

A Nodular Hidradenoma of Atypical Location in Pregnancy

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Nodular hidradenomas (NH) are benign tumors that arise from eccrine adnexal structures. NH are commonly located on the head, neck, and anterior trunk, with lower extremities being an atypical site (1, 2). NH exacerbation in pregnancy is rare and raises possibility of cytological changes occurring secondary to hormonal fluctuations.

CASE REPORT

A 36-year-old Hispanic woman, gravida 6 para 6, at 31 weeks gestation of pregnancy presented with a tender nodule on her right lower leg that was stable for 2 years but enlarged during pregnancy. There was no significant past medical history or trauma to the area.

Physical examination of the right medial lower leg revealed a well demarcated, dome-shaped light brown nodule measuring 10×15 mm (**Fig. 1**). There was no regional lymphadenopathy and systemic examination was unremarkable. We considered a wide differential diagnosis, including arteriovenous malformation, connective tissue neoplasms, adnexal tumors, skin cancers, and cutaneous metastatic tumors.

A punch biopsy was performed. Histopathologic evaluation showed a well-circumscribed solid-cystic dermal tumor consisting of a solid sheet-like arrangement of

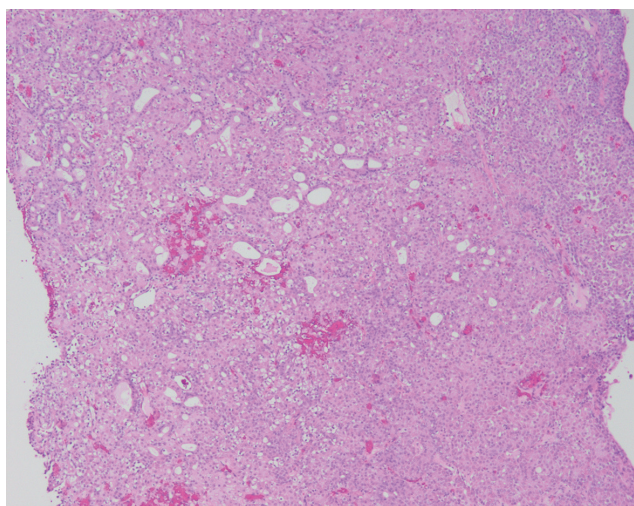


Fig. 2. The well-circumscribed dermal tumor consists of a solid sheet-like arrangement, containing eosinophilic, polygonal or fusiform cells, cystic spaces, and some cellular differentiation (Hematoxylin and eosin, 4x).

eosinophilic, polyhedral and clear cells. The tubular and cystic structures are filled with homogeneous eosinophilic material. Atypia, invasion, necrosis, and mitoses are not observed (**Figs. 2–3**). Immunohistochemical studies showed tumor cells positive for cytokeratin 7 (CK7) and epithelial membrane antigen (EMA), and negative for



Fig. 1. A well-demarcated border, dome-shaped, light brown nodule measuring 10 x 15 mm on the right lower leg.

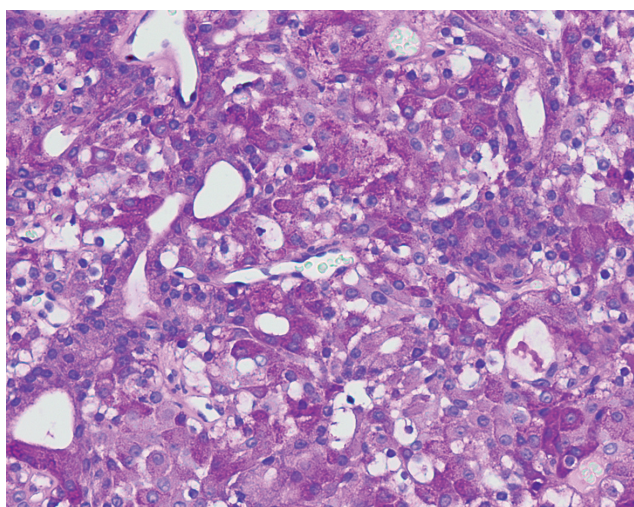


Fig. 3. The hidradenoma cells are large and polyhedral with distinct cell borders, containing round to ovoid nuclei, evenly distributed nuclear chromatin, and pale to eosinophilic cytoplasm. Tubular lamina formed by cuboidal cells are present. Hidradenoma produces mucin, which stains red with PAS stain, 20x.

renal cell carcinoma marker (RCC), paired box gene 8 (PAX8), carcino-embryonic antigen (CEA), and CD10. Tumor cells were also diffusely positive by periodic acid schiff (PAS) stain. These findings were consistent with nodular hidradenoma.

Due to its benign nature, surgical removal was postponed until after delivery when the tumor was excised with a 3 mm margin.

DISCUSSION

NH are benign tumors that arise from eccrine adnexal structures. NH was first described by Liu in 1949 as clear cell papillary carcinoma of the skin, and has been termed clear cell hidradenoma, eccrine acrospiroma, and solid-cystic hidradenoma (3).

NH has a female predominance (1.7:1) with a mean age of 37.2 years (4). NH presents as a single, small, slow-growing, well-demarcated, intra-dermal tumor that varies in size from 5–30 mm. The overlying skin varies in color, with a smooth, thickened, atrophic, or ulcerated surface (1). It is common on the head, neck, and anterior trunk, and infrequent in the lower extremities (1, 2).

Our case involved an atypical location on the leg. In a 10-year review of 89 NH cases, 4 were located on the legs (4.49%) (4). The unusual location and exacerbation during pregnancy prompted our report of this case. Pregnancy is known to increase eccrine activity with exception of at the palms. This may lead to miliaria, hyperhidrosis, and dyshidrotic eczema (5). Our case suggests that pregnancy can contribute to the growth of eccrine sweat apparatus tumors, but this relationship has not been elucidated.

NH may be asymptomatic or may present with pain, or bleeding, and rarely can show malignant transformation (6). Diagnosis is made by histopathology. NH presents as a well-circumscribed dermal nodule surrounded by a collagenous pseudocapsule, occasionally extending into subcutaneous tissue (1, 7). The tumor is comprised of multiple lobulated masses of epithelial cells and tubular lamina (1, 7). Several different cell types are seen, in-

cluding large polyhedral eosinophilic cells and fusiform cells, as well as columnar, cuboidal, squamous, and intermediate cells. Duct-like structures and cystic cavities are commonly found as is collagenous, hyalinized, and mucinous stroma (1). Dermoscopy shows a nodule of amorphous, white-pink areas, red lacuna-like areas, eccentric ulceration, as well as hairpin, dotted, and linear irregular vessels (8). Due to the variation in different cell types, histopathologic differential diagnosis is extensive, including apocrine mixed tumor, eccrine poroma, thyroid carcinoma, renal cell carcinoma, metastatic tumor, neuroendocrine carcinoma, hidradenocarcinoma, and glomus tumor.

Due to high recurrence rate, biopsy and surgical excision are warranted with follow-up to monitor for recurrence as malignant transformation has been known to occur (1). Our patient had an uncomplicated post-operative course with no evidence of recurrence at 12 months follow-up.

The authors have no conflict of interest to declare.

REFERENCES

1. Stratigos AJ, Olbricht S, Kwan TH, Bowers KE. Nodular hidradenoma: a report of three cases and review of the literature. *Dermatol Surg* 1998; 24: 387–391.
2. Winkelmann RK, Wolff K. Solid-cystic hidradenoma of the skin: clinical and histopathologic study. *Arch Dermatol* 1968; 97: 651–661.
3. Liu Y. The histogenesis of clear cell papillary carcinoma of the skin. *Am J Pathol* 1949; 25: 93–103.
4. Hernandez-Perez E, Cestoni-Parducci R. Nodular hidradenoma and hidradenocarcinoma: a 10-year review. *J Am Acad Dermatol* 1985; 12: 15–20.
5. Yang CS, Teeple M, Muglia J, Robinson-Bostom L. Inflammatory and glandular skin disease in pregnancy. *Clin Dermatol* 2016; 34: 335–343.
6. Diab M, Gabali A, Kittaneh M. Malignant acrospiroma: a case report in the era of next generation sequencing. *BMC Cancer* 2017; 17: 221.
7. Nandeesh BN, Rajalakshmi T. A study of histopathologic spectrum of nodular hidradenoma. *Am J Dermatopathol* 2012; 34: 461–470.
8. Robles-Mendez JC, Martinez-Cabriaes SA, Villarreal-Martinez A, Ayala-Cortes AS, Miranda-Maldonado I, Vazquez-Martinez O, et al. Nodular hidradenoma: dermoscopic presentation. *J Am Acad Dermatol* 2017; 76: S46–48.