Dear Editor,

Pilomatrixoma is a benign tumor of skin appendages with presumed differentiation toward hair follicle matrix cells. It was first described in 1880 by Malherbe and Chenantais as “calcifying epithelioma of sebaceous origin”. It is usually located in the head, neck, or upper extremities with female predominance. Fine needle aspiration cytopathology (FNAC) of the cutaneous lesion is rarely practiced. Although the cytomorphological features of pilomatrixoma are well described in most cytopathology books, these lesions pose a diagnostic dilemma when there is focal sampling with a predominance of one/either component. Rapid on-site evaluation (ROSE) with FNAC can be a useful adjunct for proper sampling of these lesions with the added benefit of material for cell block or special stains.

A 20-year-old female presented to our Dermatology out-patient department, with a swelling over the right suprascapular region for 3 months. There was no history of any local trauma. Personal, past, and family history were unremarkable. On examination, a single erythematous swelling was noted measuring 3 × 4 cm. On palpation, it was soft to cystic in consistency with a deep-seated nodule underneath. Atrophic stria was noted at the edge of the lesion starting from 9’o clock position. Ultra-sonography soft tissue showed a well-defined marginated hypo-echoic lesion measuring 1.2 × 1 × 1 cm with mild subcutaneous edema, and without any vascularity on color doppler, suggestive of sebaceous or dermoid cyst.

Cytology was performed using the non-aspirational method with rapid on-site evaluation using 1% aq. toluidine blue and a 23 guage needle. The aspirate was a whitish granular material. Cytosmears were showing moderate cellularity with clusters and sheets of basaloid cells along with ghost cells and foci of calcification. The basaloid cells were having a high nuclear/cytoplasm ratio and a scant basophilic cytoplasm. The nuclei were round to oval with a uniform outline and occasional nucleoli. There were numerous benign-looking nucleated and anucleated squamous cells seen singly or in clusters. Occasional amorphous dark-stained material was also seen. Numerous ghost-like cells along with a few multi-nucleated giant cells, inflammatory cells, and nuclear debris were also seen. The slides were returned to routine Giemsa and Papanicolaou staining. It also had similar features and cytological diagnosis of the benign adnexal tumor-pilomatrixoma bullous type was given. Cell block of the patient showed a few fragments of ghost cells along with mature adipocytes. The patient had undergone wide excision in an outside laboratory, and the histopathology slide was showing features of pilomatrixoma.

Pilomatrixoma is known by various synonyms, that is, “benign calcifying epithelioma”, “infundibulo-matrix tumor”, or “infundibulopilomatrix cyst”. The incidence of bullous pilomatrixoma is in the range of 3–6%. Our case is unique in the fact that we report bullous pilomatrixoma in cytopathology. There are various theories to explain

Figure 1: (a and b) Flaccid bullous lesion soft in consistency and pale-colored, with a firm nodule underneath the lesion. (c and d) Ultra-sonography: soft tissue showing a well-defined marginated hypo-echoic lesion measuring 1.2 × 1 × 1 cm

Figure 2: (a-d) Cytosmears showed moderate cellularity with basaloid cells in loose clusters melting with the benign-looking nucleated and anucleated squamous cells/ghost cells. Occasional amorphous dark-stained material was also seen [Tol blue, ×10, ×40]
the bullous appearance in pilomatrixoma. However, the main reason is attributed to lymphatic congestion and obstruction because of pressure. Hence, it leads to leakage of the lymphatic fluid with edema in the dermis producing a bullous-like lesion.[5,6]

Most patients present during the second decade with solitary firm nodules over the neck, upper back, arm, buttocks, and thigh with sizes ranging from 1 to 6 cm. They appear as a semi-transparent flaccid blister with a clear border and a hard nodule underneath. The overlying skin may be normal in color, scar-like, or discolored red, blue, or purple.[6,7] It can be associated with atrophic stria as seen in our case. Most bullous forms were reported on the shoulder and upper limbs, similar to our patient.[6,7]

FNAC is a rapid, cost-effective, and minimally invasive method used for palpable superficial swelling diagnosis. The diagnostic accuracy of pilomatrixoma in FNAC is very low and varies from 21 to 44% in various studies.[7] Cytomorphological features of a pilomatrixoma can be very characteristic comprising a cohesive cluster of basaloid cells, empty-looking shadow (ghost) cells, scattered nucleated squamous cells, and focal calcification. The foreign body giant cell reaction can be because of engulfed keratin.[8,9]

Cell block preparation of the aspirate can be a helpful adjunct with hematoxylin and eosin (H&E) sections showing the squamous ghost cells, basaloid cells, giant cells, and variable amounts of calcium which can help in the diagnosis of pilomatrixoma, and exclude malignancy.[9]

The role of FNAC in the diagnosis of pilomatrixoma is a point-of-debate/diagnostic dilemma.[10] There may be some degree of nuclear atypia in squamous cells and basaloid cells mis-interpreted as malignant. The differential diagnosis includes skin appendageal neoplasm, basal cell carcinoma, squamous cell carcinoma, and cutaneous and metastatic neuroendocrine carcinoma.[8,9] Malignant transformation of pilomatrixoma is rare. The treatment of pilomatrixoma includes wide local surgical excision with clear margins. The overall recurrence rate after complete excision is approximately 2–6%. FNAC with ROSE can provide a definitive diagnosis in cases where adequate material and cytological features are carefully looked for.

**Ethics approval and consent to participate**

This case report was conducted in accordance with the fundamental principles of the Declaration of Helsinki.

**Consent for publication**

Written consent for publication and any additional related information was taken from all the patients involved in the study.

**Authors’ contributions**

TS carried out concepts & design, literature search, participated in clinical study & manuscript preparation will stand as guarantor also. JK carried out data acquisition, & data analysis. HG, AS carried out concepts & design, literature search. All the authors have read & approved the final manuscript.

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**Conflicts of interest**

There are no conflicts of interest.

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