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Case Report

Pathology Section

Pilomatricoma with Osseous Metaplasia in the Shoulder Joint: A Common Lesion with Rare Differentiation

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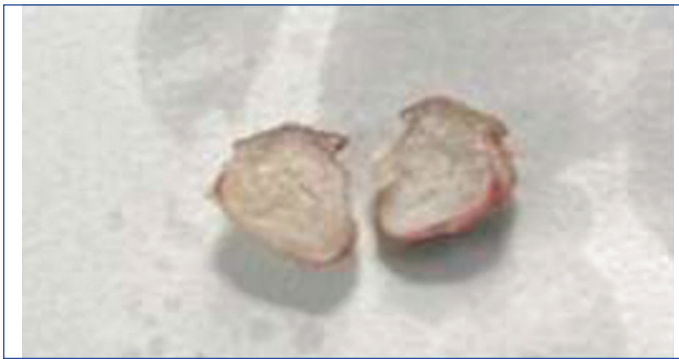
ABSTRACT

Pilomatricoma, also known as calcifying epithelioma of Malherbe, is a tumour that shows differentiation towards hair cortex cells. It was first described by Malherbe and Chenantais in 1880. It most commonly occurs in the first two decades of life, with a female preponderance, and accounts for 1% of all benign skin lesions. Pilomatricoma undergoes calcification and ossification, but ossification is rare and poorly documented. The head and neck region, as well as the upper extremities, are the most common sites. Here, the authors present a rare case of pilomatricoma with osseous metaplasia in a 15-year-old male who presented with shoulder swelling for one year. The patient had a firm to hard, mobile swelling over the right shoulder, which was not fixed to the overlying skin or underlying structures. Clinically, calcified epidermal cysts were considered. Excision was performed in a minor operation theatre, and the specimen was sent for histopathological examination. On gross examination, the specimen was a firm to hard, globular 1×0.8 cm tissue that was difficult to cut. Microscopic examination revealed a lesion composed of two types of epithelium: inner shadow cells and outer basophilic cells. Bony trabeculae with osteocyte rimming were present around the shadow cells, and giant cells were also observed. Based on the histopathological examination, the diagnosis of pilomatricoma with osseous metaplasia was made. The patient was discharged after one week and followed-up for three months, during which satisfactory wound healing and no local recurrence were observed. Pilomatricoma with osseous metaplasia should be considered as a differential diagnosis when a patient presents with a firm to hard nodule.

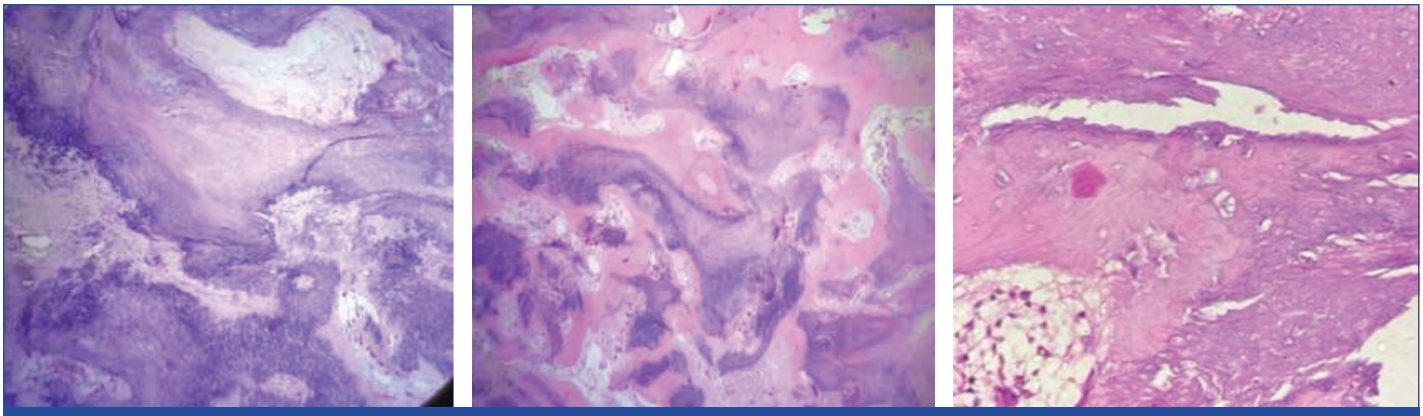
Keywords: Benign adnexal tumour, Epithilioma, Hard nodule, Osteocytes

CASE REPORT

A 15-year-old male patient presented at the surgical Outpatient Department (OPD) with complaints of a painless swelling over his right shoulder for one year, which has recently become harder. The patient was otherwise fit and not taking any medication. The swelling measured approximately 1×1 cm, was firm to hard, mobile, and not fixed to underlying structures or skin. The overlying skin appeared normal. Clinically, epidermal cysts with calcification were considered. Since the lesion was small with no superficial or deep attachments, a simple excision of the lesion without any radiological details was planned. The excision was performed in a minor operating theatre, and the specimen was sent for histopathological examination. On gross examination, the specimen was a firm to hard, globular tissue measuring 1×0.8 cm and was difficult to cut [Table/Fig-1]. Upon sectioning, the lesion appeared white and firm. The specimen was then subjected to decalcification. Microscopic examination revealed a lesion composed of two types of epithelium: inner shadow cells and outer basophilic cells. The basophilic cells, located at the periphery of the lesion, were fewer in number and exhibited mild to moderate basophilic cytoplasm and centrally placed nucleus [Table/Fig-2]. The central region consisted of shadow cells with distinct cell borders and no nuclear staining. Bony trabeculae with osteocyte rimming were observed surrounding the shadow cells [Table/Fig-3,4]. Giant cells were also present. Based on histopathological examination, a diagnosis of pilomatricoma with osseous metaplasia was made.



[Table/Fig-1]: Specimen shows firm to hard tissue.



[Table/Fig-2]: Section of pilomatricoma show shadow cells surrounded by bony trabeculae lined by osteocytes and focal giant cells (H&E, 100X). **[Table/Fig-3]:** Section showing shadow cells and basophilic cells (H&E, 200X). **[Table/Fig-4]:** Section showing bony trabeculae and shadow cells (H&E, 400X). (Images from left to right)

The treatment provided was surgical excision and suturing, along with antibiotic therapy to aid wound healing and prevent infection at the incision site. The patient was discharged after one week and followed-up for three months, during which no recurrence was observed, and complete healing of the incision site was achieved.

DISCUSSION

The most common age group for pilomatricoma is the first and second decade. The age of the patient was consistent with the literature, and there was a male preponderance compared to females [1]. The most common location for pilomatricoma is the head and neck [1-3], followed by the shoulder, as in the present case.

Pilomatricomas are mostly solitary, but multiple lesions can also occur and may be associated with syndromes. These syndromes include sarcoidosis, Turner syndrome, skull dysostosis, and Soto syndrome. Pilomatricoma typically presents as well-circumscribed nodules but can rarely manifest as large, bullous, perforating, or multiple eruptive lesions [3]. Microscopically, the common findings are basophilic cells and shadow cells, also known as ghost cells [4]. Calcification is also commonly observed, appearing as deep basophilic collections [3]. Melanin depositions are also seen [4], mostly in melanophages of the stroma and shadow cells [3]. When present in abundance, the term "melanocytic pilomatricomas" has been used. Dystrophic calcification and haemosiderin deposition are rare findings, while florid osseous metaplasia is extremely rare [4]. Osseous metaplasia can even exhibit extramedullary haematopoiesis [4]. In the present case, no haematopoiesis was observed. The metaplasia of fibroblasts to osteoblasts is considered the mechanism of ossification, occurring adjacent to shadow cells. The cytoplasm of shadow cells shows bone morphogenic protein-2, indicating its role in bone formation [3,4]. The diagnosis of pilomatricoma with osseous metaplasia is important, as different lesions require different treatment strategies. In the case of pilomatricoma, surgical excision is the treatment of choice, with very rare chances of recurrence [5].

There are four stages of pilomatricoma: early, fully developed, early regressive, and regressive. The lesion begins as infundibular matrix cysts and progresses into an ossified nodule. Malignant transformation is rare [6]. Clinically, pilomatricoma with or without osseous metaplasia presents similarly, except that those with osseous metaplasia are firmer on palpation and have a longer history of presentation [7]. The epithelial component is positive for CK-AE1 and CK-AE3, and the basaloid cells show beta-catenin positivity with both cytoplasmic and nuclear staining [8]. To the best of authors knowledge, a total of 10 cases have been reported in the literature so far [2,4,5,7,9-11] [Table/Fig-5]. The clinical differential diagnoses considered include epidermoid, dermoid, and trichilemmal cysts, foreign body granuloma, and cutaneous osteoma [4]. In the present case, the clinical diagnosis was given as an epidermoid cyst. Microscopic differential diagnoses to be considered are trichoblastoma, basal cell carcinoma, and trichoepithelioma, where basaloid cells are prominent [1,4].

Case report	Year	Age/sex	Site
Present case, Aurangabad	2023	15 y/M	Shoulder
Kambale T et al., [2], Pune	2015	52 y/F	Chest wall
Bharti S et al., [4], Jodhpur	2021	Adult /F	Thigh
Sathe PA and Agnihotri MA [5] Mumbai	2022	7 y/M	Upper back
Bhatti N et al., [7] London	2022	87 y/F	Scalp
Zaman S et al., [9] Lahore	2009	1 case out of 27 cases studied	Head and neck
Anunayi J et al., [10] Secundrabad	2013	3 cases out of 15 cases studied	Upper extremity
Lan MY et al., [11] Taiwan	2003	1 case out of 179 cases	Head and neck

[Table/Fig-5]: Previous cases of pilomatricoma with osseous differentiation at various site [2,4,5,7,9-11].

CONCLUSION(S)

A careful clinical and histopathological examination of patients presenting with firm, hard nodules is crucial for reaching a final diagnosis. Pilomatricoma and its differentials should be considered, and pilomatricoma with osseous metaplasia should be included as a possible differential diagnosis for patients presenting with firm, hard nodules, especially in the head, neck, and shoulder regions, by both clinicians and pathologists. Follow-up of the case is important, even though malignant transformation is rare, as recurrence can occur in cases of incomplete excision.

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