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Original Research Paper

Pathology

SYNRINGOCYSTADENOMA PAPILLIFERUM: A RARE CASE REPORT

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ABSTRACT Syringocystadenoma papilliferum is a rare benign hamartomatous adnexal tumour that occurs chiefly on the scalp, neck and face. It is present since birth or in early childhood. We are presenting a case of Syringocystadenoma papilliferum which was clinically diagnosed as Molluscum contagiosum. Histopathological diagnosis was crucial and confirmatory in this case since Syringocystadenoma papilliferum has the potential to transform into malignant Syringocystadenocarcinoma Papilliferum, hence, surgical excision of lesion followed by histopathology is the preferred mode of treatment.

KEYWORDS : Benign adnexal tumor, SCAP, skin

INTRODUCTION

Syringocystadenoma papilliferum, also known as papillary syringadenoma, is a rare benign hamartomatous adnexal tumour that occurs chiefly on the scalp, neck and face but may be found elsewhere on the skin. It is present since birth or in early childhood in about 50% of the cases, while in 15-30% the tumor develops later during puberty.

CASE REPORT

A 32 years old female presented with complaints of a red to skin coloured, raised lesion measuring 0.5 x 0.5 cm on the right side of her face since birth. The lesion was associated with itching and white discharge on touch which had aggravated in the past one week. On examination, the right side of cheek showed a solitary, discrete erythematous papule with overlying crusting and mild erythema, seen on a background of ill-defined skin colored plaque. It was clinically diagnosed as Inflamed Molluscum Contagiosum. A 4 mm skin punch biopsy was taken from the site of the lesion and sent for histopathological examination.

Gross findings: A single, grey white to grey brown, soft to firm, skin covered, hair bearing tissue bit was received measuring 0.4 cm.

Microscopic Findings: Microscopy showed epidermis and dermis. Papillary dermis revealed invaginated cystic structures partly covered by outer and inner lining. Outer part of the cystic structures showed lining squamous epithelium in the upper dermis. Lower part of the dermis showed variable sized cystic structures with infolding papillary structures lined by double layered epithelium. Inner layer constituted by tall columnar and outer by cuboidal epithelium.

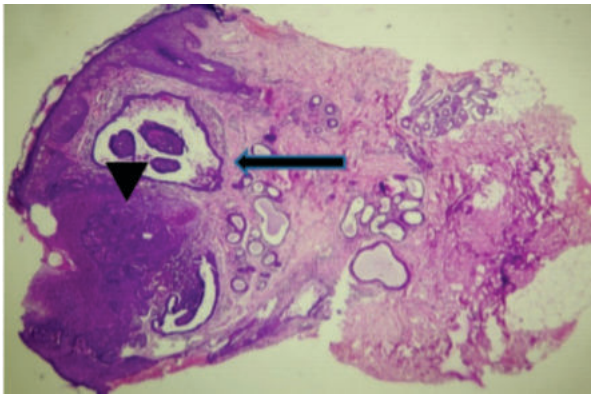

Fig. 1 Scanner view (4x) shows epidermis and dermis. Upper dermis shows invagination of cystic structures with infolded papillary structures (thin arrow) covered by an outer lining of squamous epithelium (arrowhead).

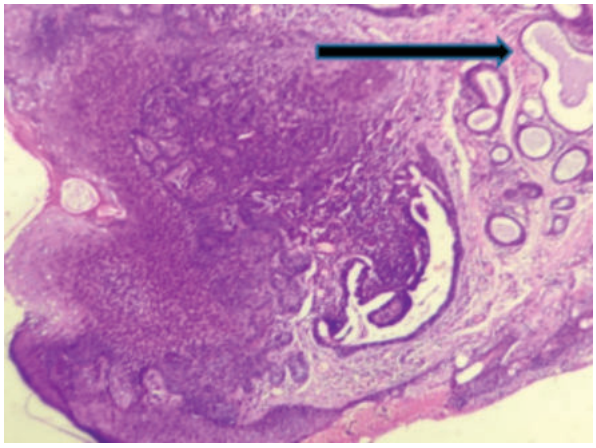

Fig. 2 Low power view (10x) shows papillary structures lined by double layered epithelium - inner layer constituted by tall columnar and outer layer by cuboidal epithelium. Cystically dilated apocrine glands (black arrow) filled with secretions are also visible.

Inner columnar lining showed apical decapitated secretions (apical snouts). The fibrous stroma of cores showed dense infiltration with sheets of plasma cells and few lymphocytes. The deeper part of the biopsy showed cystically dilated apocrine glands lined by dual lining and filled with secretions.

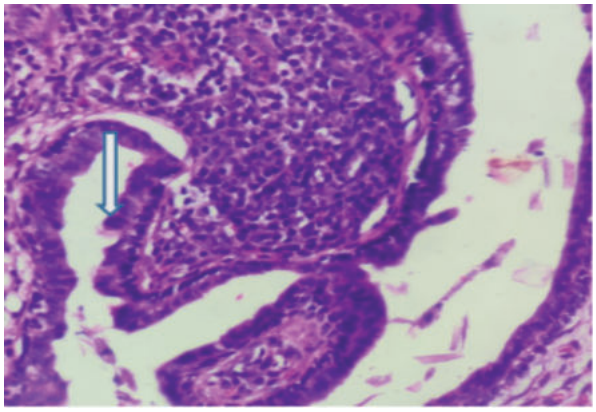

Fig. 3 Higher magnification (40x) of papillary structures

show inner layer of tall columnar cells showing apical decapitated secretions - apical snouts (white arrow).

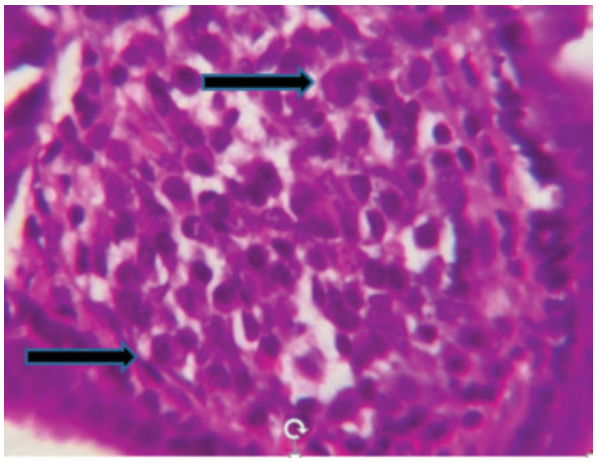

Fig. 4 Oil field immersion (100x) The stroma shows dense infiltration with sheets of plasma cells and lymphocytes. Plasma cells (black arrow) with perinuclear hof, eccentric nucleus with coarse and clock face chromatin (cart wheel pattern).

DISCUSSION

Syringocystadenoma papilliferum (or SCAP) is a rare benign hamartomous tumor of the skin. SCAP commonly occurs on the head and neck, but there have been reported cases of lesions involving abdomen, chest, breast, axillae, genitalia, arm, thigh and eyelid. The lesion may present in the form of a group of nodules, a nodular plaque or a solitary nodule. The solitary nodular type is found more commonly on the trunk whereas the nodular variety has a predilection for the scalp, head and neck.[6] The surface of the lesion can be smooth, flat, papillomatous or verrucous and crustations or change in size is noted at puberty.[2]

Due to its rare presentation, the SCAP lesion is often misdiagnosed clinically as it was in our case. The various differential diagnosis are viral warts, naevus sebaceous, linear naevus verrucosus, naevus comedonicus, apocrine poroma, apocrine hidrocystoma, tubulopapillary hidradenoma, hidradenoma papilliferum, papillary eccrine adenoma, verrucous carcinoma, apocrine acrosyringeal keratosis, poroma folliculare, linear naevus verrucosa, atypical fibroxanthoma, clear cell syringoma, basal cell epithelioma, sebaceous epithelioma, trichoepithelioma and verruca vulgaris.[6-10] In about one-third cases, SCAP is associated with naevus sebaceous, with development to Basal cell carcinoma in majority of those cases. Transformation to squamous cell carcinoma has also been reported but not that frequently.[11]

The malignant counterpart of Syringocystadenoma papilliferum is Syringocystadenocarcinoma papilliferum. Any increase in size or shape or ulceration of the lesion is indicative of malignant transformation. The benign or malignant nature of the lesion needs to be confirmed histopathologically.

The definitive treatment for Syringocystadenoma papilliferum is surgical excision to prevent malignant transformation. The excised tissue must be sent for detailed histopathological examination to ascertain the free margins.

CONCLUSION

In conclusion, Syringocystadenoma papilliferum is a rare benign adnexal tumor which may be clinically misdiagnosed as other skin lesions such as Hidradenoma papilliferum, Keratoacanthoma and Molluscum contagiosum, etc due to its clinical presentation. Histological studies are crucial in narrowing down the differentials and establishing a diagnosis in such cases, so as to arrive at definitive diagnosis and help frame a proper treatment plan for the patient.

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