

Case Report

Mammary-Like Gland Adenoma of The Vulva in An Adolescent: A Case Report

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Abstract

Background: Mammary-like gland adenomas, formerly named Hidradenoma Papilliferum (HP), are benign lesions derived from anogenital mammary-like glands [1,2]. The occurrence of these glands in the vulvar region is rare and constitutes 60% of non-malignant vulvar tumors [3].

Case: This case report describes an incident of mammary-like gland adenoma of the vulva in an adolescent female.

Conclusion: This report reviews treatment options for benign and malignant lesions while identifying topics for future research.

Keywords: Adenoma; Adolescent; Mammary-like glands; Vulva

Teaching Points

- Histopathological examination is essential in diagnosing mammary-like gland adenomas.
- Treatment modalities may differ with proper pathological identification and revolve around excisional biopsy with monitoring for recurrence.

Introduction

Mammary-like gland adenomas are benign, slow-growing lesions that represent up to 60% of non-malignant vulvar tumors. They are usually found on the anogenital region of middle-aged women. As noted by El-Khoury's et al., the most commonly affected areas include the interlabial sulcus, skin adjacent to the labia, or the perineum after analyzing 52 photographs of the tumor. In rare occurrences, ectopic sites include the ears, face, scalp, chest, and abdomen [3]. These adenomas are rare, benign, smooth-surfaced lesions approximately ranging from 0.5 to 1 cm found on

the labia. They can be soft, firm, translucent, skin-colored, pink-red, or dark colored [4]. These have been formerly named HP as it was believed to be an apocrine tumor, but it is now thought that these lesions are originally derived from anogenital Mammary-Like Glands (MLG); they have henceforth been more accurately termed as MLG adenoma, first coined by van der Putte in 1991 [3]. They are a normal finding in the male and female anogenital region, found in the subepidermal stroma lined by basophilic cells and staining positive for estrogen and progesterone receptors. In females, they are commonly located in an ellipse formed by the periclitoral region interlabial sulci, fourchette, perineum, and perianal region [5]. Histological features of MLG adenomas include tubular and cystic adenomatous structures containing apocrine-like cytological characteristics and abundant fibrous stroma in a lobular arrangement [6].

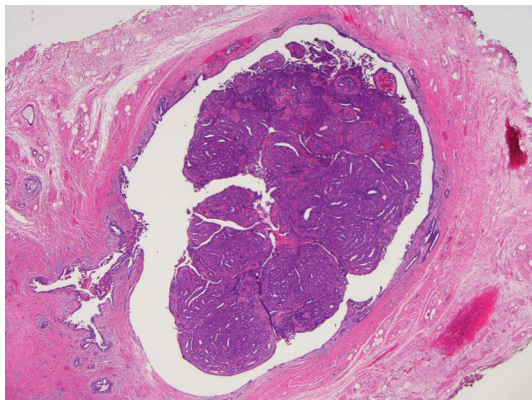
There have been few cases reported of MLG adenomas in the young adult population. As such, there is a lack of clinical trials determining definitive treatment to prevent recurrence or malignant transformation. This review aims to identify a case of MLG adenoma of the vulva in a young adult and describe treatment options and outcomes.

Case Presentation

A 21-year-old, gravida 0, presented for evaluation of a small nodule on the left vulva. The patient states she was not sexually active; and her past medical history includes dysmenorrhea, migraine headaches, hyperlipidemia, mononucleosis, and mitral valve prolapse. Her past surgeries include tonsillectomy and myringotomy. Her family history is significant for breast cancer in her mother, diagnosed at 49 years of age and treated with a lumpectomy. Genetic testing for the mother's status of BRCA gene mutation is negative.

The patient's physical exam revealed a non-tender, non-suppurative, mobile 1 cm lesion on the left vulva. Her pelvic and breast examination were benign. Initially the vulvar lesion was treated with antibiotics, with no subsequent resolution; subsequently, the lesion increased in size to 1.5 cm by the follow-up visit and was henceforth diagnosed as an epidermoid cyst of the labia majora. One month after the initial diagnosis, the patient underwent surgical resection due to increasing size.

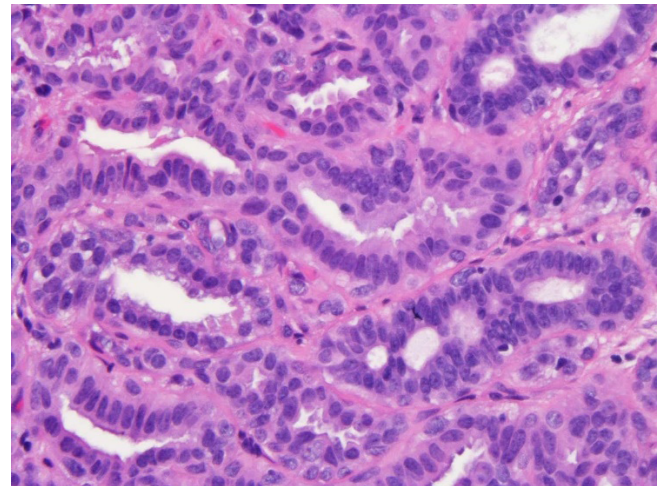
During surgery, the left vulva was prepped and draped under sterile conditions. The patient underwent sedation, and local infiltration of the vulva was initiated with 1% lidocaine in a circumferential manner. The granular lesion was palpated and a small (0.5 cm) vertical incision was made on the vulvar fold over the lesion. The nodule was lifted from its capsule using Allis clamps, and with bipolar energy the nodule was carefully removed from the cavity in order to ensure its complete excision. Hemostasis was assured throughout the procedure and the vulvar skin edges were re-approximated with 4-0 monocryl. The patient was then transferred to our recovery room and had a stable recovery. Subsequently, pathology findings were consistent with Hidradenoma Papilliferum (HP) showing characteristics of an adenomatous pattern with cystic spaces (Figure 1). At the six-month follow-up visit, the patient reported feeling scar tissue at the excision site, but denied re-growth of the lesion.



However, 11 months after the lesion was removed, the patient presented for evaluation of a recurrent left vulvar growth. She had

recently been on a course of antibiotics when she noticed the lesion had recurred. Patient underwent a second surgical resection of the left vulvar lesion. This time, the lesion was located 1-2 mm from the prior surgical resection margins. The area was again re-explored and a 1.5 cm nodule was removed from the adjacent scarred area. Again, we identified the capsule and ensured complete removal with bipolar energy. She had a uneventful recovery, and to date has been doing well without evidence of regrowth of the vulvar lesion.

Pathology results indicate that the final lesion was consistent with Mammary-Like Gland (MLG) adenoma of the vulva. Histologic findings are consistent with epithelial proliferation with features similar to sclerosing adenosis of the breast in addition to focal cystic glands with apocrine metaplasia. This histologic description is consistent with the final diagnosis of MLG adenoma of the vulva previously termed HP with mixed histopathologic features (Figure 2). These mixed features resemble mammary glands due to their glandular structures made up of branching ducts and surrounded by prominent fibrous stroma arranged around lobules [6].



Discussion

The findings from this case serve as an opportunity for discussion of effective treatment options and prevention of malignant transformation in patients with MLG adenomas of the vulva. In Scurry's review of 46 cases of MLG adenomas, their population ranged from 31-90 years with a mean age of 52 years; no lesions recurred. This case is characteristically different in that the patient is a young adult as compared to a middle-aged woman, and in that the lesion recurred. If continuous recurrence occurs, alternative treatment options must be considered. It is also of significant importance to monitor for malignant transformation in recurrent lesions. In Abbott's case of adenocarcinoma of MLGs of the vulva, success was reported with excisional biopsy and subsequent Mohs micrographic surgery in order to ensure adequate removal of the lesion. Furthermore, Butler's reported

case of primary invasive breast carcinoma arising from MLGs of the vulva, the patient had success with excision and ipsilateral groin Sentinel Lymph Node (SNL) dissection; she was also given letrozole, an aromatase inhibitor, as adjuvant treatment as the carcinoma was determined to be Estrogen Receptor (ER) positive. If malignancy with metastasis occurs, weekly paclitaxel chemotherapy has been shown to achieve a partial response with rapid and accurate diagnosis giving this therapy a greater prognosis [7]. These findings offer treatment management for recurrent and/or malignant MLG lesions.

Conclusions

MLG adenoma is a more accurate term for anogenital HP. Histopathological examination is essential in diagnosing MLG adenomas as the presence of tall columnar ductal cells with apical snouts is pathognomonic⁶. Treatment modalities revolve around excisional biopsy with monitoring for recurrence. In cases of malignancy, success has been reported using Mohs micrographic surgery, SNL dissection, letrozole, or paclitaxel as determined on a case-by-case basis. Future research may benefit from identifying cause, determining definitive treatment, preventing recurrence, and identifying a correlation with malignant breast tissue.

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