhancing lesion over left anterior neck, arising from skin with associated skin thickening and subcutaneous fat stranding; no calcification or necrotic component within the lesion.**



**Figure 4: Volume rendering images of the neck region shows a solitary well defined exophytic lesion over left anterior neck.**



**Figure 5 (a and b): Contrast enhanced CT in coronal and axial plane showing tubular enhancing lesion at left submandibular region insinuating lateral to left carotid space and medial to left sternocleidomastoid muscle; no prominent artery seen adjacent to the lesion, however a prominent vein within the lesion seen draining into the left jugular vein; focus of calcification seen in the anteromedial aspect of the lesion; clear fat plane seen between the lesion and adjacent left SCM.**

Surgical wide local excision of the swelling and split skin graft was done under general anaesthesia. Intra operative finding was pedunculated mass at anterior triangle of the left side of the neck, size 6×4 cm (base), 6 cm (height). The specimen was sent for histopathological examination.

Report was suggestive of a skin tissue measuring 4.5×3×0.5 cm and the greyish sessile polypoidal lesion measuring 5×4.5×4.5 cm. The intradermal lesion was composed of a cyst lined by stratified squamous epithelium with no granular layer containing keratinous material. Mild lymphoplasmacytic infiltration was seen around and a fragment of soft tissue with crush artifacts. No evidence of malignancy seen. A final diagnosis of TC was made.

## DISCUSSION

Trichilemmal (pilar) cysts and the commoner epidermoid (sebaceous) cysts are the two types of cutaneous cysts found in hair bearing area that arise from different parts of hair follicle unit, with a prevalence ratio of 1:4.<sup>6</sup> In earlier days, all cutaneous cysts were erroneously referred to as sebaceous cysts until Pinkus pointed out in 1969 that TC are keratin-filled cysts with a wall resembling external root sheath of a hair follicle, whereas sebaceous cysts arise from follicular infundibulum and hence are distinct entities.<sup>7</sup> TC are common intradermal or subcutaneous cysts, occur in 5-10% of the population, with 2% developing into PTTs and MPTTs are even rarer.<sup>3</sup> PTTs were originally described by Jones in 1966 as benign but locally aggressive skin neoplasms arising from the outer root sheath of hair follicles.<sup>1</sup> They usually develop in pre-existing TCs and present as rapid growth from previously singular, asymptomatic lesions.<sup>2</sup> Very rarely, they may develop into MPTTs.<sup>2</sup>

They have a female to male preponderance of 6% to 1%, with average age of 65 years at presentation and may have autosomal-dominant inheritance.<sup>3</sup> 90% are located on the scalp with increased density of hair follicles and chronic sun exposure, while the residual 10% occur mainly on the back.<sup>3</sup> They can develop anywhere on the body; other possible sites include face and neck such as the submandibular region. Most people end up having more than one TCs at any given time.

Proliferating TCs present as lobulated masses. They are usually soft in consistency and vary considerably in size from a few millimetres to large masses many centimetres in diameter. Occasionally the masses may become exophytic and may be associated with ulceration and may show malignant transformation.<sup>4</sup>

On imaging, these lesions can be either a cystic or solid mass. Histopathological, it was characterised by the absence of intercellular bridges between the epithelial

cells lining the cyst wall. The peripheral layers demonstrate a palisading arrangement, whereas cells close to the cyst cavities are swollen and filled with pale cytoplasm. The cyst cavity contains amorphous eosinophilic keratin. Foci of calcifications within the keratin occur in approximately 25% of cases.<sup>5</sup>

Satyaprakash et al reported complete surgical excision was warranted for TCs. It would be necessary to confirm, if there is a proliferative or malignant potential if it turns out to be a trichilemmal cyst, as it would affect the postoperative management and overall prognosis.<sup>2</sup>

## CONCLUSION

TC are benign but locally aggressive skin neoplasms arising from hair follicles, rarely, they can become malignant. Occurrence of TC at submandibular region is rare and must be appropriately managed to prevent recurrence and metastasis. One must have a low threshold for diagnosing this rare neoplasm.

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