



**SYRINGOCYSTADENOMA PAPILLIFERUM WITH CYLINDROMATOUS AND SEBACEOUS DIFFERENTIATION: A RARE CASE REPORT**

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**ABSTRACT**

Syringocystadenomappilliferum (SP) is a rare adnexal neoplasm seen in the head and neck region. Occurrence of the lesion in other sites is uncommon. A 60yr old woman presented with a solitary swelling in the breast. The histopathological diagnosis of syringocystadenomappilliferum with cylindromatous and sebaceous differentiation was made. We report a case of sp in unusual location with multiple adnexal differentiation for the first time to the best of our knowledge.

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**INTRODUCTION**

Syringocystadenomappilliferum (SP) is a rare adnexal neoplasm seen in the head and neck region. Occurrence of the lesion in other sites is uncommon. We report such a rare tumor with multiple differentiation.

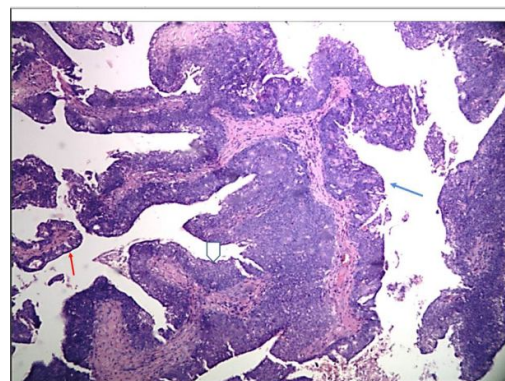
**Case Report**

A 60yr old woman presented with a solitary, non-tender, non-pruritic lump in upper outer quadrant of the left breast gradually increasing in size from four month. There was no history of discharge or nipple retraction. She was hypertensive on tablet enalapril 5mg from 4yrs. On examination a freely mobile lump in the left upper outer quadrant of the left breast measuring 5x4cms was palpated. Skin over the lump was normal with no erosion or discharge. Excisional biopsy was performed and sent for histopathological examination. Histopathological examination showed cystic mass with multiple gray-white tissue pieces. Microscopic examination of the biopsy showed a tumour composed of an inner layer of cuboidal epithelial cells and an outer layer of basaloid myoepithelial cells arranged in papillary configuration projecting within the cystic spaces. The tumour groups showed cells lining hyaline globules in the form of rosettes, eccrine ducts and basement membrane material within and outside tumour groups. Focal areas of sebaceous differentiation was also present as in Figure 1. The diagnosis of benign adnexal tumor of syringocystadenomappilliferum with cylindromatous and sebaceous differentiation was made.

**DISCUSSION**

Syringocystadenomappilliferum is a benign hamartomatous adnexal tumor of apocrine differentiation. It is commonly seen in head and neck region in the scalp and few cases outside the head and neck region have been reported. Various authors have reported SP in different sites like vulva, breast, trunk and perineum. Mammino *et al.*

reviewed 145 cases with syringocystadenomappilliferum and found 108 cases (75.0%) occurring on the head and neck region, 29 cases (20.0%) on the trunk, and 8 cases (5.0%) on the "extremities" (Mammino and Vidmar 1991). SP can arise de novo or from pre-existing nevus sebaceous. It can present since birth or in childhood. Occurrence of the tumor in the elderly female is unusual. SP can be linear, nodular or plaque like. Our patient presented with nodular type of SP as a lump in the breast (Sudip *et al.*, 2012). Microscopically SP shows cystic structures and papillary projections with keratinized stratified squamous epithelium in the upper part and double lining epithelium, an inner columnar cells with apocrine differentiations and basal layer of cuboidal epithelium in the lower part of the dermis. Our patient showed features of sp with areas of sebaceous and cylindromatous differentiation. There is been only one case report of sp with sebaceous differentiation by vizmitel *et al* who suggested the occurrence due to close embryological relationship of the folliculosebaceous-apocrine unit (Vazmitel *et al.*, 2008).



**Figure 1.** syringocystadenoma papilliferum showing bilayered epithelium (as shown in arrow), areas of sebaceous differentiation (as in arrow head) and areas of cylindromatous differentiation (as in blue arrow). H&E 10 x.

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We report the first case of sp with sebaceous and cylindromatous differentiation to the best of our knowledge.

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