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Papillary Eccrine Adenoma: A Recent Review of Literature

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Abstract Background: Papillary eccrine adenoma (PEA) is a very rare benign cutaneous sweat gland tumor, it usually presents as an isolated well-circumscribed dermal nodule existent for a prolonged duration of time. Objectives: We report a case of PEA in a 74-year-old woman from Mexico who presented with a rare case of papillary eccrine adenoma. Materials and methods: We gathered our information by using PubMed and Scopus portal, in total there have been only 48 cases reported so far in English Literature. We present detailed review of findings of all cases reported so far including our case report. Results: Our results indicated that the median age of presentation was 45 years amongst males and females. The mean size of the tumor is approximately 16 mm amongst males and females with more than double the incidence was noted in females, along with a predominant occurrence noted in the Black population. The most common location for presentation was the lower extremities, followed by the upper extremities, abdomen and face respectively. Conclusion: In summary, PEA is a rare benign cutaneous sweat gland tumor representing intradermal proliferation of sweat gland ducts. It is slow growing in nature, with a greater incidence in women, and predominantly presents on the extremities. Despite its typical histological features, at times it can resemble other cutaneous neoplasms, thus it may present with diagnostic challenges. Moh's technique has lately been reported to have increased success rates in the management of this tumor.

Keywords: *Rare benign cutaneous tumor, sweat gland origin, histopathological findings, diagnostic challenges, Moh's technique*

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1. Introduction

Papillary eccrine adenoma (PEA) is a rare benign cutaneous tumor of sweat gland origin. It was first described by Rulon and Helwig in 1977. [1,2,4] It most commonly presents as a slow growing solitary dermal nodule on the extremities, less frequently involving the trunk or face. [2,3] These tumors are well circumscribed, unencapsulated and composed of numerous dilated intraluminal papillae. [1,2,3] PEA is most frequently seen in women and are often located on the extremities. [1,2,3,7] The majority of PEA's reported in the literature have followed a benign clinical course; the recommended treatment is conservative surgical excision with clear margins. [7] Our paper entails a case report and a detailed review of recent medical literature on PEA. Through our review we plan to discuss histopathological findings, diagnostic challenges, and recent advances in management of this rare tumor.

2. Materials and Methods

We gathered our information by using PubMed and Scopus portal, in total there have been only 48 cases reported so far in English Literature. Our review involves a specific focus on case reports between the time period of 1994-2014, i.e. those case reports after that of the review of literature by Mizuoka et.al. in 1998. [5] We additionally report a case of this rare cutaneous tumor, studied by light microscopy.

3. Case Report

A 74-year-old woman from Mexico presented with a solitary lesion on her right shin that she first noticed two years ago. She claimed no change in size of the mass over that time period. The only complaint was its unaesthetic appearance. On examination, the lesion measuring 4 mm x 3 mm x 2 mm was diagnosed by biopsy as papillary eccrine adenoma. The skin mass was further excised with

local wide resection. Histopathological findings showed papillary eccrine adenoma with negative margins. Upon follow up visits, our patient showed no recurrence of PEA.

4. Light Microscopy Findings

Hematoxylin and Eosin microscopic examination revealed multiple dilated ducts of various sizes in the dermis. Several intraluminal papillae extending into the lumen were present. The ducts as well as the papillae were lined by 2 to 5 layers of bland cuboidal cells. The cells showed minimal cytoplasm. Features of apocrine differentiation were not noted. These cells were covered by an outer layer of spindled myoepithelial cells. The surrounding dermis showed dense fibrosis. No nuclear pleomorphism, cytological atypia or mitotic figures were evident. There was no necrosis of epithelial elements, nor was there any evidence of decapitation secretion These features were diagnostic of papillary eccrine adenoma.

5. Discussion

PEA usually presents as an isolated well-circumscribed dermal nodule existent for a prolonged duration of time. Mizuoka etal (1998), reported a total of 37 cases in English literature. In their review, it was found that PEA frequently occurs within a wide age range: between 9-78 years with a median age of 46 years. A greater preferred occurrence in women is noted, with an approximate male to female ratio of 7:27. [5] A majority of PEA's were located on extremities, with a size varying between 5 mm-40 mm in diameter (median size= 13 mm) and clinical duration between 5 months-20 years (from diagnosis). [5]

Demographic and Tumor Characteristics

Our study reports a total of 48 cases of papillary eccrine adenoma (Table 1).

Age: The age range varied between 9 to 78 years, with median age of 45 years. Median age at time of diagnosis amongst males and females 40 and 49 years respectively (Table 2, Figure 2).

Table 1. Reported cases of papillary eccrine adenoma

Case	Age (yrs)	Sex	Location	Size (mm)	Duration (mos)	Follow-up (mos)	Race/Ethnicity
1	12	F	Left middle finger	20	144	99	African American
2	12	F	Left wrist	10	ND	109	African American
3	30	M	Left hand	10	Several	35	African American
4	20	M	Abdomen	6	11	37	American Indian
5	49	F	Foot	5	ND	16	African American
6	61	F	Nose	15	18	30	White
7	62	F	Lateral hip	5	Many	26	African American
8	63	F	Shoulder	7	ND	None	ND
9	14	F	Foot	14	ND	24	African American
10	69	F	Foot	10	Many	14	ND
11	52	F	Great toe	13	72	13	ND
12	9	F	Fifth toe	9	5	12	African American
13	52	M	Upper thigh	ND	Many	None	African American
14	40	F	Leg	13	ND	None	ND
15	43	F	Right leg	10	240	2	ND
16	66	M	Right thigh	30	168	24	African American
17	68	F	Right forehand	12	Many	108	African American
18	ND	ND	ND	ND	ND	ND	ND
19	57	F	Right arm	12	ND	26	African American
20	25	F	Right foot	25	132	ND	ND
21	44	F	Right foot	20	132	48	Japanese
22	44	F	Left elbow	30	120	108	African American
23	64	F	Left heel	ND	ND	96	Caucasian
24	12	F	Left middle finger	20	ND	ND	African American
25	35	F	Left wrist	ND	Several	24	ND
26	43	F	ND	20	240	ND	ND
27	22	F	Right hand	20	72	ND	ND
28	ND	ND	Face	ND	ND	ND	African American
29	ND	ND	Face	ND	ND	ND	White
30	29	F	Right leg	10	60	ND	ND
31	53	F	Left leg	15	24	ND	African American
32	45	M	Right cheek	20	24	24	ND
33	63	F	Right thigh	40	48	36	ND
34	31	M	Right heel	28	24	ND	African American
35	52	F	Left foot	15	36	ND	ND
36	60	M	Left knee	15	24	36	ND
37	78	F	Right forearm	13	120	None	Japanese
38	28	M	Right hand	10	6	ND	ND
39	47	F	Left forearm	20	at least 180	lost to f/u	African American
40	49	F	Left forearm	10	192	lost to f/u	Hispanic
41	51	F	Left hand	32	2	12	African American
42	35	M	Left heel	20 x 15 x10	at least 12	18	White
43	67	M	Scrotum	13	36	ND	ND
44	45	M	Right thigh	9	Many years	ND	African American
45	58	M	Left upper arm	ND	24	24	Japanese
46	30	M	Left leg	10 x 10	60	12	Japanese
47	30	M	Left abdominal skin	27 x 21	180	12	Japanese
48	74	F	Right shin	4	24	12	Hispanic

ND=not done

Table 2.

	All	Female	Male
Number	48	31 (64%)	14 (29%)
Median Age in years (Range)	45 (9-78)	49 (9-78)	40 (20-67)
Median Duration prior to diagnosis in month (Range)	54 (2-240)	72 (2-240)	24 (6-180)
Mean Follow up (mo)	37.9+33	44.8+36.2	24.7+9.1
Race			
African American	19 (61.3%)		
Caucasian	4 (12.9%)		
Hispanic	2 (6.5%)		
Japanese	5 (16.1%)		
Other	1 (3.2%)		
Missing	17		
Size in mm (Range)	15.8+8.1 (4-40)	15.5+8.2	16.5+7.9
Location			
Extremity	39 (85%)	29 (96.7%)	10 (71.4%)
Lower	23 (50 %)	16 (53.3%)	7 (50%)
Upper	16 (35%)	13 (43.4%)	3 (21.4%)
Abdomen/Midsection	3 (6%)	0	3 (21.4%)
Face	4 (9%)	1 (3.3%)	1 (7.1%)
Missing	2	1	0

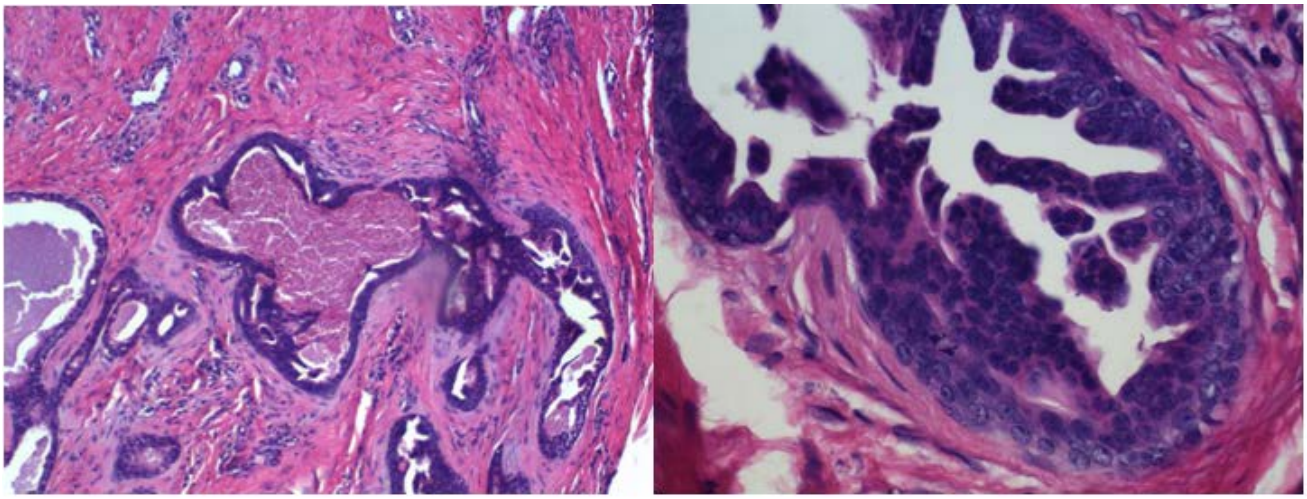

Figure 1. A shows dilated ducts within the dermis, B shows intraluminal papillary structures lined by cuboidal cells

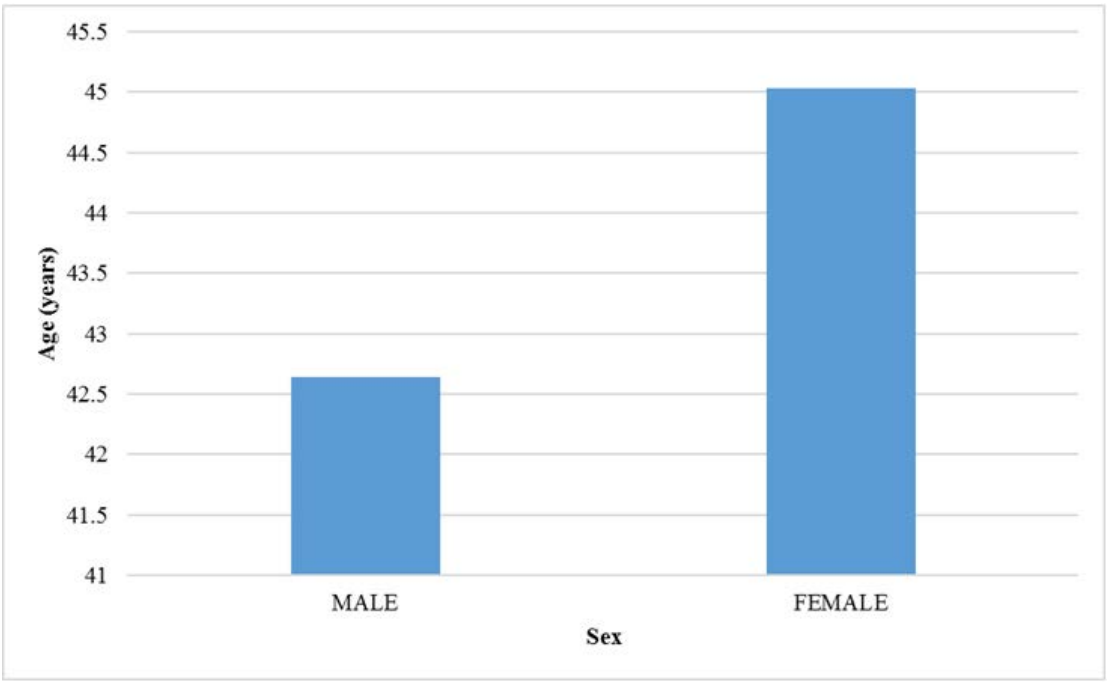

Figure 2. Mean Age of occurrence of PEA in males and females

Sex: The male to female ratio in our study is 1:2.2, indicating more than double the incidence in females.

Race: In 17 reported cases, the race of the patient was not documented, however amongst the remaining cases predominant occurrence is in the Black population (61.3% i.e. 19/31 cases).

Location: The most common location for presentation is extremities 39/46 cases (85%). Of these 39 cases, 23 (59%) were reported in lower extremity and the rest of them were located in upper extremities. Abdomen and face only accounted for 3 (approximately 6%) and 4 (9%) respectively (Table 2, Figure 3).

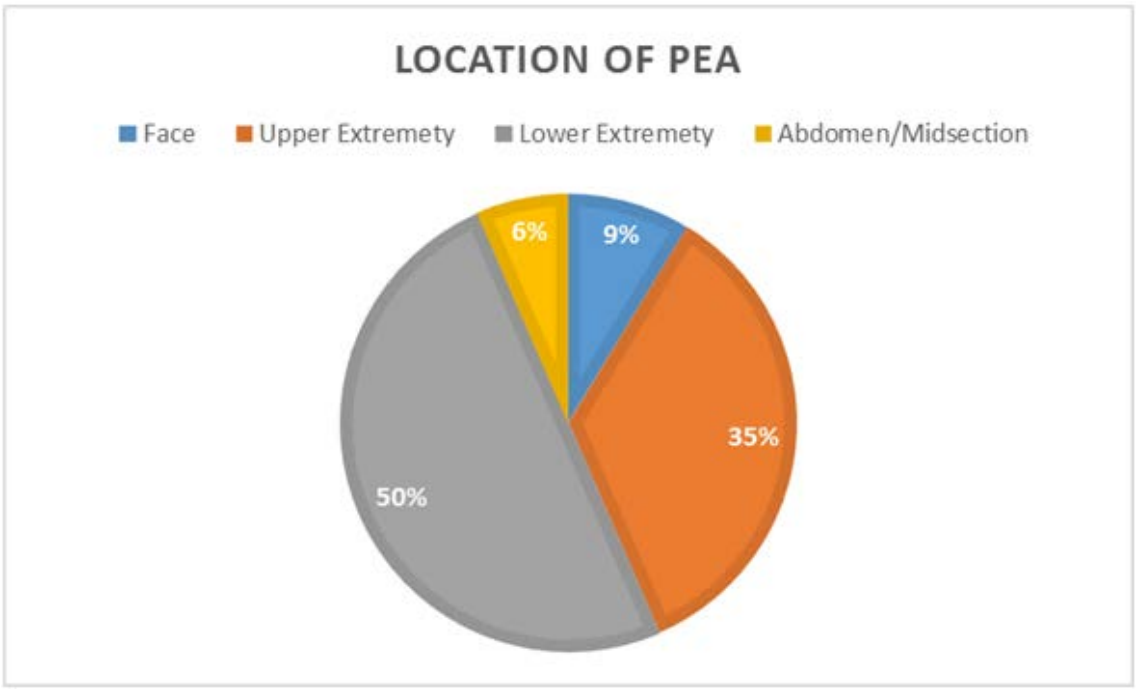

Figure 3. Location of PEA in males and females

Size: The size of the tumor varied from 4 to 40 mm in diameter with a mean size of approximately 16 mm. The majority of tumors were within the size range of 6 mm to 15 mm in males and females (Figure 4).

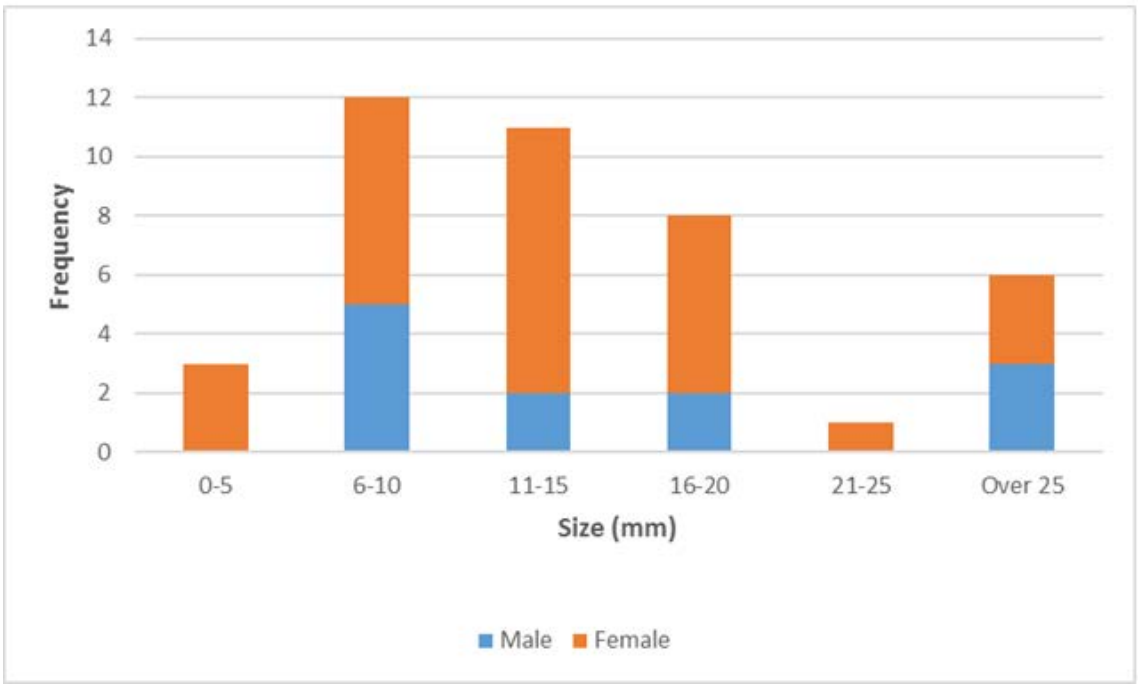

Figure 4. Size of PEA amongst males and females

Duration prior to diagnosis: The median duration of the PEA prior to presentation was 54 months, with duration ranging from 2 months to 20 years. Median duration of PEA in males was shorter by 48 months than females.

Histology

PEA represent intradermal benign proliferation of sweat gland ducts. [1,2,7] These sweat gland tumor ducts are often dilated and lined by a double layer of tumor cells with characteristic intraluminal papillations formed by

proliferation of the inner layers of epithelial cells lining these ducts. [1,3]

Immunohistochemistry

Immunohistochemistry is helpful to confirm the diagnosis; positive S-100 protein, CEA and EMA usually favor differentiation towards secretory epithelium of the sweat glands. [3,5] S-100 protein reaction strongly favors eccrine differentiation, however it is not present in all cases and therefore cannot be relied upon as a confirmatory marker, when used alone. [5,11] Markers such as actin (alpha-SMA), keratin 8 and keratin 14 are more reliable in suggesting an eccrine origin of the tumor. [5]

Differential

In most cases, PEA's have clear cut benign histological features but there are reported cases of diagnostic challenges in distinguishing this tumor from aggressive types of neoplasms such as aggressive digital papillary adenomas (ADPA), adenocarcinoma of the skin and basal cell carcinoma with eccrine differentiation. [7,17,19,20]

1. Tubular Apocrine Adenoma (TAA): PEA is considered the eccrine variant of TAA. Most authors consider PEA and TAA within the same spectrum of tumors due to their histopathological similarities. [3,11,12,13] Both tumor types can differentiate into both eccrine and apocrine forms. There has been no universal agreement about their origin based on current electron microscopic and immunohistochemical studies. [2,14] Despite their similarities, these two adenoma's have distinctive features: tubular apocrine adenoma's tend to occur most frequently on the scalp, they tend to show features such as decapitation secretion, shorter inward papillary projections into lumen, lack connections with epidermis, continuity of follicular infundibula and may contain infiltration by plasma cells as well as tending to co-occur with syringocystadenoma papilliferum. [2,11,12,13,15,16]
2. Aggressive Digital Papillary Adenoma (ADPA): Other cutaneous neoplasms that can present as a diagnostic challenge while diagnosing PEA are ADPA and adenocarcinomas. ADPA's are more infiltrative in nature with a nodular and solid configuration most frequently involving the fingers and toes of elderly population. [3,17] Additionally, it may have mild to moderate degree of atypia with pleopmorphism. [3,17]
3. Adenocarcinomas: Adenocarcinomas consistently have histological features of atypical mitosis, pleomorphism, necrosis, and vascular or perineural invasion. [7,17,18] It also has been observed that aggressive digital papillary adenomas and adenocarcinomas share a greater tendency for local recurrence and deep soft tissue infiltration. [7,19,20]
4. Basal Cell Carcinoma of Eccrine Differentiation (eccrine epithelioma): On histology, this carcinoma may appear similar to a PEA in that it contains tubules consisting of one or two layers of cuboidal cells. These layers are surrounded by fibrous stroma. Eosiniphilic granular material may be found intraluminally. However, unlike the PEA, these carcinomas do not contain intracystic papillations. Many of these carcinomas also contain additional histological differences such as cystic, alveolar, or cribriform areas. These carcinomas tend to be aggressive and often recur. [7]

Treatment

The current treatment of choice involves complete surgical excision of the neoplasm, and confirmation of clear margins of the tissue affected by the PEA. [4] In a recent case report by Jackson and cook (2002), Mohs micrographic surgery (MMS) was used to resect PEA for the first time. They emphasized the benefit of Mohs micrographic surgery in the management of cutaneous neoplasms, with reportedly high cure rates, greater tissue conservation and smaller post-surgical defects. [7] MMS has been reported to have success as a treatment for PEA, especially in cases where the neoplasm has borders that are difficult to determine, and when the lesion presents in a location that requires very careful and precise excision in order to preserve the function of nearby structures. [7] MMS has the benefit of thoroughly examining the surgical margins, without necessarily needing a wider excision, therefore leading to greater anatomical and functional conservation. [7] MMS also enables the surgical excision to contain smaller margins than that which is needed with the standard surgical removal of a PEA. [7]

Follow up

Although many cases reported lacked information on post-surgical follow up, mean follow duration amongst 48 reported cases was 38 months approx., with only one case of recurrence reported. [5] In our review of literature there were no cases of malignant tumors or metastases reported to date.

Recurrence and Distant Metastases

Papillary eccrine adenoma is a benign tumor that as of yet does not have any evidence or reports of metastasis. [4]

In summary, PEA is a rare benign cutaneous sweat gland tumor representing intradermal proliferation of sweat gland ducts. It is slow growing in nature, with a greater incidence in women, and predominantly presents on the extremities. Despite its typical histological features, at times it can resemble other cutaneous neoplasms, thus it may present with diagnostic challenges. Although metastasis has never been reported to occur, the treatment of choice is total surgical excision with complete removal of all affected margins. Moh's technique has lately been reported to have increased success rates in the management of this tumor.

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