

Case and Review

Poroid Hidradenoma: Case Report and Comprehensive Review of the Literature

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Keywords

Poroid hidradenoma · Eccrine · Sweat gland tumor · Adnexal neoplasm

Abstract

Poroid hidradenoma (PH) is a rare benign adnexal tumor of eccrine differentiation. It is the rarest of the four described variants of poroid neoplasms. PHs characteristically share a hybrid of the architectural features of the hidradenoma, namely, tumor cells are entirely intradermal with both solid and cystic components, and the cytologic characteristics of the poroid neoplasms, containing predominantly poroid and cuticular cells. Many published reports of PH since its original discovery in 1990 state that “very few” cases of PH can be found in the literature. Here, we have identified a total of 75 published accounts of PH, including the case presented here, as well as the associated patient demographics, lesion characteristics, treatment, and outcomes. We suggest that while uncommon, PH is likely not exceptionally rare and could be an underreported diagnosis.

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Introduction and Historical Background

Cutaneous adnexal tumors are benign or malignant growths of one of the four normal skin appendages: hair follicles, sebaceous glands, apocrine glands, and eccrine glands. They are classified based on morphological differentiation toward one or more of the listed adnexal structures [1]. Sweat gland tumors, both eccrine and apocrine, are generally considered an

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uncommon group of benign adnexal neoplasms with a wide histological spectrum [2]. One subcategory of sweat gland tumors is the poroid tumors, including hidroacanthoma simplex, classic poroma, dermal duct tumor, and poroid hidradenoma (PH) [3]. Poroid tumors are characterized by poroid cells which are small round, monomorphous cells with small amounts of cytoplasm that exhibit ductal differentiation (Fig. 1). Here, we further describe the poroid hidradenoma which displays a hybrid of the architectural features of a hidradenoma, namely, tumor cells are entirely intradermal with solid and cystic components and nodular deep expansile growth, but the cytologic characteristics of a poroid neoplasm, meaning they contain predominantly poroid and cuticular cells akin to the small round cells that make up normal eccrine ducts [4, 5].

Although originally described in 1990 [6], the benign adnexal neoplasm PH is understood to be a rare diagnosis in the literature. Therefore, currently no official guidelines exist for its management, and minimal information is accessible regarding PH's true incidence, natural clinical history, or treatment outcomes. These cutaneous tumors develop primarily from eccrine sweat glands and represent the newest variant of eccrine gland neoplasms [5]. They typically present as a painless, well-circumscribed solitary nodule or papule that often grows over the course of months to years and can have associated itching or mild tenderness [4, 7, 8]. PH can affect a wide range of ages, but with a predilection for the sixth to seventh decade [4]. It has been reported that PHs were most frequently located on the head and neck [7]. However, a systematic review outlining all published cases between 1990 and 2017 [8] demonstrated that nearly 40% of published PHs were on the head and neck, and an equal number (~40%) of cases were situated on the limbs. The case presented here is that of a PH on the posterior shoulder in a 67-year-old woman. Additionally, we have included a systematic review of the literature describing PH including its presentation, patient demographics, treatment, recurrence, and outcomes (see Table 1 for a comprehensive list of all published reports of PH).

Case Report

The patient is a 67-year-old woman with an extensive past medical history including (but not limited to) diabetes, cirrhosis, hyperlipidemia, hypertension, hypothyroidism, multinodular goiter, memory loss, chronic joint and low back pain, and iron and vitamin deficiencies secondary to gastric bypass surgery, presenting with a right posterior shoulder lesion. The patient first noticed the lesion 5 years prior and initially described it as a flat, red, scaly lesion. Over the course of 1 year, the lesion increased in size and began to have some concurrent drainage. Over the preceding 5-months, the lesion also changed in color from red to purple. The patient did not have complaints of itching and burning at the site of the lesion and denies any associated pain or weight loss. There was no personal or family history of skin cancer.

On physical exam, a 5-mm diameter, light brown, irregular but well-circumscribed lesion was observed on the right posterior shoulder (Fig. 2). Shave biopsy to the level of the dermis was performed and specimen was sent for pathology in formalin. The skin fragment measuring 0.9 × 0.7 × 0.3 cm revealed an adnexal neoplasm extending to the peripheral margin and to biopsy base. Histological evaluation found a dermal predominant adnexal neoplasm with features of both poroma and hidradenoma. The tumor did not display significant nuclear pleomorphism, significantly elevated mitotic rate or infiltrative growth pattern (Fig. 3 and 4). Although the histological features suggested a nonmalignant PH, the patient's age, the potential for tumor heterogeneity within adnexal neoplasia, and positive margins of the biopsy, conservative re-excision was recommended. Surgical excision was then performed with 1 cm margins for a total excision area of 3.9 × 1.7 × 1.0 cm. Residual lesion contained PH with

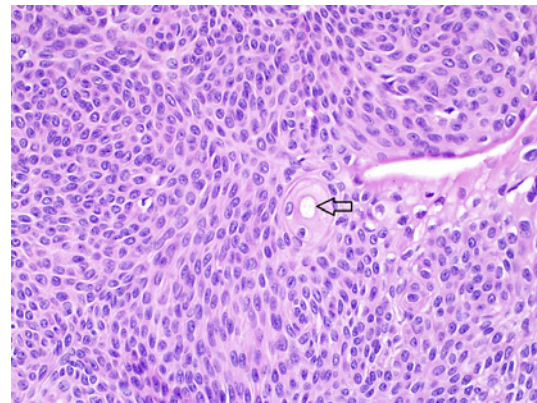


Fig. 1. H&E stain, ×400. Typical poroid cells with small round to streaming uniform nuclei, pink cytoplasm, and focal ductal lumina (arrow).

negative margins. Within the wide excision specimen, there were some atypical features including focally increased mitotic rate, and scant necrosis but not fulfilling criteria for malignancy. Overall, the lesion remained well-defined and symmetrical without infiltrative borders.

Methods

A systematic review of the literature was performed to identify all published reports of PH. PubMed and Google Scholar databases were used to identify papers. The search term “poroid hidradenoma” in PubMed yielded in 41 results. Papers were limited to the English language. Titles and abstracts screened to identify papers with confirmed PH diagnoses based on histology. Seven papers were excluded based on titles or abstracts not related to true PH cases (such as other poroma subtypes, hidradenomas of apocrine differentiation, or malignant sweat gland tumors). To ensure all published cases of PH, they were captured in this systematic review, all cited articles (within the 34 articles identified) were also located if not a result of the original search. Articles were further analyzed for the following parameters: patient age, patient sex, location of lesion, duration of lesion, associated symptoms, gross appearance of lesion, diagnostic workup results, differentials stated, treatment, and recurrence of lesion. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see <https://doi.org/10.1159/000531052>).

Systematic Review

A total of 32 articles and 74 cases were identified that met the inclusion criteria plus the 1 patient described here (see Table 1). We found that PH affected a wide range of ages (13–81 years) with a mean age of 57 years. The male:female ratio was 1.28:1. PH has also been found at a variety of locations, involving the hands, feet, limbs, trunk, head/neck including face and scalp, back, chest/breasts, abdomen, buttock, and vulva. Signs and symptoms of PH were also broad, but often presented as a slow-growing, painless subcutaneous mass, sometimes with overlying normal pink skin but frequently associated with a dark and/or bluish discoloration. The cystic components of PH can impart a blue hue to the tumors, a characteristic known as Tyndall phenomenon [11, 12, 37]. Treatment was generally excision, and recurrence was found to be uncommon.

Table 1. Systematic review of published cases of PH

Age, years	Sex	Location	Duration	Symptoms/examination/differentials	Treatment	Recurrence	References
46	F	Vulva	1 yr	Gradually enlarging, dark lesion causing dyspareunia for 2 w Size: 1.4 × 1.1 × 1.0 cm	Excision	Unknown	Alowami [9]
65	M	Middle finger, flexor surface	2 yr	Tender, soft, moveable, nodule covered in normal skin Size: 1.1 × 0.9 × 0.2 cm Differential: sebaceous cyst	Excision	None at 9 m	Arwyn-Jones, et al. [10]
Mean age, years (range): 55.5 (18–86)	M:F	Trunk 14 Upper limb 7 Lower limb 5	Mean duration (range): 3.9 years (0.1–10 years)	Nonspecific cutaneous and/or subcutaneous nodules (74% of cases), polypoid nodules (7%), recurrent cysts (19%); ulceration (14%), erythematous (90%), pigmented (5%), bluish (5%), pain (4.5%) Size: ≥1 cm: 17; <1 cm: 10	Unknown	Unknown	Battistella, et al. [11]
15: 18	Head/neck 5 Other 3			Differential: dermatofibrosis, epidermal cyst, benign adnexal tumor, dermal melanocytic nevus, basal cell carcinoma, lipoma, angiolipoma (3/34 cases were PH coexisting with dermal duct tumor)			Case series (34 cases)
78	M	Cheek	2 yr	Skin-colored, asymptomatic lesion Differential: BCC	Biopsied, no further treatment stated	Unknown	Chen, et al. [12] Liu, et al. [13]
64	M	Scalp	5 yr	Painful and hemorrhagic, dark-colored nodule Size: 0.6 × 0.6 cm Differential: pyogenic granuloma	Biopsied, no further treatment stated	Unknown	Chen, et al. [12] Liu, et al. [13]
45	M	Temporal area	2 yr	Asymptomatic, skin-colored lesion Size: 1.2 × 1.2 cm Differential: trichilemmal cyst	Biopsied, no further treatment stated	Unknown	Chen, et al. [12] Liu, et al. [13]

Table 1 (continued)

Age, years	Sex	Location	Duration	Symptoms/examination/differentials	Treatment	Recurrence	References
64	M	Chest	6 mo	Painless, bluish nodule Size: 3.0 × 2.0 cm Differential: pyogenic granuloma	Biopsied, no further treatment stated	Unknown	Chen, et al. [12] Liu, et al. [13]
65	M	Abdomen	3–4 mo	Asymptomatic, skin-colored lesion Size: 1.0 × 1.5 cm Differential: soft fibroma (PH coexisting with eccrine poroma)	Biopsied, no further treatment stated	Unknown	Chen, et al. [12] Liu, et al. [13]
69	M	Buttock	6 yr	Asymptomatic, brown to black-colored lesion Size: 1.5 × 1.5 cm Differential: seborrheic keratosis (PH coexisting with hydroacanthoma simplex)	Biopsied, no further treatment stated	Unknown	Chen, et al. [12] Liu, et al. [13]
55	F	Upper back	7 yr	Irregularly shaped, soft, brownish pigmented nodule with several black protruding papules on the surface Size: 1.5 × 2 cm Differential: nevus (PH coexisting with eccrine poroma)	Unknown	Unknown	Chiu, et al. [14]
36	M	Buttock	1 yr	Enlarging, soft, tender, skin-colored nodule Size: 1.3 × 0.9 cm	Excision	Unknown	Cho, et al. [15]
66	F	Breast	2 yr	Dark bluish dome-shaped nodule Differential: papillary neoplasm Mammography showed well-circumscribed round isodense mass Ultrasound showed oval complex cystic and solid mass abutting the dermis	Excision	Nil at 1 yr	Choi, et al. [16]

Table 1 (continued)

Age, years	Sex	Location	Duration	Symptoms/examination/differentials	Treatment	Recurrence	References
35	M	Posterior distal thigh	3 yr	Enlarging, tender, soft, moveable mass, originally covered with normal skin that changed to a gray blue color Size: 4.3 × 3.1 cm Differential: fibrolipoma Ultrasound showed nodular, anechoic soft tissue mass, with hyperechoic spots in its contents and multiple internal septations	Excision	None at 2 wk, long-term unknown	Delfino, et al. [17]
34	M	Chest, below nipple	18 mo	Tense, cystic, painless swelling Size: 5.0 × 5.0 × 3.0 cm Ultrasound showed heterogeneous mass with anechoic and cystic components Differential: cyst with malignancy	Excision	Unknown	Dravid, et al. [18]
65	M	Leg, posterior aspect	10 yr	Red and blue dome-shaped nodule that slowly enlarged over 6 m Size: 2.6 × 2.2 cm	Unknown	Unknown	Goksugur & Yilmaz [19]
66	F	Superior most portion of scalp	Not stated	Mobile, firm, nontender nodule Differential: lipoma, sebaceous cyst	Excision	Unknown	Grant & Awad [20]
77	F	Elbow		Slightly elevated, tender, reddish nodule Size: 2.7 × 2.4 cm Differential: epidermal cyst (previous lesion had been excised at the same site and had recurred, enlarging over years)	Excision	None at 1 yr	Hoshida, et al. [21]
61	M	Back	1 yr	Enlarging, painless, soft mass covered with pink skin Size: 3.0 × 2.5 cm Differential: cystic lesion Ultrasound showed oval-shaped hypoechoic and cystic mass with clear borders	Excision	None at 1 yr	Ichioika, & Yamada [22]

Table 1 (continued)

Age, years	Sex	Location	Duration	Symptoms/examination/differentials	Treatment	Recurrence	References
43	F	Scalp	Unknown	Nodule - no further description	Unknown	Unknown	Kazakov, et al. [23]
78	M	Scalp	Unknown	Basal cell carcinoma-like features	Unknown	Unknown	Kazakov, et al. [23]
63	M	Hand	Unknown	Tumor - no further description	Unknown	Unknown	Kazakov, et al. [23]
19	F	Submandibular area	7 mo	Enlarging, painless nodule Size: 1.0 × 0.8 × 0.7 cm	Excision	Unknown	Koo & Chang, et al. [24]
36	M	Forearm	3 yr	Firm, solitary, growing nodule with pigmented and multifaceted surface Differential: apocrine hidradenoma, lipoma, fibrolipoma, epidermal inclusion cyst, basal cell carcinoma, pyogenic granuloma, malignant eccrine poroma	Excision	Unknown	Kumar, et al. [4]
68	M	Neck	Unknown	Firm, intradermal nodule Differential: squamous cell carcinoma, basal cell carcinoma, pilomatixoma, sebaceous carcinoma	Unknown	Unknown	Layfield & Mooney [25]
74	M	Breast	Not stated	Slow growing, painless, hyperpigmented and nodular mass Mammography showed 46 mm × 40 mm mass with macro-lobulated margins in the upper outer quadrant Ultrasound showed complex cystic and solid mass 4.0 cm from the nipple	Modified radical mastectomy (suspected breast malignancy)	Unknown	Liaquat, et al. [26]
67	F	Heel	1 yr	Solitary, blueish, indurated, and tender nodule	Excision	Nil at 3 mo	Lim, et al. [27]

Table 1 (continued)

Age, years	Sex	Location	Duration	Symptoms/examination/differentials	Treatment	Recurrence	References
50	M	Temple	6 mo	Solitary, skin-colored nodule Size: 1.5 cm diameter	Excision	Nil at 3 mo	Lim, et al. [27]
13	M	Forearm	2 yr	Tender, soft, erythematous, and violaceous nodule Size: 1.0 cm diameter	Excision	None at unspecified follow-up	Lopez, et al. [28]
57	F	Lateral surface distal thigh	4 yr	Stable, erythematous, desquamating papule with central scabbing Size: 7 mm diameter superficial papule; 15 mm diameter rubbery nodule deep to papule Ultrasound showed well-defined, hypoechoic lesion in the subcutaneous tissue with cystic appearance	Excision	Unknown	Martínez-Morán, et al. [29]
61	F	Posterior aspect of knee	Unknown	Soft, light brown nodule containing a pigmented papular lesion in the center	Unknown	Unknown	Misago & Kohda [30]
79	M	Lumbar area	8 yr	Painless, enlarging, pedunculated 4 cm mass with 2.0 cm stem that was smooth, reddish, hemorrhagic in some places and had purulent smell	Excision	Nil at 3 yr	Michalinos, et al. [31]
65	M	Middle finger, ulnar side	1 yr	Small, round, painful nodule Size: 0.1 × 0.9 × 0.2 cm Differential: sebaceous cyst	Excision	15 mo	Miller, et al. [8]
74	F	Scalp	40 yr	Growing mass over 3 years with ulceration and pigmented nodular lesions Size: 2.0 × 3.0 × 0.5 cm Differential: nevus with pyogenic granuloma, basal cell carcinoma	Excision	Nil at 1 yr	Min, et al. [32]

Table 1 (continued)

Age, years	Sex	Location	Duration	Symptoms/examination/differentials	Treatment	Recurrence	References
81	M	Parasternal	"few" years 1 year	Enlarging, hard, painless, pedunculated mass Size: 6.0 cm diameter Differential: vascular tumor Ultrasound showed Heterogenous tumor with anechoic areas and cystic components CT showed presternal subcutaneous mass containing apparent cystic and bleeding components	Excision	None at 2 wk, long-term unknown	Mlika, et al. [33]
58	M	Parietal scalp		Painless, raised lesion Size: 1.5 × 0.9 × 0.3 cm Differential: epidermal inclusion cyst, pilar cyst, lipoma	Excision	Unknown	Mukit, et al. [34]
34	M	Chest	18 mo	Tense, cystic, painless swelling Size: 5.0 × 5.0 × 3.0 cm Differential: cyst with malignancy Ultrasound showed heterogenous mass with anechoic and cystic components with mild vascularity CT showed subcutaneous mass with dual solid and cystic components	Excision	Unknown	Piana, et al. [35]
65	M	Knee, anterior aspect	3 yr	Mildly painful nodule with normal overlying skin Differential: leiomyoma, dermatofibroma	Excision	Unknown	Requena & Sanchez [5]
50	F	Breast	Not stated	Painless breast lump Size: 3.4 × 2.5 × 2.3 cm Mammography showed solid-cystic lesion Ultrasound showed 42 × 22 mm well-defined, complex cystic mass with increased vascularity	Excision	Unknown	Sharma, et al. [36]

Table 1 (continued)

Age, years	Sex	Location	Duration	Symptoms/examination/differentials	Treatment	Recurrence	References
34	M	Leg, anterior aspect	6 yr	Slowly enlarging, dark bluish, dome-shaped nodule Size: 2.1 × 1.3 cm	Excision	Unknown	Ueno, et al. [37]
56	F	Great toe, plantar lateral side	12 yr	Enlarging, painless, lobulated mass with underlying blue hue Size: 4.5 × 3.0 × 2.5 cm Differential: fibrous tumor, nodular tenosynovitis, fibroma, lipoma, unspecified soft tissue neoplasm Ultrasound showed solid soft tissue mass with multiple internal septations CT showed large, soft tissue mass, not eroding into bony structures, no enhancement	Excision	None at 4 yr	Whitmore, et al. [38]
67	F	Upper back	5 yr	Enlarging, itchy, and painless nodule that has changed in color over 5 m period Size: 0.9 × 0.7 × 0.3 cm Differential: benign versus atypical nevus	Excision	None at week 1 visit, will continue to follow	Case presented
Average (range) age: 57.0 (13–81)	M:F	Head/neck: 16 Trunk: 28 Limb: 21 41: Hand: 3 32 Foot: 2 Other: 4	Average (range) duration: 4.4 yr (0.1–40 yr)				
Clinical data included: age of patient in years, sex of patient, location of lesion, how long the patient stated the lesion had been present, brief description of the lesion, results of any noninvasive diagnostic studies performed, differential before pathology, treatment performed, and recurrence of lesion.							

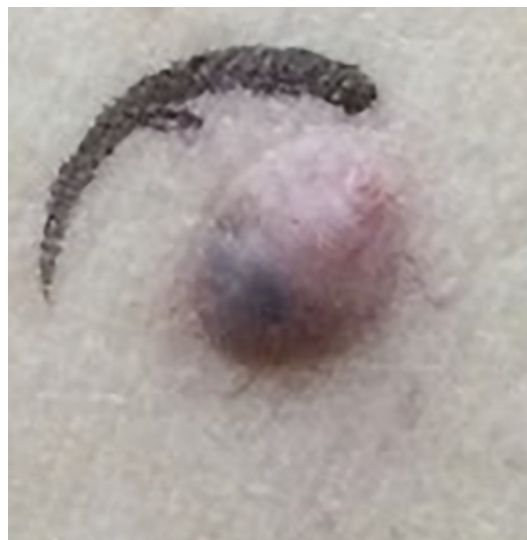


Fig. 2. Photograph of skin lesion pre-shave biopsy. Solitary, firm, and well-circumscribed nodule with variable pigmentation on the patient's right upper posterior shoulder.

Discussion

PH is one of the four variants of poroid neoplasms characterized by a single or multi-lobulated nodule contained within the dermis with no connection to the overlying epidermis [27, 38]. These tumors are composed of two types of cells: small, round, basophilic poroid cells and eosinophilic, squamoid cuticular cells with eccrine differentiation [6]. PH is thought to be a benign cutaneous neoplasia with low (<1%) risk of malignant transformation [32]. However, complete surgical excision remains the standard treatment due to possible advancement of pathology from a benign eccrine poroma to eccrine porocarcinoma [39–41] or development of carcinomatous changes to hidradenocarcinoma [42], as well as to prevent recurrence, and due to its common misdiagnosis for malignant subcutaneous neoplasms.

In fact, although, generally benign most eccrine sweat gland tumors of all types are excised due to misdiagnosis, patient complaints, or malignant potential. These poroid neoplasms may be clinically difficult to differentiate from other cutaneous lesions, as well as from each other, without histological examination. Eccrine poroma, hidroacanthoma simplex, dermal duct tumor, and PH share a common cellular composition of poroid and cuticular cells and have been suggested to exist more on a spectrum of poroid neoplasms with eccrine differentiation rather than as independent entities [6, 43]. In their pioneer article, Abenza and Ackerman defined PH as a fourth additional variant of the poroid neoplasms and defined each subtype according to location of the neoplastic cells and their architecture. Lesions were termed hidroacanthoma simplex when the neoplastic poroid cells were confined entirely within the epidermis and are arranged in ovoid nests. Tumors were given the designation eccrine poroma when the cells involved basal layer of the epidermis and extended into the superficial part of the dermis. Neoplasms were named dermal duct tumor when neoplastic poroid cells were restricted to the dermis and formed of discrete nodules, widely spaced and devoid of cysts. Lastly, when the neoplastic poroid cells are in a tumor with solid and cystic components, contained within the dermis without connection to the epidermis, the neoplasm is called PH. Among the group of eccrine adnexal tumors, PH is thought to be the rarest type [6, 36, 20].

While PH is repeatedly quoted to be a rare diagnosis with very few reported cases, our systematic review reveals the incidence of PH may be underestimated. Here, we have presented the most comprehensive systematic review of reported PH to date (as of January

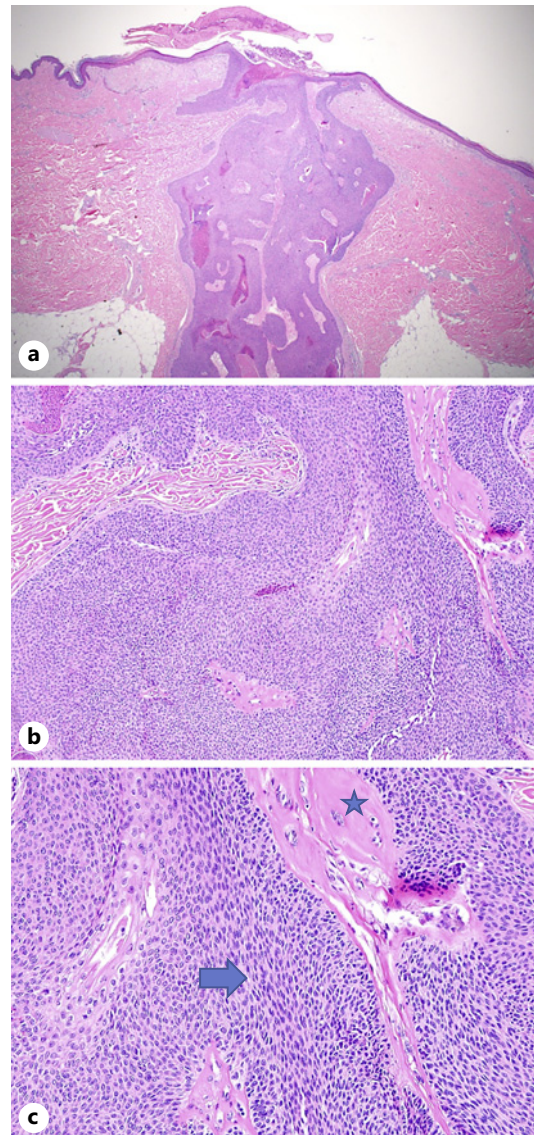


Fig. 3. **a** H&E stain, $\times 20$. Fusiform, nodular adnexal neoplasm emanating from the epidermis, with dermal and subcutaneous extension. **b** H&E $\times 100$. Well-circumscribed lobules of PH. **c** H&E $\times 200$. Bland round poroid cells (arrow) within a hyalinized vessel-rich stroma (star).

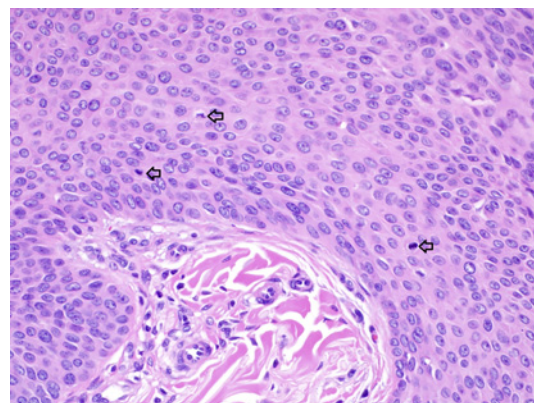


Fig. 4. H&E stain, $\times 400$. Focally increased mitotic rate but no cytologic atypia.

2022). While <100 cases of PH have been reported since its initial recognition in 1990, the close relationship between poroid neoplasms may complicate recording its true incidence. We have found multiple reports of composite tumors, including PH mixed with eccrine poroma [12, 44], hidrocanthoma simplex [12], and dermal duct tumor [11]. Additionally, we have shown that PH presents with high variability in both characteristics of the presenting lesion and patient population. The slow-growing and often painless nature of PH may lead to patient delay in presentation until size or color change raises concern, or not at all. All of these factors may be leading to underdiagnosis and PH may be more prevalent than previously thought.

Statement of Ethics

Ethical approval is not required for this study in accordance with local or national guidelines. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Funding Sources

No funding source for this study.

Author Contributions

Arrin Brooks: completed the review of the literature and primary author of the manuscript. Mariah Morris: obtained histological data and edited manuscript. Jonathan Cuda: analyzed histological samples and wrote histological descriptions. Armein Rahimpour: edited manuscript. Semeret Munie: performed lesion excision and oversaw the study.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material files. Further inquiries can be directed to the corresponding or senior author.

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