

Inguinal Papule with a Milky Secretion

Hulya Cenk^a Meral Uner^b Gulbahar Sarac^c

^aDepartment of Dermatology, Pamukkale University Hospital, Denizli, Turkey; ^bDepartment of Pathology, Hacettepe University, Ankara, Turkey; ^cDepartment of Dermatology, Inonu University, Malatya, Turkey

Question

A 56-year-old man applied to our polyclinic with the complaint of a mass on his left groin. He had noticed the mass for the last 7 months, and there was a milky fluid coming from a small pore in the middle of the mass for the last 2 weeks. He was otherwise healthy, and he did not have a familial history. At the dermatological examination, there was a nipple-like papule with a central pore-like opening plugged with a keratinous material, and it was surrounded with darker velvety skin like an areola (shown in Fig. 1) When it was pressured, there was a milky fluid coming from the pore. A culture test and a biopsy were performed.

Possible answers:

- Dilated pore of Winer
- Pilar sheath acanthoma
- Trichofolliculoma
- Accessory nipple

What is your diagnosis?



Fig. 1. Nipple-like papule surrounded with darker velvety skin like an areola and a milky fluid.

This case was documented as a poster presentation at IDEA CONGRESS 2019, 24–28 April 2019, in Istanbul with the title “Is this an AN with a milky secretion.” Presentation at a meeting: IDEA CONGRESS 2019. Organization: IDEA CONGRESS 2019. Place: Istanbul. Date: 24–28 April 2019.

Answer

Pilar Sheath Acanthoma

Accessory nipples (ANs) are minor congenital malformations located along the embryonic milk lines [1]. These lines extend bilaterally from the axillae, through the chest and abdomen toward the groin [2]. ANs may be small and, therefore, unnoticed [3] or, sometimes, noticed at puberty, menstruation, or pregnancy when hormonal changes result in milk production [4].

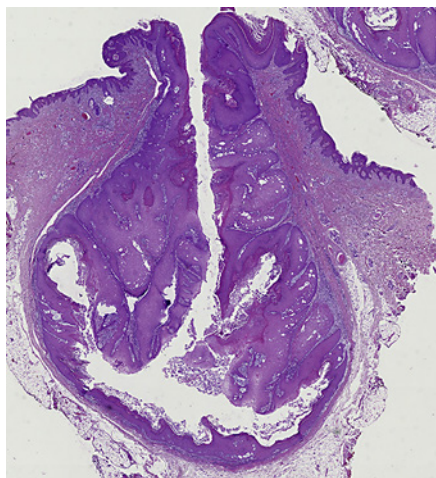


Fig. 2. Low power view of the lesion having solid lobules of eosinophilic squamous cells connected to a crater-like cavity in the dermis with a central pore opening (H&E; $\times 15$).

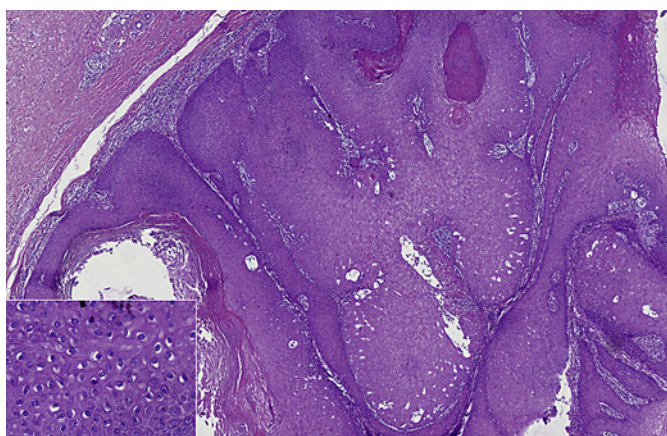


Fig. 3. Lobular proliferation of benign squamous epithelium (H&E; $\times 100$); there are areas of clear cells, indicating outer root sheath differentiation (**inset**).

Pilar sheath acanthoma (PSA) is a rare, benign facial neoplasm seen in typically middle-aged and elderly patients. It presents as an asymptomatic, solitary papule or nodule with a central opening plugged with keratin [5, 6]. Generally, they are found on the upper lip, although they can be found anywhere on the head and neck [7].

The biopsy revealed solid lobules of eosinophilic squamous cells connected to a crater-like cavity in the dermis with a central pore opening (shown in Fig. 2), and lobular proliferation of benign squamous epithelium with areas of clear cells, indicating outer root sheath differentiation (shown in Fig. 3). The lobules composed of bland keratinocytes around small cystic spaces and were surrounded by a prominent basement membrane (shown in Fig. 4). These findings were consistent with PSA.

Differential diagnoses include trichofolliculoma, dilated pore of Winer, keratoacanthoma, and inverted follicular keratosis. An excisional biopsy of the lesion helps confirm the diagnosis [5, 7, 8]. PSA has a central pore with keratinous material, while in trichofolliculoma, there are small hair follicles radiating from the wall of central cystic cavity [8]. Also, histopathologically, trichofolliculomas contain many small hair follicles radiating from the wall of a central cystic structure in a well-formed stroma which are absent in PSA [5, 9]. Dilated pore of Winer has a central cavity and an acanthotic cystic wall with thin strands or finger-like projections of the epithelium into the surrounding connective tissue; addition-

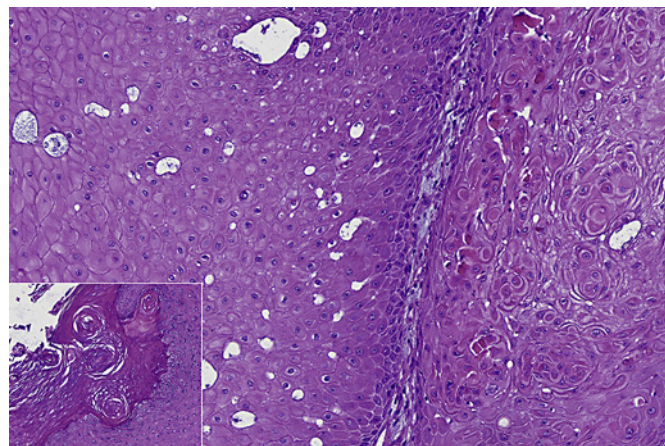


Fig. 4. Lobules are composed of bland keratinocytes surrounded by a prominent basement membrane, and these lobules surround small cystic spaces (H&E; $\times 200$); there is a granular layer similar to an epidermoid cyst with prominent surface keratin (**inset**).

ally, in the histopathology of PSA, there are no hair shafts and sebaceous glands and fibrovascular stroma [5, 9]. PSA is less mature than dilated pore of Winer [8]. Keratoacanthoma is among the differential diagnoses in terms of clinical features. PSA shows no spontaneous regression contrary to keratoacanthoma [5, 8]. Inverted follicular keratosis may represent an irritated seborrheic keratosis or a distinct neoplasm derived from the infundibular portion of the hair follicle [10]. Neoplasms arising from pilar infundibular apparatus have some common histological features, such as superficial nature of growth, connection with the epidermis, pore-like opening proliferation of the outer sheath epithelium, infundibular keratinization, and connection with the pilosebaceous structure [8]. The presence of whorled keratinocytes without keratinization or atypia (squamous eddies) and the lack of squamous pearls or cytologic atypia are the features in favor of inverted follicular keratosis [10]. Our case had more prominent solid lobules of eosinophilic cells than squamous eddies, suggesting a PSA diagnosis. Also, it was connected to the surface with a characteristic crater-like cavity, in favor of PSA as well. We did not see any cytopathic change (koilocytosis, etc.) suggestive of an HPV infection, precluding the prediagnosis of verruca.

In our case, there was a lesion with a subtle central opening and a milky secretion coming from it. Our provisional diagnoses were AN and lymphoma. The cultural test indicated that the lesion was infected with *Klebsiella oxytoca*, and the biopsy result was consistent with a PSA. Our patient has given a written informed consent to publish his case, including publication of images on medical platforms.

Men also can have AN or supernumerary breasts. Since the lesion looked very similar to an AN, it was our first prediagnosis. Also, strangely, there was milky secretion. Men with prolactin-secreting pituitary adenomas may have galactorrhea [11]. Despite a very low probability, the patient would have had a hormonal disorder causing galactorrhea.

In this case, if it were not for the male gender, the clinical picture suggested that the lesion could be an AN. And also, the biopsy result turned out to be totally different from what we have thought; therefore, we found this case valuable to share.

Statement of Ethics

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

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Funding Sources

There is no funding.

Author Contributions

Hulya Cen: conception or design of the work; acquisition, analysis, and interpretation of data, drafting the work and revising it critically for important intellectual content, final approval of the version to be published, and agreement to be accountable for all aspects of the work. Meral Uner: providing histopathological image and relevant text, interpretation of data, drafting the work or revising it critically for important intellectual content, final approval of the version to be published, and agreement to be accountable for all aspects of the work. Gulbahar Sarac: conception and design of the work; the acquisition, analysis, and interpretation of data, drafting the work and revising it critically for important intellectual content, final approval of the version to be published, and agreement to be accountable for all aspects of the work.

Keywords

Accessory nipple · Pilar sheath acanthoma · Milky fluid · Inguinal pilar sheath acanthoma

References

- 1 Brown J, Schwartz RA. Supernumerary nipples: an overview. *Cutis*. 2003;71(5):344–6.
- 2 Marinopoulos S, Arampatzis I, Zagouri F, Dimitrakakis C. Pseudomamma of the inguinal region in a female patient: a case report. *Int J Surg Case Rep*. 2015;12:71–4.
- 3 Paul CR, Harris MA. Physical examination of the newborn. In: Hertz DE, editor. *Care of the newborn: a handbook for primary care*. 1st ed. Philadelphia, PA: Lippincott Williams & Wilkins; 2005. p. 23–46.
- 4 Lakkawar NJ, Maran G, Srinivasan S, Rangaswamy T. Accessory breast tissue in the axilla in a puerperal woman- case study. *Acta Med Medianae*. 2010;49(4):45–8.
- 5 Ozgur T, Temel M. Pilar sheath acanthoma presenting as a nevus. *Int J Trichology*. 2015;7(4):187–8.
- 6 Mehregan AH, Brownstein MH. Pilar sheath acanthoma. *Arch Dermatol*. 1978;114(10):1495.
- 7 Kushner JA, Thomas RS, Young RJ. An unusual location of a pilar sheath acanthoma. *Int J Trichology*. 2014;6(4):185–6.
- 8 Bavikar RR, Gaopande V, Deshmukh SD. Postauricular pilar sheath acanthoma. *Int J Trichology*. 2011;3(1):39–40.
- 9 Ba W, Wang W, Li C. An unusual location of a pilar sheath acanthoma. *Int J Trichology*. 2015;7(4):179–81.
- 10 Ullman D, DiCarlo CM, Ferringer T. Verrucoid lesion on the eyelid. *Cutis*. 2017;100(4):216. 227;228.
- 11 Serri O, Somma M, Rasio E, Beauregard H, Hardy J. Prolactin-secreting pituitary adenomas in males: transphenoidal microsurgical treatment. *Can Med Assoc J*. 1980;122(9):1007–13.