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Pilomatrixoma of Anterior Chest Wall – A Case Report

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Abstract:
Introduction :*Pilomatrixoma, also known as pilomatricoma or calcifying epithelioma of Malherbe, is a benign neoplasm that derives from hair follicle matrix cells. In this case report, we present a case of a male patient with pilomatrixoma arising from the chest wall.*
Case presentation :*A 71 year old gentleman presented with painless swelling over anterior aspect of the chest for the past 1 year, which was insidious in onset and progressed gradually. On examination, a single, oval, nodular, mobile, hard swelling in the anterior aspect of the midline chest was noted. Excision biopsy was done and biopsy revealed features suggestive of Pilomatrixoma. Patient recovered uneventfully.*
Discussion :*Pilomatrixomas can show malignant transformation. Pilomatrix carcinoma is extremely uncommon and has traditionally been considered as having a low malignant potential. Despite the availability of various imaging techniques, histopathological evaluation remains as the diagnostic procedure of choice and surgical excision remains as the treatment of choice.*
Conclusion :*Pilomatrixoma needs to be considered as one of the differential diagnoses of benign soft tissue swellings of skin and subcutaneous tissue.*
Keywords: *Pilomatrixoma;Calcifying epithelioma; Skin adnexal tumours; Soft tissue tumours.*

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I. Background

Pilomatrixoma, also known as pilomatricoma or calcifying epithelioma of Malherbe, is a benign neoplasm that derives from hair follicle matrix cells.(1)Pilomatrixomas are generally asymptomatic and found in the head and neck area and, upper extremities, but rarely identified on the chest, trunk, or lower extremities. They are reported to occur in people of all ages.(2)Pilomatrixoma is classified within the family of skin adnexal tumours.(3) These tumours most frequently appear in the first or second decade of life. Pilomatrixoma occurs as a solitary lesion; however, multiple lesions and familial patterns have been reported. Hereditary types have been associated with myotonic dystrophy.(4) The lesions are typically painless, slow-growing, deep-seated hard subcutaneous nodules arising most commonly within the face and upper extremities. In rare instances, pilomatrixomas can show malignant transformation. Pilomatrix carcinoma is extremely uncommon and has traditionally been considered as having a low malignant potential; however, a high local recurrence rate has been reported.(5)This article describes a case of a male patient with pilomatrixoma arising from his midline chest, and the clinical and pathologic features of this rare tumour.

II. Case presentation

A 71 year old gentleman came to General Surgery Out Patient Department with the complaints of swelling over the anterior aspect of chest for the past 1 year. The swelling was insidious in onset, started as small (approximately 0.5 x 0.5 cm), later progressed over 1 year to the current size (approximately 4 x 4 cm). The swelling was not associated with pain. No history of trauma. No history of fever, cough, hemoptysis. No history of loss of weight or loss of appetite. On examination,patient's general condition was fair. Vitals were stable. Systemic examination wasunremarkable.

Local examination :

A single, oval, nodular swelling of size 4 x 4 cm present in the anterior aspect of chest wall in the midline. The swelling was hard in consistency, no warmth, non tender, freely mobile, with well defined margins, without any punctum. The skin over the swelling was non-pinchable.

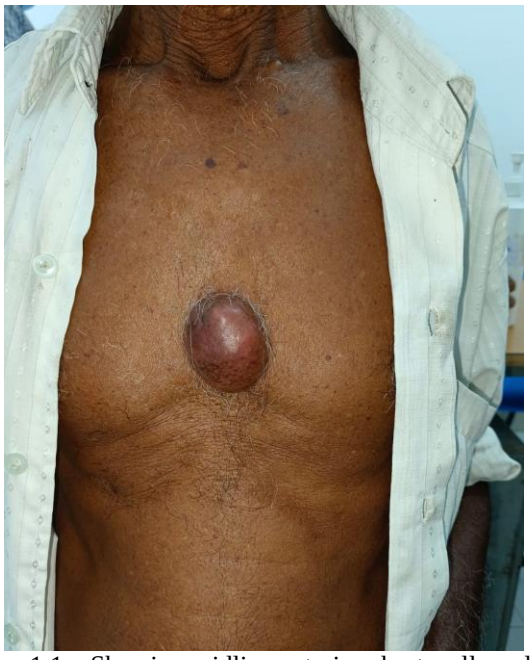

Figure 1.1 – Showing midline anterior chest wall swelling

Differential diagnosis :

1. Calcified Lipoma
2. Soft tissue tumour of anterior chest wall
3. Sebaceous cyst

III. Intraoperative findings

Since it was a benign, solitary swelling, we proceeded with excision biopsydirectly. An elliptical skin incision was made. Layers dissected along the lateral sides and a 5 x 4 x 3 cm nodular, hard mass was noted (Fig. 1.2). Mass with intact wall excised intoto along with overlying skin. Excised specimen was sent for biopsy.

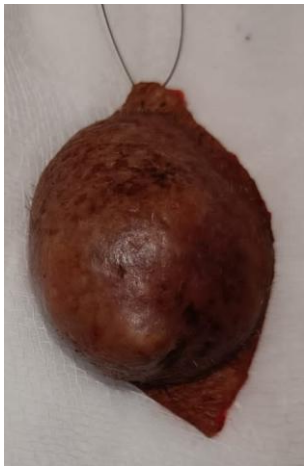

Figure : 1.2 – Showing 5 x 4 x 3 cm nodular mass

IV. Histopathology

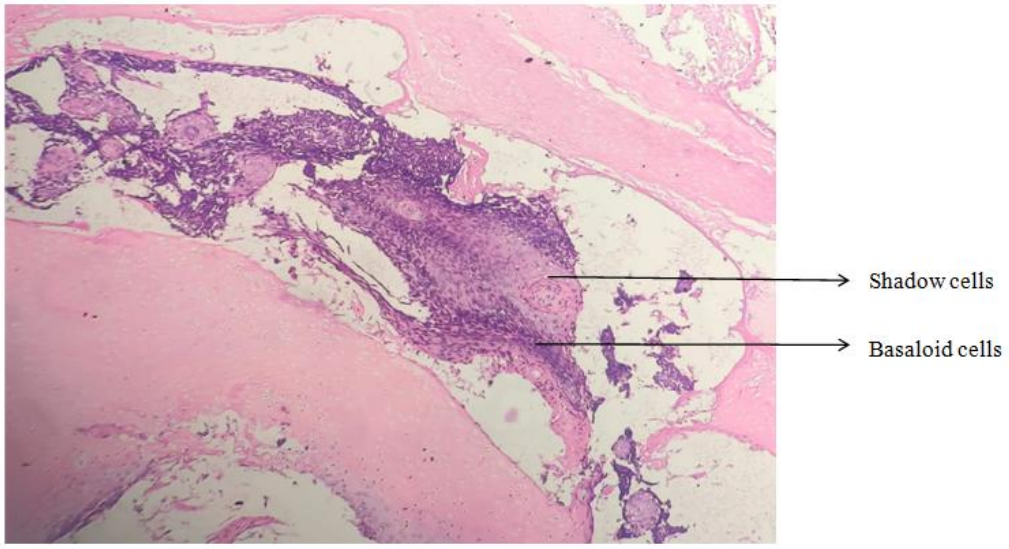

Figure 1.3 – Showing basaloid cells and shadow cells

Examined sections revealed skin with an underlying well demarcated lesion, composed of epithelial cell islands embedded in cellular stroma. The lesion was composed of two types of cells – basophilic cells and shadow cells (Fig. 1.3). The basophilic cells had round or elongated deeply basophilic nuclei and scanty cytoplasm. Features of abrupt keratinization were noted. The shadow cells showed loss of nuclei and appeared as faintly eosinophilic, keratinized cells. Foreign body giant cell reaction and foci of calcification were seen. A few clear cells were seen. The lesion was seen reaching upto deep surgical margin. Mitotic activity was present, but no atypical mitosis or cytological atypia were evident, with features suggestive of ***Pilomatrixoma***.

V. Discussion

Pilomatrixomas were first described by Malherbe and Chenantais in 1880 as a benign neoplasm of sebaceous gland origin.[6]It was not until later that the lesion was understood to be a calcifying epithelioma that arises from hair follicle matrix, hair cortex, follicular infundibulum, outer root sheath, and hair bulge. Pilomatrixoma is a term first used by Jones and Campbell in 1969, when they discovered presentations of subcutaneous lesions in a paediatric population, with similar unique histological features occurring in adults. Pilomatrixomas manifest as a benign, cutaneous, firm, solitary lesions of the face, neck, and upper extremities. The difficulty with diagnosing pilomatrixoma lies with their variant morphology and sometimes unusual appearance similar to more common lesions. The presentation of these subcutaneous nodules may resemble benign lesions such as a keratoacanthoma, ossifying hematoma, and fibroxanthoma, or malignant lesions such as squamous cell carcinoma. Diagnostic identification of this lesion is important because, although extremely rare with fewer than 20 cases described in the literature, pilomatrixomas can undergo malignant transformation into a pilomatrix carcinoma. Most pilomatrix carcinomas occur on the head and neck of middle age to elderly patients. The rate of malignant transformation is difficult to assess due to overall disease rarity and lack of specific features that can distinguish whether a malignant pilomatrixoma has arisen de novo or if it represents the malignant transformation from a pre-existing pilomatrixoma.

Ultrasound has demonstrated the highest accuracy rates between 28.9 and 46 percent. There is also literature citing some diagnostic discriminative findings on ultrasound that may distinguish pilomatrixomas from other subcutaneous tumours : Heterogeneous echo texture, internal echogenic foci in scattered-dot pattern, and a hypoechoic rim or posterior shadowing.[7] Ultrasound imaging of pilomatrixomas usually demonstrates lesions with an ovoid complex mass at the junction of the dermis and subcutaneous fat with focal thinning of the overlying dermis.[8]Computed tomography (CT) can also be used to study pilomatrixomas. They appear as sharply demarcated subcutaneous tumours containing micro-calcifications. The predicament, however, is the same as the clinical diagnosis. There are many lesions that share these CT characteristics, including sebaceous cysts, foreign body reaction, and metastatic bone formations.[9] There have also been reports of the use of magnetic resonance imaging (MRI). Most demonstrate uniform, homogeneous signal on T1 weighted signal, with varying results on T1 with contrast and T2 imaging.[10,11] Therefore, the nonspecific features of pilomatrixoma on MRI, do not allow for its use as a definitive diagnostic tool.

The only truly reliable means of diagnosis remains pathological evaluation. The classic histology is said to be defined by the presence of ghost or shadow cells and basophilic cells. At low power the histological pattern usually seen in pilomatrixoma is of a well-circumscribed nodulo-cystic tumour. While predominantly seen within the lower dermis, extension into the subcutaneous tissue is not uncommon. Pilomatrixomas are traditionally regarded as benign tumours with limited understanding of any possible transition to pilomatrix carcinoma. The clinical difficulty distinguishing pilomatrixomas from more common skin lesions, combined with a patient population that does not have ideal medical access, enforces the importance of incorporating these lesions in the differential diagnosis. Treatment is surgical resection with wide margins of 1–2cm. Following excision, pilomatrixoma recurrences are relatively rare, with an overall rate of 2.6%.[12,13] The rarity of this lesion provides little evidence for follow-up recommendations after excision. One study's treatment population remained free of recurrence during follow up that ranged from 3 to 37 months. If the lesion were to recur, the physician should have increased suspicion of pilomatrix carcinoma, however aggressive surgical excision remains the treatment of choice.[14–17] Surgical excision of this tumour is a sufficient and curative treatment, with excellent postoperative prognosis for both cosmesis and to prevent the possibility of malignant transformation.

VI. Conclusion

Pilomatrixoma is a rare, benign neoplasm that derives from hair follicle matrix cell. It is generally asymptomatic and commonly arises from head and neck region and upper extremities. Malignant transformation is rare. Yet, it should be considered as one of the differentials when suspecting benign swellings arising from hair follicles, skin and subcutaneous tissue. Inspite of several imaging modalities available, histopathological evaluation is considered to be the investigation of choice in coming to a conclusive diagnosis. Surgical excision is the treatment of choice, since the rate of recurrence is rare.

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