

Case Report

Anterior Neck Hidradenoma Papilliferum– Rare Case Report in Khartoum Teaching Hospital, Sudan

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Background: Hidradenoma papilliferum is a rare tumor that occurs almost exclusively in females on the anogenital area. Rare cases of ectopic (nongenital) hidradenoma papilliferum have been described. The lesions usually present as an asymptomatic for a long time before resection.

Case report: A 52-year-old man presented to Khartoum teaching hospital on July 2017 with fungated anterior neck a painless swelling for six months. The nodule was excised and the histology revealed a hidradenoma papilliferum. The diagnosis and treatment of hidradenoma papilliferum is possible with surgical removal and histopathological evaluation of nodules

Conclusion: When an adult woman presents with a nodular lesion in the anogenital area, sexually transmitted diseases and other benign and malignant vulvar lesions, as well as malignant transformation is very rare but, should be kept in mind; however, because it has been reported and long-term clinical follow-up is suggested.

Keywords: Hidradenoma papilliferum; Excisional biopsy; Sudan.

Introduction

Hidradenoma papilliferum is a slow-growing benign adnexal tumor with apocrine differentiation, and for some authors it can be considered an analog of intraductal papilloma of the breast [1]. The tumor primarily affects vulvar, perineal and perianal skin of middle-aged women with rare cases being reported in other skin localizations (ectopic hidradenoma papilliferum) [2,3]. To our knowledge, only a perianal, hidradenoma papilliferum has been described in a man [4]. The clinical presentation as well as the pathologic features, treatment, and prognosis are similar in both forms. It usually presents as a slow-growing, solitary, asymptomatic skin colored or red nodule less than 1 cm in diameter [5]. However, a giant ectopic hidradenoma papilliferum on the scalp has been recently described [6]. The most common site of ectopic hidradenoma papilliferum is the head and neck. The diagnosis can be made only by histopathological examination since they clinically mimic other cutaneous neoplasms.

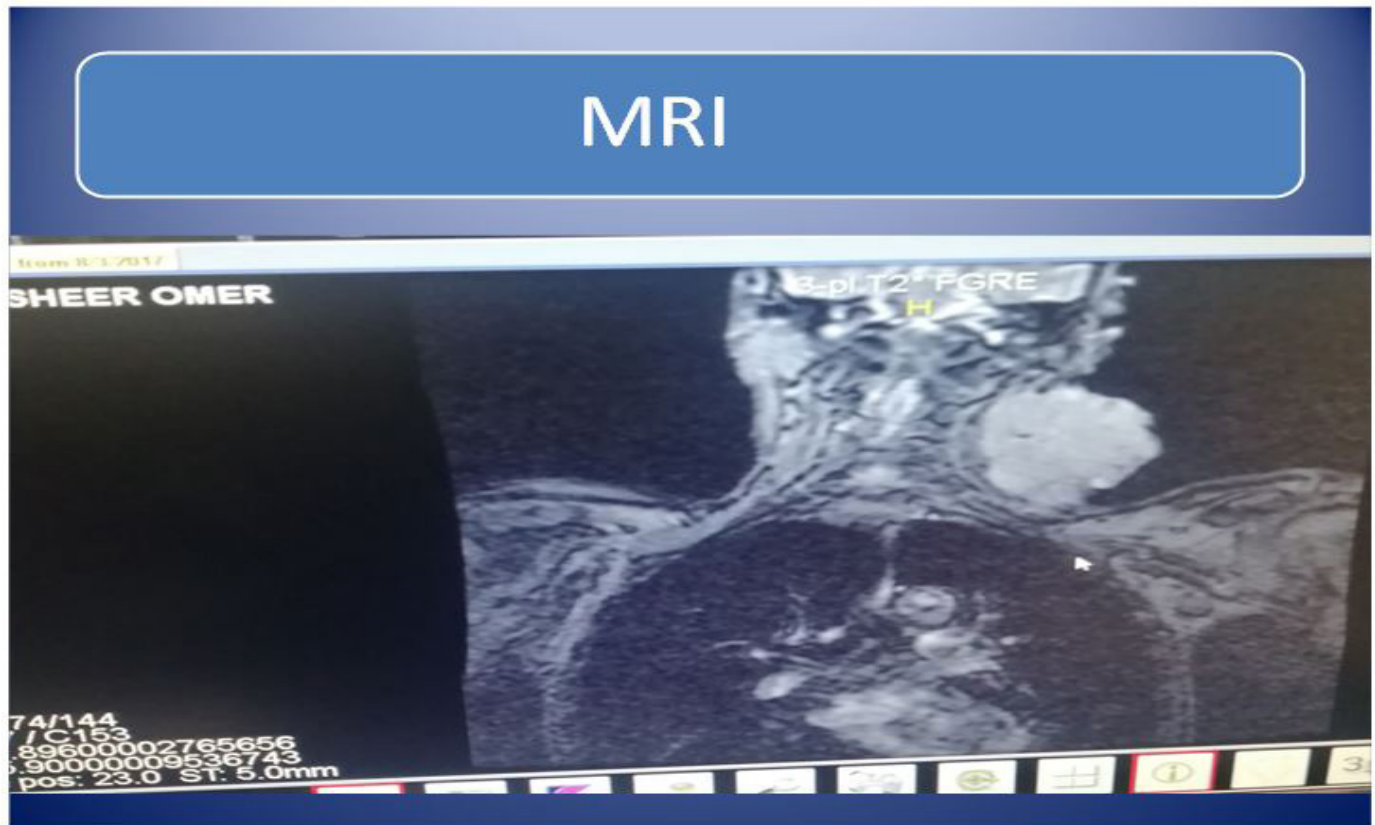
Case Report

A 52-year-old man presented to Khartoum teaching hospital

on July 2017 with fungated anterior neck a painless swelling for six months. Actually, the story started 6 months before the patient attends to us. complaining of anterior neck swelling started small in size increased with time. referred to our unit from Omdurman teaching hospital and they diagnosed him as cervical lymphadenopathy based on neck us. We saw the patient in the refer clinic with his neck covered by plaster after we take full history at time of examination, he said the lesion may bleed unfortunately the patient develop bleeding large amount from the site of the lesion we compress the site and start resuscitation. after that when we review his investigation there was MRI reported as (left parotid pleomorphic adenoma). Biopsy result show un specific skin lesion .so plan finally excisional biopsy was done; Consistent with Hidradenoma papilliferum.

Discussion

Hidradenoma papilliferum is originated in the apocrine glands, which are mainly concentrated in the anogenital region, axillae, and periumbilical areas. The tumor occurs mainly in those areas with ectopic localization being rarely reported [2]. The distribution of the ectopic forms corresponds to the areas con-



taining heterotrophic and modified apocrine glands. According to a medline search, only 20 reports of ectopic hidradenoma papilliferum have been described in the English language and only 3 were localized to the eyelid [7–9]. Independently of being typical or ectopic the tumor occurs mostly in white women. However, in contrast to anogenital hidradenoma papilliferum, nearly one-half of the patients with ectopic hidradenoma papilliferum are men [2]. The head and neck are the most frequent localization for the ectopic presentation, mainly the eyelid and external ear, where modified apocrine glands (Moll and ceruminous glands) are found normally [2, 7]. Sometimes ectopic apocrine tumors are also found in the scalp within lesions such as the nevus sebaceous of Jadassohn. There are 4 reported cases of hidradenoma papilliferum on the head and neck region in males [7]. Other ectopic localizations included arm, thigh, back, and nipple [2, 7, 10]. The age range reported is between 8 to 78 years [7]. The clinical presentation of both forms is similar to most lesions, being asymptomatic and growing for a long time before excision. Pain, pruritus or, ulceration can occur. Like other adnexal skin tumors, they clinically mimic other neoplasms such as basal cell carcinoma (as in our case report) and spinocellular carcinoma. Thus, histological examination is required for the correct diagnosis. Histologically the tumor is characterized by a cystic space containing eosinophilic material and papillary folds projected from the cyst wall. Tumor epithelium is composed by a basal layer of cuboidal cells and a luminal layer of larger columnar cells showing decapitation secretion [11]. The epidermis may be normal, acanthotic, or ulcerated and may sometimes show continuity with the overlying epithelium [12, 13]. In some cases, the tumor displays a histopathology similar to syringocystadenoma papilliferum since they are closely related tumors which originate from apocrine glands [14]. Aggregates of lymphocytes and plasma cells have been described in the stroma of ectopic lesions [2]. Some authors considered the presence of focal areas infiltrated by plasma cells and lymphocytes as a sign of a mixed differentia-

tion between hidradenoma papilliferum and syringocystadenoma papilliferum [7]. A report of an ectopic hidradenoma papilliferum with sebaceous differentiation has been documented [8]. Other histopathological differential diagnosis includes tubular apocrine adenoma and clear cell (apocrine) adenoma. The prognosis is good with local excision being the treatment of choice. Recurrence of the lesions is attributed to incomplete excision of the primary tumor and there is no report of recurrence for the ectopic form [2]. Malignant transformation in anogenital hidradenoma papilliferum has been documented (intraductal carcinoma resembling apocrine carcinoma and invasive Aden squamous carcinoma) but not in the ectopic presentation [2, 15, 16]. It is speculated that HPV may play a role in inducing malignancy, but the association still needs to be proved [17]. We presented a new case of ectopic hidradenoma papilliferum with features of a mixed differentiation arising in the eyelid.

Conclusion

When an adult woman presents with a nodular lesion in the anogenital area, sexually transmitted diseases and other benign and malignant vulvar lesions, as well as Hidradenoma papilliferum should be kept in mind. Patient history and clinical findings are not specific for hidradenoma papilliferum and surgical removal of the lesion and its histopathological evaluation are required to make a definitive diagnosis.

Declarations: Ethics approval and consent to participate Informed consent was obtained from the patient after the study authors fully explained why his condition is being submitted as a medical case report.

Consent for publication: Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conflicts of Interest: None

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