Two Cases of Hidradenoma Papilliferum of the Nose

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Hidradenoma papilliferum is a rare benign neoplasm originating from the apocrine sweat glands. It has been reported primarily to affect Caucasian females, typically during the third to fifth decades of life. The lesions are usually solitary, well-demarcated papules or nodules covered with normal skin, and are generally less than 10 mm in diameter (1). This tumour is frequently asymptomatic, though may be associated with a pruritic, burning sensation or pain. Hidradenoma papilliferum usually occurs in a vulval, perineal or perianal location. When these tumours are not located in the anogenital area, they are termed ectopic hidradenoma papilliferum (2). We report here two cases of ectopic hidradenoma papilliferum of the nose.

CASE REPORTS

Case one

A 66-year-old female presented with a 5–6-year history of a slowly growing asymptomatic lesion on the nose. Physical examination revealed a 7 × 7 mm, soft dome-shaped papule with normal overlying skin (Fig. 1a). Routine laboratory investigations and physical examination were unremarkable. Differential diagnosis included an intradermal naevus, fibrous papule, or other benign neoplasm. A 3-mm punch biopsy showed a well-circumscribed tumour composed of complex patterns of anastomosing papillary structures without epidermal connection. The papillary structures were lined with a single or double layer of cuboidal or columnar cells. The outer layer was a layer of columnar cells with faintly eosinophilic cytoplasm, showing active decapitation (Fig. 1c). The inner layer consisted of small cuboidal cells with basophilic nuclei. On immunohistochemical study, gross cystic disease fluid protein-15 (G-CDFP15) was expressed and Periodic acid-Schiff (PAS)-stained secretory material was observed. However, the staining reaction for diastase-PAS (D-PAS) was negative. These findings are consistent with ectopic hidradenoma papilliferum, and the patient was referred to plastic surgery for complete excision of the lesion.

Case two

The second case was a 46-year-old woman who presented with a 2-year history of a gradually enlarging asymptomatic nasal cutaneous lesion. Physical exami-

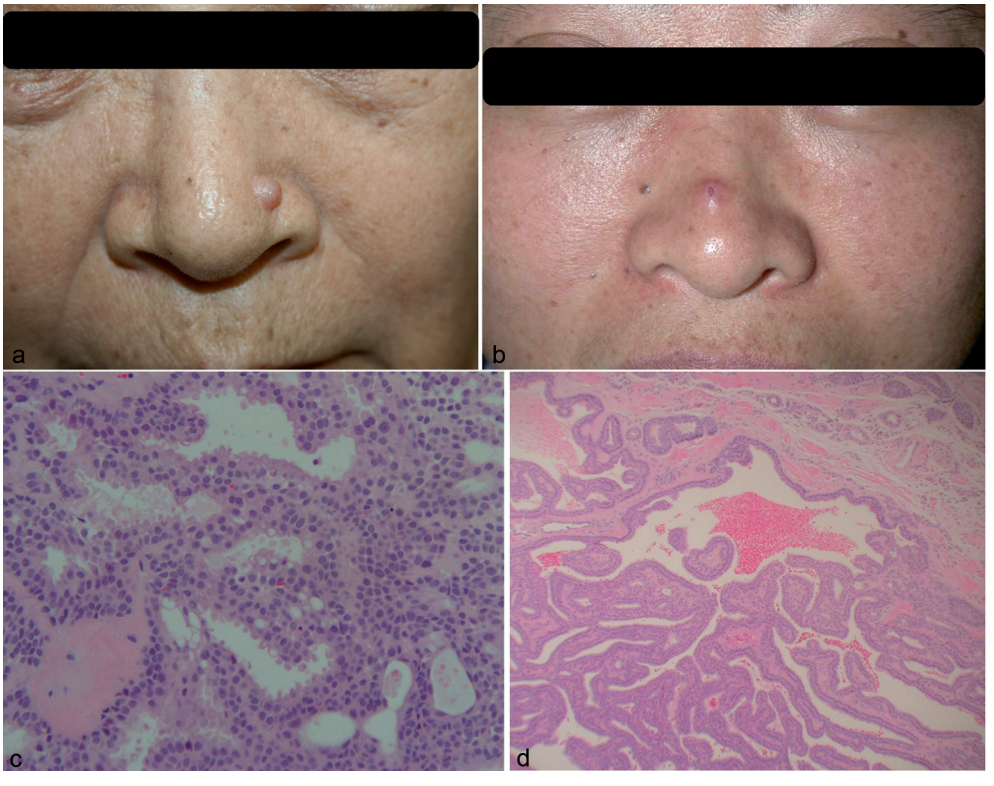

Fig. 1. (a) A 7 × 7 mm sized, soft, dome-shaped mass with normal skin colour on the left lateral side of nose (case 1). (b) A 10 × 10 mm sized, slightly erythematous, dome-shaped nodule with central ulceration (case 2). (c) The lumen of the papillary glandular structure was lined with two layers of columnar and cuboidal cell with active decapitation (case 1, haematoxylin and eosin (HE) × 200). (d) Complex anastomosing papillary glandular structure (case 2, HE × 40).

nation revealed a well-demarcated, slightly erythematous, dome-shaped nodule with central ulceration, 10 mm in diameter (Fig. 1b). Routine laboratory investigations and physical examination were unremarkable. Clinical differential diagnosis included an epidermal cyst, basal cell carcinoma, keratoacanthoma or another benign adnexal tumour. A diagnostic 3-mm punch showed a well-encapsulated cystic neoplasm consisting of an anastomosing papillary, glandular structure without epidermal connection (Fig. 1d). The lumen of the papillary glandular structure was lined with two layers of columnar and cuboidal cells. Active decapitation was observed in the outer layer of the lining cells. The secretory material was positive for PAS staining, but the staining reaction for D-PAS was negative. Immunohistochemically, the epithelial cells expressed G-CDFP15. These findings were consistent with a diagnosis of ectopic hidradenoma papilliferum.

DISCUSSION

Apocrine glands are heavily concentrated in the anogenital, axillary, and peri-umbilical regions, with most cases of hidradenoma papilliferum located in the anogenital region. However, there are heterotopic apocrine glands on the scalp and face, and modified apocrine glands are found in the external ear canal, eyelids and breasts. These heterotopic, modified apocrine glands are thought to be the potential origin of ectopic hidradenoma papilliferum (2). The main clinical differences between ectopic hidradenoma papilliferum and anogenital hidradenoma papilliferum are body site, sex and age of onset. Ectopic hidradenoma papilliferum occurs on non-anogenital sites in an older group of patients, with an equal incidence in males and females (2). Anogenital hidradenoma papilliferum occurs almost exclusively in females (2).

It is not clear whether ectopic hidradenoma papilliferum is indeed a true variant of hidradenoma papilliferum or should be considered as an apocrine papillary cystadenoma. We favour the former. Apocrine papillary cystadenoma consists of a large unilocular or multilocular cystic space with focal papillary proliferation (3). Its cystic characteristic, focal papillary proliferation, nuclear atypia and mitotic activity differ from hidradenoma papilliferum (3, 4). Clinically, unlike the skin-coloured nodule of hidradenoma papilliferum, apocrine papillary cystadenoma presents as a translucent, blue or purple cystic nodule (3). In addition, hidradenoma papilliferum forms a fairly well-demarcated nodule in the dermis, and consists primarily of typically double-layered epithelium-covered papillary processes. Occasionally, the lining is only one cell thick, and focal sebaceous differentiation may be a feature (3). The clinical and histological findings of our two patients are consistent with ectopic hidradenoma papilliferum. Worldwide, only 19 cases of ectopic hidradenoma papilliferum have been reported in the English language literature, located on the thigh (1 case), external auditory canal (1 case), eyelid (3 cases), eyebrow (1 case), axilla (2 cases), face (5 cases), head (1 case), back (2 cases), upper limb (1 case), auricular area (1 case), and nose (1 case) (2, 5–7). Thus, only one case on the nose has been reported previously among 19 cases of ectopic hidradenoma papilliferum. Given that the face was the most common previously reported site for ectopic hidradenoma papilliferum, we suggest that, in cases of a skin-coloured papule on the facial region, ectopic hidradenoma papilliferum should be included in the clinical differential diagnosis.

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