



Nipple adenoma in infancy^{☆,☆☆}

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Received 14 June 2009; revised 11 August 2009; accepted 12 August 2009

Key words:

Adenoma;
 Areola;
 Breast;
 Infant;
 Lesion;
 Nipple;
 Nipple adenoma;
 Pediatric;
 Reconstruction;
 Tumor

Abstract We report the first patient with a nipple adenoma presenting in infancy. Nipple adenoma is a benign lesion typically affecting women between 45 and 55 years of age. This lesion can occur in the pediatric population and should be included in the differential diagnosis of an infantile breast lesion. Management of children with nipple adenoma requires consideration for breast development; excision before maturity may cause nipple-areola deformity or injury to the breast bud.

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Nipple adenoma is a rare, benign lesion that may be confused with Paget disease or well-differentiated adenocarcinoma [1–4]. Although this condition is most commonly referred to as nipple adenoma [5,6], other terms have been used: florid papillomatosis [2,3], subareolar duct papillomatosis [7], erosive adenomatosis [7,8], and papillary adenoma [1]. Nipple adenoma typically presents in middle-aged women as a crusted mass with induration and serous or bloody discharge; men are rarely affected [7,9–13]. Treatment options include excision [14], cryosurgery [15], or Mohs micrographic surgery [16]. Because nipple adenoma is

easily confused with malignancy, unnecessary radical excision with deformation of the breast can occur [5–7,17,18]. Although nipple adenoma commonly affects adult women, 5 cases affecting the pediatric population have been reported [7,19,20]. We describe the first patient with a nipple adenoma presenting in infancy.

1. Case report

A 5-month-old female infant presented to an outside hospital with a new-onset nipple lesion. The mass slowly increased in size, and at 23 months of age, it was removed by shave excision. Histopathology showed nipple adenoma with positive margins. One month postoperatively, the growth was enlarged, and it was resected, without margins, 5 months after the shave excision at the same outside institution. Histopathology again showed nipple adenoma

[☆] Statement of financial interest: No financial support or benefits were given to the authors from any source that is related to the scientific work reported in this article.

^{☆☆} List of products used: No products, devices, or drugs were used in this article.

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with positive margins. Shortly after the second excision, the adenoma recurred to involve the entire nipple-areola complex.

At 3 years of age, the child presented to our center for management because the lesion was tender and bleeding. Our differential diagnosis on clinical examination included pilomatrixoma, breast duct anomaly, and tumor. The patient's medical records and histopathology slides were reviewed from the outside hospital, and the diagnosis of nipple adenoma was confirmed. At 3.5 years of age, the lesion involving the nipple-areola complex was resected with 2-mm margins. The circular wound was closed using a purse-string technique to limit the length of the scar (Fig. 1).



Fig. 1 Three year-old girl with a left nipple adenoma since 5 months of age. A, Preoperative appearance after 2 previous excisions with recurrence. B, Resection of the tumor with 2-mm margins and purse-string closure.

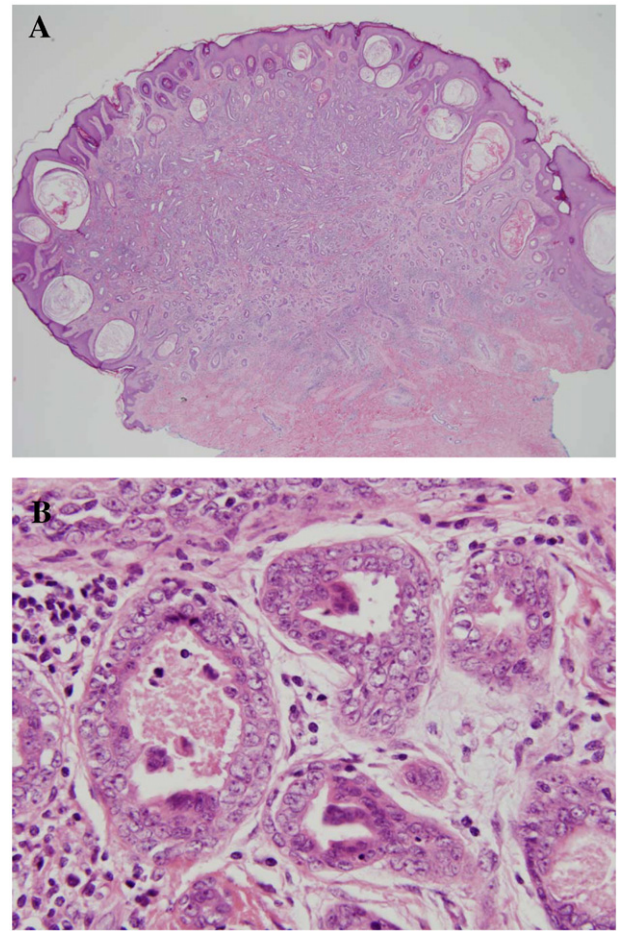


Fig. 2 Histopathology showing nipple adenoma. A, At scanning magnification, the nodule consists of ducts/glands and some dilated lactiferous ducts subepidermally, which are also involved by the process. B, The glands/ducts consist of a bilayered epithelium with enlarged benign cells.

Histopathology showed nipple adenoma with negative margins. A fairly well-circumscribed nodule of benign tubular/glandular structures, separated by a fibrous stroma, with lymphocytes was present. The process blended with and involved some lactiferous ducts subepidermally as well as distal breast ducts. The tubular/glandular structures were lined by plump cells with an inner epithelial and an outer myoepithelial layer, and focally, there was minor pseudopillary epithelial hyperplasia. No significant cytologic atypia was present (Fig. 2). The lesion has not recurred after 7 months of follow-up.

2. Discussion

Nipple adenoma is a rare, benign neoplasm of adulthood; fewer than 500 cases have been reported [4,6,21]. Although the lesion typically occurs in women between 45 and 55 years of age [7], 5 children with nipple adenoma have been described in the literature; all were female between 8 and

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