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Case Report

Pathology Section

Ectopic Hidradenoma Papilliferum of Axilla: A Case Report

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ABSTRACT

A benign adnexal tumour is a non cancerous growth located in the uterine tubes, ovaries, or near the uterus, including the surrounding connective tissue. While adnexal tumours can also be cancerous, they are predominantly benign. Moreover, adnexal masses can be non gynaecological, such as bladder diverticulum, appendicitis, apocrine gland tumours, sebaceous gland tumours, eccrine gland tumours and nerve sheath tumours. These tumours can occur at any age due to various causes. Some adnexal tumours are sporadic, while others may be linked to conditions such as Brooke-Spiegler syndrome and Birt-Hogg-Dubé syndrome. The tumours can be solid or fluid-filled and while some resolve spontaneously, others require therapy or surgical intervention. This case describes one such benign adnexal tumour: hidradenoma papilliferum, or papillary hidradenoma, which is a rare subcutaneous benign adnexal tumour of the apocrine glands in the anogenital regions. It typically occurs in females aged 30 to 50 years but is rarely reported in males. Hidradenoma papilliferum that is localised outside the anogenital region is referred to as ectopic. Ectopic presentations are primarily reported in the head and neck regions, especially on the external ear and eyelid, where modified apocrine glands are present. It is rarely reported in the nose, arm and axilla. This case report details a 53-year-old female who presented to the Surgery Outpatient Department (OPD) with a small swelling in her axilla accompanied by mild pain. Histopathological examination confirmed the nodular lesion as hidradenoma papilliferum. The location of the tumour was extremely rare and is often misdiagnosed as syringocystadenoma papilliferum and trichoepithelioma.

Keywords: Benign, Ectopic, Trichoepithelioma, Tumour

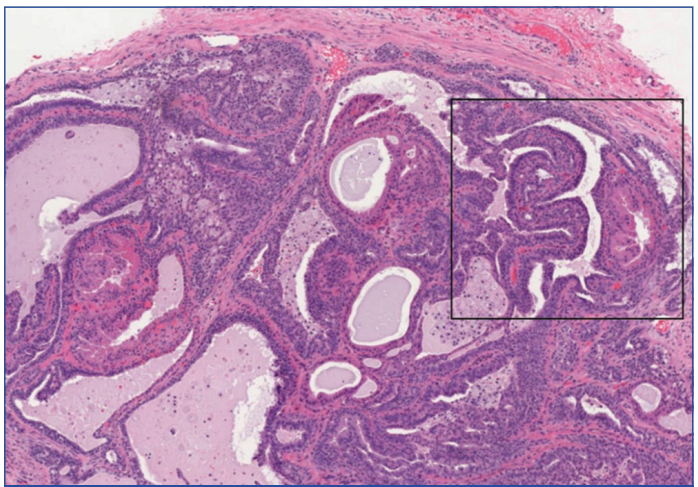
CASE REPORT

A 53-year-old woman presented to our hospital with a swelling in her right axilla. It began as a small swelling five months ago and progressively increased in size. She experienced mild pain for the last two months and during surgical excision, the size was measured at 4.5×3×1.5 cm, with adjacent adipose tissue, while the lesion itself measured 1.5×1×0.5 cm. Grossly, a small, well-circumscribed, smooth, tan-coloured, firm lesion was observed. [Table/Fig-1] shows the resected specimen.

[Table/Fig-1]: The gross image of the resected specimen showing smooth and tan-coloured tissue.

There was no history of irritation, bleeding, discharge, or ulceration. The size of the nodule was spherical and it was skin-coloured. She visited a local physician when the pain started and although she took the prescribed medications, the pain and the tumour did not subside. The patient's past medical and family histories were insignificant. She reported no history of hypertension, hypotension, diabetes, asthma, or allergies. The provisional diagnosis was thought to be reactive lymphadenitis. The patient underwent Fine Needle Aspiration Cytology (FNAC) but claimed to have misplaced the documents. Subsequently, she visited the Surgery Outpatient Department (OPD) of our hospital, where a clinical examination and surgical excision were performed.

Histopathological examination of the biopsy revealed a sharply circumscribed dermal nodule with a well-defined maze-like arborising proliferation. It displayed papillary, tubular and cystic structures lined with cuboidal myoepithelial and glandular cells. The papillae and micropapillae projected into the cyst cavity, featuring broad fibrous cores with a few plasma cells and lymphocytic aggregates. The broad tubules and fronds were lined by a double layer of epithelium. The outer layer consisted of columnar apocrine cells with eosinophilic cytoplasm, while the inner layer comprised cuboidal myoepithelial cells. The tumour may have been surrounded by fibrous tissue, termed a pseudocapsule. These microscopic features confirmed the diagnosis of hidradenoma papilliferum. Malignant changes were absent [Table/Fig-2].



[Table/Fig-2]: Histopathologic section showing features of hidradenoma papilliferum (H&E, 4x).

The surgeon obtained consent from the patient and performed the surgical excision; however, they later lost the opportunity to follow-up with the patient.

DISCUSSION

Werth described hidradenoma papilliferum in 1878 [1]. It is a benign adnexal tumour of the apocrine glands, typically presenting on the perineal, perianal and vulvar skin. Middle-aged women are primarily affected, although it occurs rarely in males [2]. These lesions are solid, isolated, circumscribed nodules that are firm, mobile and usually red or skin-coloured and they are mostly asymptomatic. They may be associated with symptoms such as pain, pruritus, ulceration, discharge, or bleeding. The average diameter is approximately 1 cm, but occasionally, it can exceed 1 cm. A case of giant ectopic hidradenoma papilliferum of the scalp has been recently recognised [2,3].

While malignant changes can occur in hidradenoma papilliferum, they have never been reported in ectopic hidradenoma papilliferum [3]. A similar case of a 61-year-old man affected by ectopic hidradenoma papilliferum of the axilla was published in English in 1966 by Ioannides [4]. Since then, no further records of ectopic hidradenoma papilliferum in the axilla have been found. The aetiology remains unclear, although some literature suggests that it generally affects sexually active females, indicating that sexual transmission factors may be a contributing cause [5]. It is treated by surgical excision.

Hidradenoma papilliferum originates from apocrine glands and predominantly affects the anogenital region of females. It can occur in females aged between 20 and 90 years [5]. Medical research indicates that oestrogen secretion contributes to tumour formation, which explains why females during and after puberty are more prone to this tumour [6]. Some researchers propose that it originates from ectopic vulvar breast tissue, glandular rudimentary structures and eccrine sweat glands [7]. Ectopic neoplasms are mainly distributed in areas with fewer apocrine or modified apocrine glands. Modified apocrine glands are found in the axilla, auditory canal, eyelid, nasal skin, scalp and face. Although rare, ectopic locations have been reported in the eyelids, nose, breast, ear, abdomen, chest, axilla, auditory canal, postauricular region, orbit and scalp [8]. The abundant concentration of apocrine glands in the axilla may account for the development of ectopic hidradenoma papilliferum in index patient. While the aetiology remains unclear, Human Papillomavirus (HPV) DNA has been identified in some patients. However, HPV is not involved in the pathogenesis of hidradenoma papilliferum [9]. Contrary to hidradenoma papilliferum, the prevalence of ectopic hidradenoma papilliferum is approximately equal in males and females [10]. The symptoms of ectopic hidradenoma papilliferum are similar to those of adnexal hidradenoma papilliferum [11].

Syringocystadenoma papilliferum originates from the sebaceous glands of Jadassohn and typically occurs in exposed areas, with secondary changes being recognised. Trichoepithelioma is a benign neoplasm of hair follicles commonly found on the face and scalp. Six rare cases of syringocystadenoma papilliferum occurring in unexposed areas, like axilla and perineum, along with two cases of trichoepithelioma, have previously been misdiagnosed as ectopic hidradenoma papilliferum. Secondary changes were absent in the slides of syringocystadenoma papilliferum that were mislabelled as hidradenoma papilliferum. Earlier reports indicated that both anogenital and ectopic hidradenoma papilliferum occur in the dermis and have no connection with the overlying epithelium. However, recent studies suggest that they can be connected via a duct to the overlying epithelium [12].

A tiny hidradenoma in a tumour-like cutaneous malformation of Jadassohn indicates the presence of two related or nosologically associated lesions [13]. Differential diagnoses include syringocystadenoma papilliferum, tubular apocrine adenoma, mammary-like gland adenocarcinoma, lipoma, hidradenocarcinoma papilliferum, papillary eccrine adenoma, basal cell carcinoma and abscess [14,15]. Ectopic hidradenoma papilliferum in the axilla can also be mislabelled as axillary lymphadenopathy.

It has been recorded that malignancies are rare in hidradenoma papilliferum and absent in ectopic hidradenoma papilliferum. According to some hypotheses, HPV may induce malignancies [6]. Histological appearances often show focal areas infiltrated by plasma cells and lymphocytes. Some authors suggest there is mixed differentiation between syringocystadenoma papilliferum and hidradenoma papilliferum [16]. The treatment is surgical excision and recurrence can occur due to incomplete tumour excision; however, recurrence of ectopic hidradenoma papilliferum has not yet been reported [10].

CONCLUSION(S)

Hidradenoma papilliferum is a rare benign neoplasm of apocrine sweat glands. The occurrence of ectopic hidradenoma papilliferum is infrequent. The abundance of sweat glands in the axilla is the probable cause of the ectopic hidradenoma papilliferum. The prognosis was good and surgical excision is the definitive treatment. Recurrence is extremely rare and typically occurs due to incomplete excision. Occasionally, syringocystadenoma papilliferum and trichoepithelioma are misdiagnosed as hidradenoma papilliferum, leading to instances of over-presentation. Breast carcinoma generally spreads to axillary lymph nodes, leading to its enlargement. Therefore, it is crucial to differentiate ectopic hidradenoma papilliferum in the axilla from carcinomas associated with the axilla, as the latter can be life-threatening.

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