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Sebaceoma in a 58 Years old Male Diagnosed on Cytology and Confirmed on Biopsy- A Case Report

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Abstract

Background: Sebaceoma is a benign adnexal tumor that usually presents as a yellowish nodule or plaque, most commonly seen in the head and neck region. The occurence of sebaceoma has been associated with Muir–Torre syndrome which is an autosomal dominant disorder and predisposes to other visceral malignancies. **Case Report:** We hereby, report a case of Sebaceoma involving the left axillary region in a 58 yrs old male. FNAC smears were hypercellular showing many clusters of basaloid cells, few showing anisonucleosis along with scattered cells of sebaceous differentiation, having vacuolated cytoplasm. The provisional diagnosis of Sebaceoma was suggested on cytology. These features were later confirmed by histopathological sections showing small, monomorphous basaloid cells admixed with cells showing sebaceous differentiation, having vacuolated cytoplasm and scalloped nuclei. We have attempted to review the literature on Sebaceoma and emphasized on its differential diagnoses, particularly sebaceous carcinoma and basal cell carcinoma with sebaceous differentiation. **Conclusion:** Sebaceoma is a rarely reported tumour and its diagnosis on cytology is way more challenging. Awareness of the cytological features of Sebaceoma will help in early specific diagnosis of this tumor at the time of aspiration, rather than a blanket term of benign adnexal tumour . Also, its association with Muir–Torre syndrome, warrants its correct and timely identification and thus, genetic testing and surveillance for its other syndromic associations.

Keywords: Adnexal tumor, Muir–Torre syndrome, cytology, basaloid.

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Introduction

Sebaceoma is a rare, benign adnexal skin neoplasm which is most commonly found in the head and neck region of older individuals.[1,2] Sebaceomas may be associated with the Muir-Torre syndrome, in which they may be multiple and associated with other sebaceous neoplasms, multiple adenomatous polyps and visceral malignancies.[3]

Case Report

A 58-year-old male presented to us with left chest swelling since 1 year. Swelling was not associated with pain or increase in size and no history of trauma to the site was present. Patient was on medications for hypothyroidism. There was no history of tuberculosis or tubercular contact. The family history was not significant and was negative for any malignancy.

On examination, swelling measured 1.5X1 cm, swelling was firm, round, mobile with distinct borders [Figure 1].

Overlying skin was hyperpigmented with yellowish discharge with no overlying skin infection. The remainder of the head and neck examination was unremarkable with no other swelling present. X Ray chest was within normal limits.

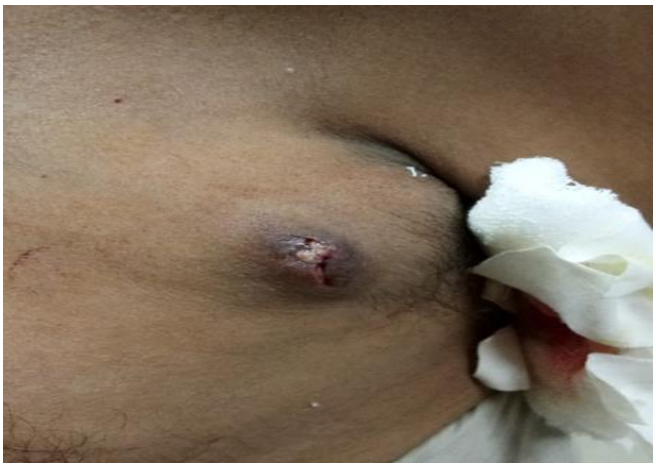

Figure 1: Firm, round and mobile left chest swelling with distinct borders, measuring 1.5X1 cm

Overlying skin was hyperpigmented with yellowish

discharge with no overlying skin infection. The remainder of the head and neck examination was unremarkable with no other swelling present. X Ray chest was within normal limits

FNAC was performed. Smears were hypercellular showing many clusters of basaloid cells having small, monomorphous, oval nuclei, few showing anisonucleosis without prominent nucleoli, along with scattered cells of sebaceous differentiation, having vacuolated cytoplasm in a background of chondromyxoid stroma [Figure 2 to 3]. No mitotic figures were noted. Provisional diagnosis of adnexal tumour with sebaceous differentiation was suggested.

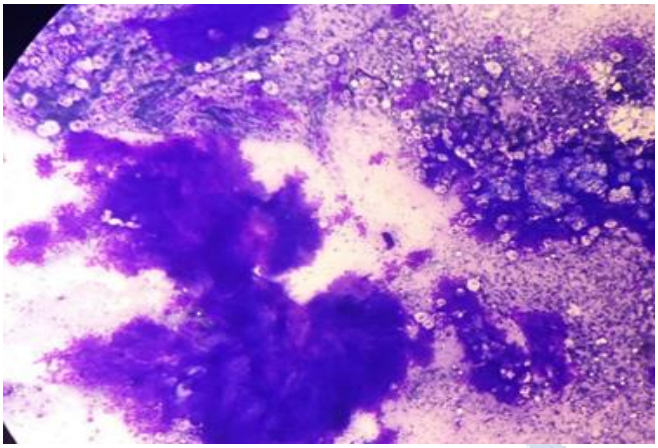

Figure 2: Hypercellular smears showing many clusters of basaloid cells in a background of chondromyxoid stroma

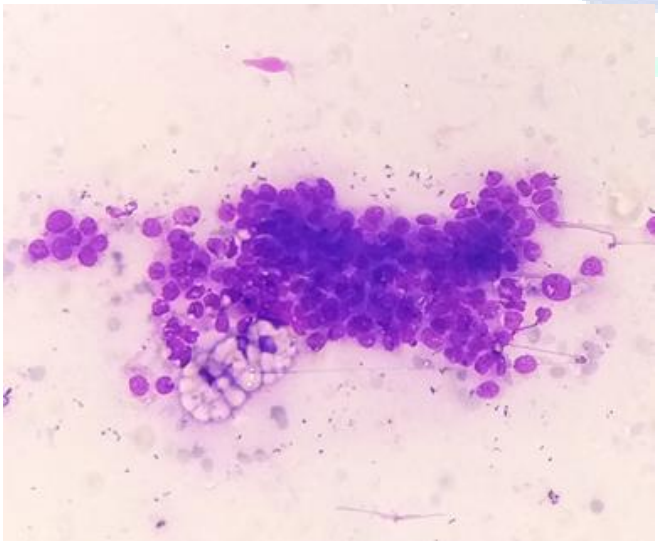

Figure 3: Clusters of basaloid cells having small, monomorphous, oval nuclei, few showing anisonucleosis without prominent nucleoli, along with scattered cells of sebaceous differentiation, having vacuolated cytoplasm

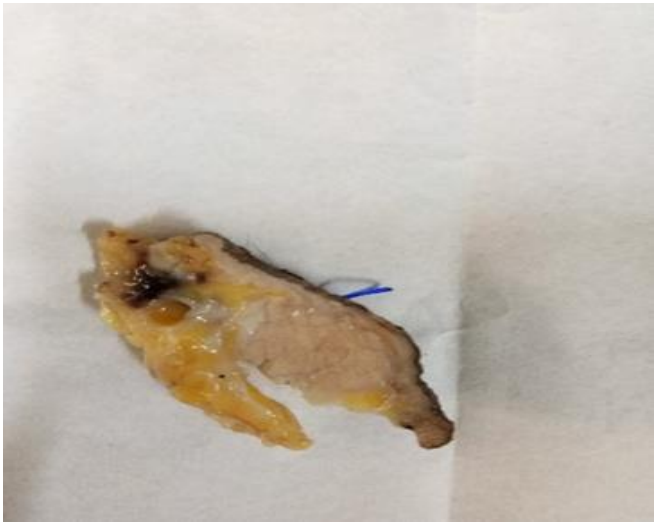

Figure 4: Gross examination showed a single soft tissue piece covered with skin measuring 4X3X3.5cm with cut surface showing greyish white nodular lesion

Wide local excision of the tumour was performed and margins were submitted. Gross examination showed a single soft tissue piece covered with skin measuring 4X3X3.5cm [Figure 4]. Overlying skin showed ulceration. Cut surface showed greyish white nodular lesion measuring 2X2 cm with areas of haemorrhage.

Histopathological sections showed a relatively sharply circumscribed tumour, comprising of a benign, basaloid adnexal neoplasm admixed with sebocytes and sebaceous duct-like structures located in the dermis without connection to the epidermis, consistent with a Sebaceoma. Few cystic structures lined by sebaceous cells were noted with cellular intervening stroma. Few cystic spaces contained eosinophilic homogenous material. There was focal breach of the overlying epidermis. No pleomorphism or mitotic activity were noted. No nuclear atypia and invasion into the deep subcutaneous tissue was noticed. Also, no palisading of the cells at the periphery of the aggregates was seen [Figure 5 to 8].

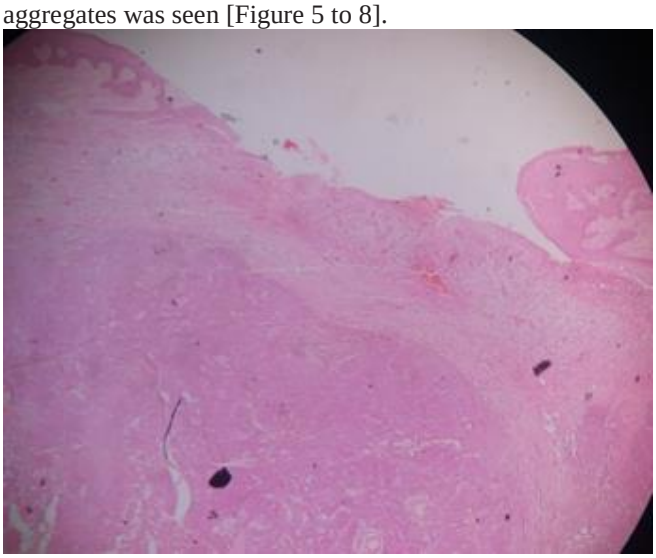

Figure 5: Low power view showing relatively sharply circumscribed tumour with focal breach of the overlying epidermis.

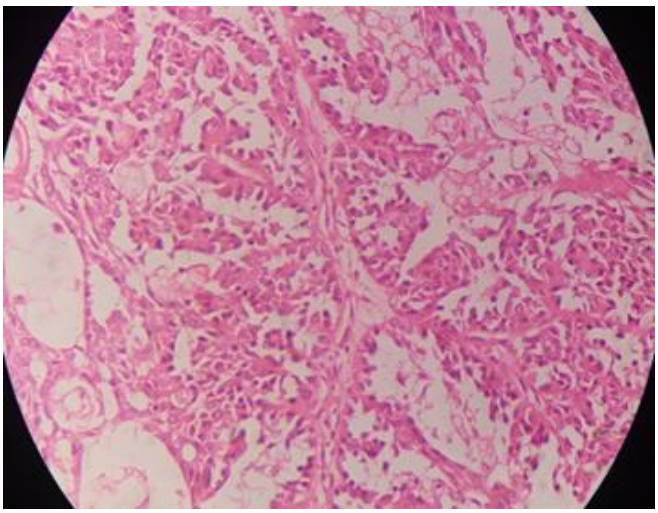

Figure 6: Histopathological sections showed basaloid adnexal neoplasm admixed with sebocytes and sebaceous duct-like structures located in the dermis without connection to the epidermis, consistent with a Sebaceoma

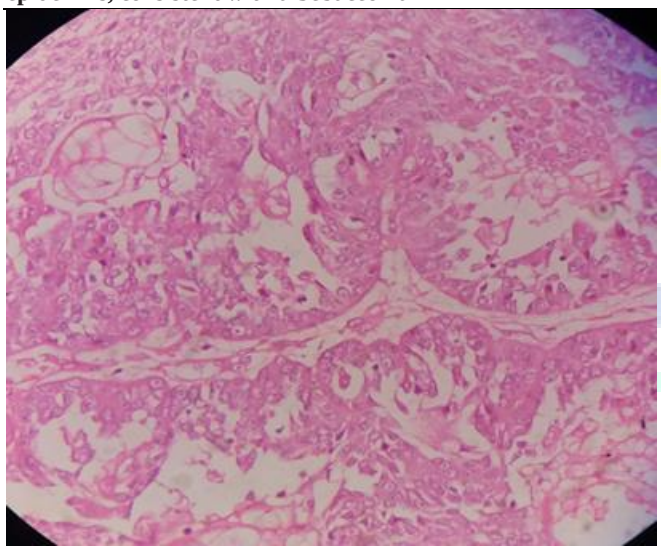

Figure 7: Histopathological sections showed basaloid adnexal neoplasm admixed with sebocytes and sebaceous duct-like structures located in the dermis without connection to the epidermis, consistent with a Sebaceoma

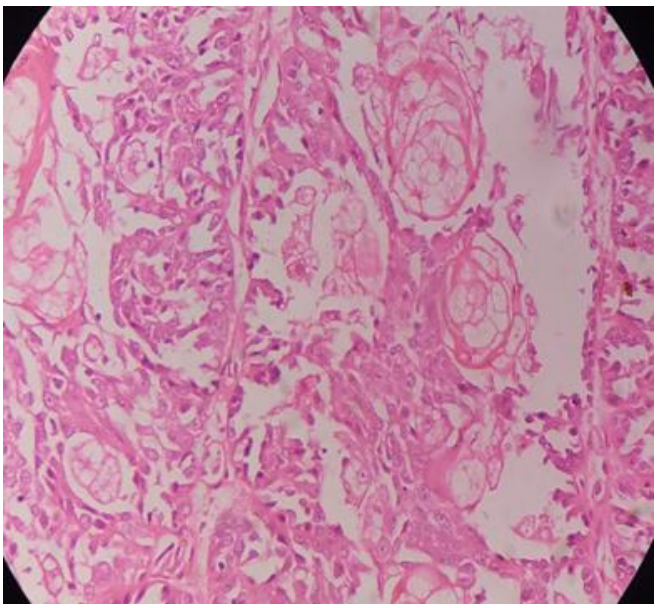

Figure 8: Histopathological sections showed basaloid adnexal neoplasm admixed with sebocytes and sebaceous duct-like structures located in the dermis without connection to the epidermis, consistent with a Sebaceoma

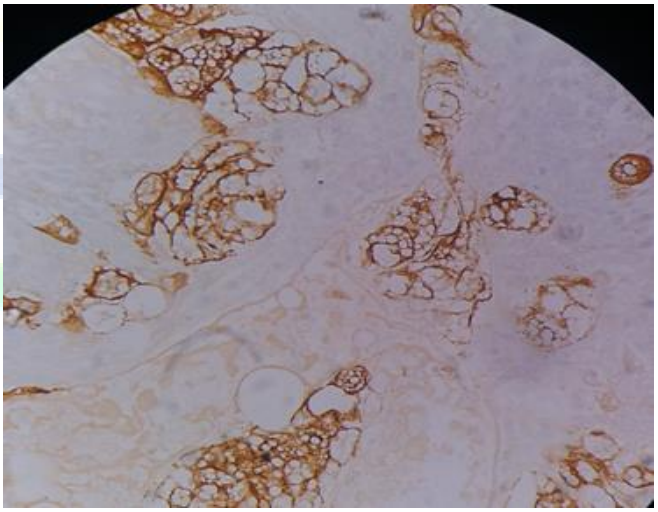

Figure 9: On immunohistochemistry, Epithelial Membrane Antigen (EMA) was positive

The margins were reported negative for the tumour. On immunohistochemistry, Epithelial Membrane Antigen was positive, while other immunohistochemical markers including S-100, Vimentin, Carcinoembryonic Antigen, Cytokeratin, BCL-2, Ber EP4, Ki-67, CD34 and CD10 were negative [Figure 9]. Special stains including Periodic acid Schiff with and without Diastase were negative.

However, genetic studies could not be performed as the patient was lost to follow up.

Discussion

Troy and Ackerman coined the term, Sebaceoma in 1984, and simplified the criteria for Sebaceoma.[4] Sebaceous lesions have been described arising from the nose, face,

scalp, neck and external auditory canal.[1,2] Infact, these neoplasms can occur in any location containing hair and sebaceous glands, and their prevalence on the face and scalp reflects the abundant sebaceous glands and hair in these areas.[5]

Fan et al showed that out of 25 sebaceomas studied, the majority (80%) were located in the head and neck region, predominantly the nose and cheek.[2]

Sebaceoma usually presents as an asymptomatic, slow growing, solitary, yellowish papule or nodule with rolled borders.[5] Surface ulceration and bleeding has also been reported.[1,2]

Sebaceoma has been described as a benign tumour involving the basaloid or germinative sebaceous cells, with basaloid, undifferentiated sebocytes mixed with mature sebocytes.[2] These basaloid cells can exhibit various patterns, including cribriform, reticular or ripple arrangements.[6] In our case, papillary formations were seen. It usually has well circumscribed lobulated architecture, lacks nuclear atypia and frequent mitotic figures.[2]

Sebaceous differentiation is defined as the presence of multiple cytoplasmic fat vacuoles, sometimes indenting the nucleus, and immunohistochemical expression of EMA.[7]

The distinction between sebaceous adenoma and sebaceoma is that in sebaceous adenoma, more than half of the lobules are composed of mature sebocytes.[8]

Therefore, neoplasms with sebaceous differentiation including benign entities, such as sebaceous adenoma, the malignant ones including sebaceous carcinoma, nodular basal cell carcinoma, trichoblastoma and apocrine poroma should always be considered in the differential.[1]

Trichoblastoma with sebaceous differentiation, apocrine poroma with sebaceous differentiation, and basal cell carcinoma with sebaceous differentiation are easily confused with the authentic neoplasms with sebaceous differentiation.[9]

Sebaceoma has well preserved architecture and nuclear nuclear atypia, contrary to its more aggressive counterpart, sebaceous carcinoma and hence differentiated.[2] Also, palisading of nuclei at the periphery of the neoplastic aggregates and separation of the cell groups from adjacent stroma by clefts is not seen in sebaceoma, unlike its close differential basal cell carcinoma.[1,10]

However, sometimes only histological differentiation from basal cell carcinoma does not suffice, and we have to resort to aid of immunohistochemistry and molecular markers such as Ber-EP4 and epithelial membrane antigen (EMA) in confirming the diagnosis.[2] Fan et al,[2] reported that Ber EP4 could make the distinction between basal cell carcinoma with sebaceous differentiation and sebaceoma with 100% specificity and sensitivity, being expressed in basal cell carcinoma but not in sebaceoma.

Usually, Sebaceoma is easily distinguished from trichoblastoma as it is a neoplasm with follicular differentiation,[11,12] however, sometimes, sebaceomas have only scattered mature sebaceous cells, and trichoblastoma may present without prominent follicular differentiation.[9,13] Histological features like rudimentary follicular papillae, a palisading border in the tumor cell aggregates and the presence of fibrotic stroma with clefts suggests towards trichoblastoma rather than Sebaceoma.[13]

Another diagnostic differential is seborrheic keratosis and is distinguished from sebaceoma due to its characteristic features like typical flat-base, stuck on appearance, prominent keratin horn cysts and lack of sebaceous differentiation and EMA expression.[14]

There is well established association of sebaceoma with Muir–Torre syndrome, which is an autosomal dominant disorder with variable penetrance and expression and is considered a variant of hereditary nonpolyposis colorectal cancer.[11] Patients with this syndrome show sebaceous neoplasms or keratoacanthomas along with visceral malignancies, most commonly of the gastrointestinal or genitourinary system.

Genetic mutations in several DNA mismatch repair proteins and microsatellite instability have been etiologically associated, therefore, immunohistochemical and germline mutation analysis can aid in the diagnosis of Muir-Torre syndrome is suspected.[11,12]

Also, one should always maintain a high index of clinical suspicion for the development of other malignancies in patients diagnosed with Sebaceoma, and the patient should therefore undergo strict surveillance with serial physical examinations, imaging and other relevant investigations.[3,11]

Conclusion

Sebaceoma is a rarely encountered, benign adnexal neoplasm which most commonly presents in the head and neck region. Treatment is local excision, and recurrence is uncommon. Fine needle aspiration cytology (FNAC) is a simple, safe, quick, cost effective and informative procedure. Awareness of the cytological features of Sebaceoma will help in early specific diagnosis of this tumor at the time of aspiration, rather than a blanket term of benign adnexal tumour. Correct and timely diagnosis of a sebaceous-derived neoplasm is crucial due to its association with Muir–Torre syndrome. The diagnosis of sebaceoma in a patient should warrant a thorough search for underlying visceral malignancies, and essential genetic testing and surveillance for visceral malignancies.

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