Poroid Hidradenoma Presenting as a Malignant Neoplasm

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Abstract Poroid hidradenoma is a rare adnexal neoplasm that presents with slow-growing solitary dermal or subcutaneous nodules with pink, red, or blue coloration. Malignant transformation has been reported in about 1%; however, clinical and histologic characteristics may present similarly with malignant neoplasms. A 60-year-old, Filipino, female presented with a 1-month history of a solitary, brownish-black papule on the right cheek progressing to a translucent friable nodule with associated bleeding after minimal trauma. The patient was initially assessed as a case of basal cell carcinoma due to its rapid growth. Hence, the patient was referred to a dermatologic surgeon for biopsy. Poroid hidradenoma was confirmed histologically and was managed with local excision. Poroid hidradenoma presenting with rapid growth is a very rare occurrence.

Keywords: Adnexal neoplasm, malignancy, poroid hidradenoma

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Submitted: 23-01-2024, Revised: 21-04-2024, Accepted: 22-04-2024, Published: 31-05-2024.

INTRODUCTION

Poroma is a benign eccrine neoplasm initially described by Jones J et al. in 1956 and is further subclassified into four entities: poroma, hidroacanthoma simplex, dermal duct tumor, and the recent addition, poroid hidradenoma.^[1] It is a relatively new variant hence there are limited reported cases worldwide. From 2011 to 2021, the Philippine Dermatological Society documented only one case of poroid hidradenoma.^[2] Poroid hidradenoma is a rare case among all skin adnexal tumors being the newest addition to the poroma family. In a study by Kang et al., they observed that the most common benign skin adnexal tumors were clear cell hidradenoma and pilomatricoma, followed by eccrine poroma.^[3] We present a rare case of poroid hidradenoma in the Philippines demonstrating an atypical presentation of rapid growth on the unusual site on the cheek.

Access this article online	
Quick Response Code:	
	Website: https://journals.lww.com/jpds
	DOI: 10.4103/JPDS.JPDS_6_24

CASE REPORT

We report a case of a 60-year-old Filipino female who consulted her dermatologist with an asymptomatic, solitary, brownish-to-black papule with a dimension of approximately 0.5 cm \times 0.5 cm on her right cheek. The patient denied the application of topical medication and cosmetics. There was no known history of trauma and chemical exposure. However, the patient claimed to have recreational chronic sun exposure and denied the use of sunscreen. No systemic symptoms were noted. Over time, the patient noted a sudden increase in the dimension of the lesion progressing to an erythematous to brownish translucent nodule, with some scaling and crust, associated with 3/10 pruritus, with noted bleeding following minimal trauma. There was no pain or discharge noted. The patient was hypertensive and maintained on losartan

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How to cite this article: Olitoquit KD, Gutierrez-Villaroman EM, Gulmatico-Flores Z. Poroid hidradenoma presenting as a malignant neoplasm. J Philipp Dermatol Soc 2024;33:17-20.

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50 mg and amlodipine 5 mg. Family history, obstetrical history, and review of systems were unremarkable. Physical examination revealed a solitary, nontender, firm, well-circumscribed, dome-shaped, translucent, erythematous to brownish nodule, with telangiectasias, measuring 1 cm \times 1 cm \times 0.5 cm on the right maxillary area of the face [Figure 1]. Regional lymphadenopathy was absent. Dermoscopy showed a vascular pattern of leaf- and flower-like features, interlacing white cords, and vascular blush, as shown in the Figure 2. The patient was initially assessed as a case of basal cell carcinoma versus Merkel-cell carcinoma and was immediately referred to a dermatologic surgeon for an excision biopsy.

Histopathological examination on scanning view revealed an asymmetrical nodulocystic tumor. The epidermis showed orthohyperkeratosis while the dermis showed a solitary, large, slightly asymmetrical neoplasm showing irregular borders without epidermal connection. There was a largely cystic component with small foci of tumor lobules within the cyst cavity. The cyst wall and tumor nests were composed of monomorphous small dark cells (poroid) and large pale (cuticular) cells, with associated intercellular vacuolization (early ductal differentiation) and ductal structures. Most have nuclear polymorphism, hyperchromatism, and crowding with mitotic figures, some of which were atypical. Areas of necrosis were seen within the tumor and cystic space, with few foci of shadow cells. There was no epidermotropism, neural or perineural and lymphatic space involvement, or satellite metastasis. Immunohistochemical staining showed negative carcinoembryonic antigen (CEA). Alcian blue was also done and showed abundant basophilic material between the tumor and connective tissue.

A wide excision of the lesion with 4 mm margins was done and revealed the histopathologic findings

mentioned. An elliptical excision was performed using an aseptic technique and hemostasis was ensured. Primary closure using absorbable deep sutures and running nonabsorbable superficial sutures was done [Figure 3]. The patient was also given postoperative medications such as cefalexin 500 mg/cap given every 8 h for 1 week, celecoxib 400 mg/tab once daily for 3 days, Vitamin C 500 mg/tab once daily for 1 month, and mupirocin 2% ointment twice daily for 2 weeks. Follow-up was done after 2 weeks for the removal of sutures with no noted signs of infection, dehiscence, hematoma, or suture reaction. Subsequent follow-up 2 1/2 months later was done through teledermatology with no reported recurrence or any subjective complaints such as pain, bleeding, or discharge. Periodic follow-up every 12 months for 2 years was advised to the patient.

DISCUSSION

Poroid hidradenoma is a rare cutaneous adnexal tumor and the newest addition to the group of poromas, derived from eccrine sweat ducts, with both solid and cystic components confined entirely to the dermis, with no connection to the epidermis.^[3] There have been 50 cases reported in the literature. Age varies between 13 and 86 years with a peak incidence in the sixth and seventh decade. Several articles have stated its equal male-to-female ratio, however, based on our literature review, it occurs more frequently in males. Some studies have hypothesized a predilection of poroid hidradenoma in Asian population due to its increased incidence in Korean and Japanese nationality. Poroid hidradenoma presents as a subcutaneous nodule with pink, red, or blue coloration and is frequently found on the trunk.^[4] The usual size ranges from 1 to 2 cm. Tenderness of the tumor is an occasional symptom. Other



Figure 1: Preoperative photo (left), presenting with a solitary, dome-shaped, firm, well-demarcated, erythematous nodule on the right maxillary cheek. Postexcision photo (right), wound closure done using simple running suture

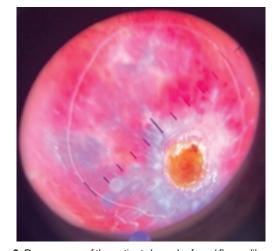


Figure 2: Dermoscopy of the patient shows leaf- and flower-like pattern, interlacing white cords, and vascular blush

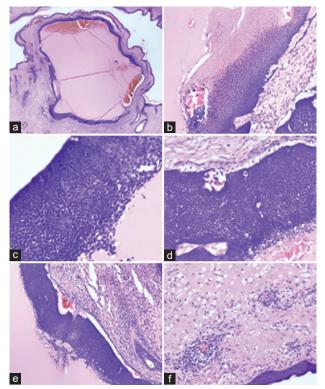


Figure 3: Biopsy of a solitary nodule on the right maxillary area of the face. (a). A solitary neoplasm with no epidermal connection to the overlying dermis. Hematoxylin and Eosin. (b-d) The cyst wall and tumor nests are composed of shadow cells, pale round cells, dark monomorphic cuboidal cells, and ductal structures. (e) Clefts are noted between the stroma and the tumor island. (f) Areas of necrosis are seen within the tumor. The surrounding stroma shows proliferation of small and dilated blood vessels, marked edema, and mixed infiltrates of lymphohistiocytes and neutrophils. (a) Scanning magnification, (b) 10x magnification, (c) 20x magnification, (d and e) 10x magnification, (f) 20x magnification

less frequent sites include the axilla, trunk, and extremities.^[5] Several case reports showed the typical duration of lesions ranging from 5 months to 8 years and the majority are treated with excision. Chaudhari and Rajput reported a case of a gradually increasing swelling in the chest wall for 5 months which was noted to be the shortest duration seen in the literature. To date, a rapid onset of 1 month associated with bleeding as seen in our case is a rare occurrence owing to the typical chronic duration of poroid hidradenoma. Malignant forms of poromas may sometimes present with a rapidly expanding mass associated with bleeding. However, local ulceration and lymph node involvement should also be noted. There have been reported cases arising from preexisting benign poromas.^[6]

The clinical presentation may present similarly with cutaneous neoplasms such as basal cell carcinoma, Merkel-cell carcinoma, nodular melanoma, and inflammatory lesions such as pyogenic granuloma. Hence, a thorough clinical history and physical examination along with histological confirmation of tissue is indeed crucial for its diagnosis. Excision biopsy may be used as a diagnostic method in cystic lesions in some reports in the literature.^[7]

Our case was diagnosed based on routine H and E stains. Diagnostic uncertainty may arise due to its similarities with malignant neoplasms and other benign tumors. Histologic confirmation can be aided by immunohistochemical stains such as CEA, being a marker for cutaneous adnexal tumors.^[8] CEA was done to highlight the ductal structures of the tumor, however, showed negative results. Poroid hidradenoma presents as a dermal nodule with no epidermal connection and is composed of poroid and cuticular cells with eccrine differentiation. Epithelial membrane antigen, a marker for eccrine differentiation, can also be requested, however, this was not done.^[8] Alcian blue was requested to detect mucin to rule out malignant transformation. However, strong stromal mucin as seen in two-thirds of sweat gland tumors and adenocarcinomas of eccrine and apocrine origin has not been established as a reliable indicator for malignancy. Histologic features suggestive of malignancy such as cellular atypia and increased mitotic figures may also be observed in atypical cases of poroid hidradenoma. In addition, areas of necrosis (atypical poroid hidradenoma) may be seen distinguishing them from hidradenoma of apocrine origin.^[7] Although histopathologic findings indicative of malignancy were present, the neoplastic processes were confined within the tumor with no evidence of invasion. Nevertheless, the absence of (1) circumscription with infiltrative borders; (2) vascular and neural invasion; and, (3) lymph node metastases will support its benign process.^[8]

Dermoscopic findings would show a vascular pattern presenting with polymorphic, glomerular, linear-irregular, looped, or hairpin. Other features suggestive of poromas are interlacing white cords, vascular blush, and leaf- and flower-like which were noted in our case.^[9]

There are no reports of malignant cases of poroid hidradenoma; however, several reports suggest malignant transformation rate of 1%.^[8] Due to its rare occurrence, there is limited data about its prognosis. Poroid hidradenoma is thought to have a good prognosis since recurrence is uncommon. There was one reported case in 2018 where the appearance of the same tumor was noted on the area where surgical excision was done 8 years prior.^[10] However, this was not substantiated by data. In the case reported by Whitmore *et al.*, excision biopsy was stated to be curative and was done with an uneventful postoperative course and revealed no evidence of recurrence 4 years after the initial diagnosis. Moreover, a follow-up for 6–12 months was suggested similar to our patient. Narrow margins are also recommended given

the unconfirmed case of recurrence. Nevertheless, not much has been investigated to support its treatment guide.^[7]

CONCLUSION

In this report, we add to the literature a case of poroid hidradenoma in a Filipino female presenting with rapid growth and bleeding with minimal trauma which could be mistaken for a malignant neoplasm. To our knowledge, this is the first case of a benign poroid hidradenoma who presents with a rapid onset. Poroid hidradenoma being a relatively rare case and a new subclassification of the poroma group is now being known due to the increasing reported cases yearly.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship Nil.

Conflicts of interest

There are no conflicts of interest.

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