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Case Report

Unmasking the error: Syringocystadenoma papilliferum mistaken for squamous cell carcinoma

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Abstract

Syringocystadenoma Papilliferum (SCAP) is a rare benign adnexal tumor. We report a rare case of Syringocystadenoma Papilliferum (SCAP) in a 20-year-old male, initially misdiagnosed as Squamous Cell Carcinoma (SCC) based on clinical history, atypical site of the lesion, examination and family history of breast carcinoma. However, histopathological examination revealed typical features of SCAP, thus, emphasizing the critical role of histopathology in distinguishing benign conditions from malignancies and avoiding unnecessary intervention. It also highlights a *potential molecular link involving the MAPK pathway*, shared between SCAP and breast carcinoma, warranting further research.

Keywords: Syringocystadenoma papilliferum, SCC, Histopathology, Dermoscopy

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1. Introduction

Syringocystadenoma papilliferum (SCAP) is a rare benign hamartomatous tumor of the adnexal glands arising from pleuripotent cells, usually developing on a pre-existing Nevus Sebaceus.[1] It typically manifests at birth, in infancy, or during puberty, and is frequently found on the head and neck.[2] Given SCAP's potential for malignancy and its resemblance to other malignant conditions, prompt and accurate diagnosis is imperative. We report a rare case of SCAP manifesting with an atypical morphology initially misdiagnosed as squamous cell carcinoma (SCC).

2. Case Report

A 20-year-old male presented to Dermatology OPD with complaints of a solitary lesion present on lower back since birth. Initially, the patient had a pin head sized, raised, hyperpigmented, rough surfaced lesion which increased in size over the past 6 months, associated with change in texture and bleeding on manipulation. The site coincided with the waistband of clothing making it prone to frequent friction and trauma. Family history revealed that his mother had a history of invasive breast carcinoma 1 year ago. There was no other significant personal history, treatment history or history of any other genetic condition.

On examination, solitary, firm, well defined greyish black plaque with verrucous surface and few erosions, measuring 2.5cm * 1.5cm was present on the lumbar region (L2-L3) with overlying whitish, adherent scaling (**Figure 1**). Regional lymph nodes were not palpable. Based on this history and examination, an initial diagnosis of SCC arising from Verrucous Epidermal Nevus (VEN) was made.

Dermoscopy revealed milky red papillomatous growth with central erosions and polymorphic vessels. Some areas displayed white circles over a background of whitish blue hue. There were no structureless areas or perivascular halo.(**Figure 2**)

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On ultrasonography, two anechoic cystic lesions with the largest measuring 6*5*2 (vol0.03cc) were seen.(**Figure 3**)

Histopathology revealed numerous papillae lined by double layered to multilayered epithelium. The basal layer is flat to cuboidal, while outer layer is columnar showing decapitation at places. The central stromal core of these papillae showed collection of plasma cells. The superficial dermis showed cystic dilation of hair follicles, filled with eosinophilic material with no evidence of atypia/malignancy. PAS stain was done, and it did not show any breach in basement membrane.(**Figure 4**)

Based on clinical, histopathological, dermoscopic and ultrasonographic findings, a final diagnosis of SCAP was established.

Table 1: Clinical variants of SCAP[2]

Clinical Variant	Clinical Presentation
Plaque Type	Appears as a hairless patch on scalp that grows into wart-like, nodular, or crusty plaques during puberty
Linear Type	Comprises several reddish, firm papules, or indented nodules of 1-10 mm on face and neck
Nodular Type	Raised, stalked nodules measuring 5-10 mm, typically found around trunk, shoulders, and armpits

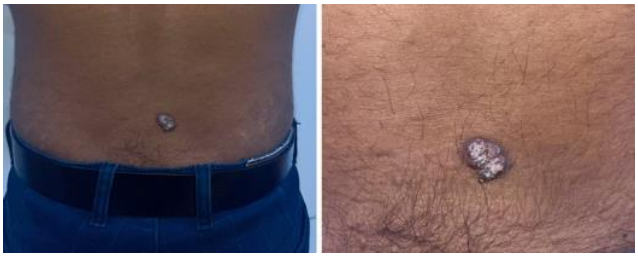

Figure 1: Solitary, well defined greyish black plaque with verrucous surface measuring 2.5cm * 1.5cm present in the lumbar region (L2-L3) with yellowish white dirty scaling

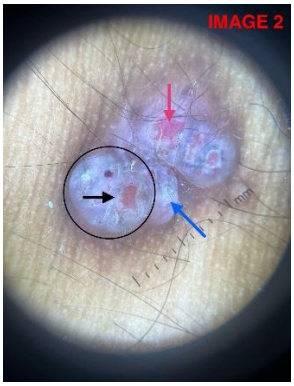

Figure 2: Dermoscopic findings using DL200 hybrid; Black Circle – Milky white papillomatous growth; Black arrow – Central erosion; Blue Arrow – White circles; Red Arrow – Polymorphic vessels.

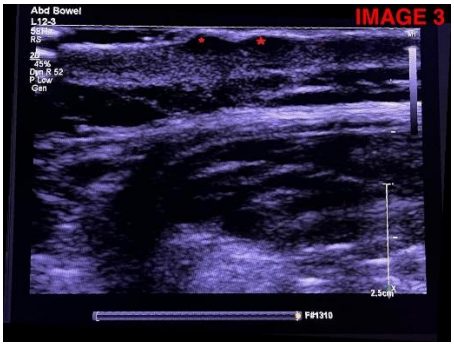

Figure 3: Ultrasonographic image showing two anechoic cystic lesions

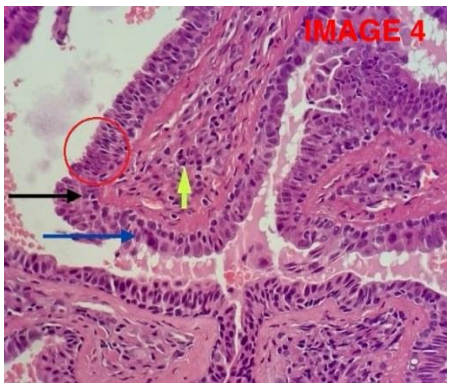

Figure 4: H&E stain (10x); Exophytic lesion comprising of numerous papillae lined by double layered to multilayered epithelium with a flat to cuboidal basal layer (Black arrow) and a columnar outer layer (Blue arrow) with decapitation at places (Red circle). Central stromal core of these papillae revealed collection of plasma cells (yellow arrow)

3. Discussion

Syringocystadenoma papilliferum (SCAP) is an uncommon hamartomatous adnexal tumor frequently associated with Nevus Sebaceus (30%).[1] Other adnexal tumors such as trichoblastoma, apocrine adenoma, hidradenoma papilliferum, and trichilemmoma, can also develop over nevus sebaceous.[3] Although head and neck are the most common sites, it can also appear in atypical locations such as face, chest, abdomen, arms, thighs, and perineum.[2]

Clinically, there are three variants of SCAP as mentioned in **Table 1**. While plaque type of SCAP typically manifests on scalp, our case involved an atypical occurrence situated on lower back—a location more commonly associated with nodular variant of the condition. Dermoscopy revealed papillomatous growth exhibiting a milky-red hue, accompanied by central erosion. Certain regions showed presence of white circles over a background of whitish blue hue, which was similar to findings described by Chauhan et al.[4].

Histopathologically, as described by Ahmed A. Alhumidi, the condition involves formation of cystic structures protruding downwards from outer layer of skin, creating small cavities lined with two layers of epithelial and myoepithelial cells. These cells exhibit evidence of

decapitation secretion. Additionally, there is presence of numerous plasma cells within the surrounding tissue, characteristic of the condition as was also seen in our case.[5]

Syringocystadenocarcinoma papilliferum, the malignant form of SCAP, presents as an asymmetrical, exophytic, flesh-coloured to hyperpigmented lesion, lacking distinct boundaries and often penetrates deeply into the subcutaneous fat layer.[2,6] A distinctive characteristic is the crust on the surface, which is due to the secretions from the apocrine gland.[6] Other conditions arising from SCAP include basal cell carcinoma, metastatic carcinoma, and ductal carcinoma.[2]

In our case, verrucous appearance of the lesion, presence since birth, history of repeated trauma or friction, recent increase in size and change in morphology combined with a positive family history of breast carcinoma in the patient's mother, led to a provisional diagnosis of SCC arising from VEN. Although there is no published literature directly linking breast carcinoma with SCC or SCAP, it is noteworthy that p53 gene, which is mutated in almost 30% of all breast cancer patients,[7] is also frequently mutated in cutaneous SCC.[8] Additionally, the Mitogen-Activated Protein Kinase (MAPK) pathway, a crucial cell signalling mechanism, is often disrupted in various cancers, including breast cancer, as highlighted by Rameshwari et al.[9] Moreover, *abnormal signalling of the RAS-MAPK pathway has been associated with benign precursors such as nevus sebaceous and SCAP*.[10]

4. Conclusion

In this study, the unusual presentation of SCAP in this case, coupled with its resemblance to SCC, emphasizes the importance of a meticulous diagnostic approach wherein dermoscopy and histopathology were key to identifying the benign nature of the lesion and directing the course of further management. The potential involvement of the MAPK pathway, shared with breast carcinoma, suggests a compelling molecular link that warrants further investigation into its oncogenic implications.

5. Conflict of Interest

None.

6. Source of Funding

None.

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