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# Juvenile papillomatosis of the breast in young male: a case report

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SUMMARY: **Juvenile papillomatosis of the breast in young male: a case report.**

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*Juvenile papillomatosis of the breast ("Swiss cheese disease") is a benign localized proliferative condition of the breast which occurs almost exclusively in young adult women. Patients with this lesion often have a family history of breast carcinoma, but rarely carcinoma may coexist with the lesion at the time of diagnosis. We present a case of a young male with juvenile papillomatosis of the breast. The pathology and clinical management of this rare lesion is discussed.*

RIASSUNTO: **Papillomatosi giovanile della mammella in giovane maschio: descrizione di un caso.**

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*La papillomatosi giovanile della mammella è una patologia quasi esclusiva di giovani donne. Le pazienti hanno spesso storia familiare di cancro della mammella, ma raramente il carcinoma coesiste con la papillomatosi al momento della diagnosi di quest'ultima. Presentiamo un caso di papillomatosi della mammella in un giovane maschio adulto. L'inquadramento della patologia e il relativo trattamento sono ancora controversi.*

KEY WORDS: Juvenile papillomatosis - Adult male.
Papillomatosi giovanile - Uomo adulto.

## Introduction

Juvenile papillomatosis of the breast ("Swiss cheese disease") is a benign localized proliferative condition first described by Rosen and colleagues in 1980 (5). Patients with this lesion often have a family history of breast carcinoma. Rarely carcinoma has been found to coexist with the lesion at the time of diagnosis, although the long-term risk for development of carcinoma in patients with this lesion remains unknown. Macroscopically, this lesion is well circumscribed and contains numerous cysts. Microscopically, this lesion consists of ductal papillomatosis, apocrine and nonapocrine cysts, papillary apocrine hyperplasia, and duct stasis.

This lesion occurs almost exclusively in women younger than 30 years of age. By review of the literature, we have only noted four cases of juvenile papillomatosis in male children, all of whom over age 11 years. We present a case of young male with juvenile papillomatosis of the breast, which represents uncommon description of papillomatosis lesions in either young infants or in young males.

## Case report

A 17 years-old male presented with 2±3 month history of intermittent bloody discharge from the right nipple and a slowly growing mass in the upper outer quadrant of the right breast. This mass measured approximately 2/3 cm, and was firm and well circumscribed. The mass was localized to the subcutaneous tissue, and had a blue discoloration. There was no history of previous breast lesions, and there was no family history of breast carcinoma.

Due to concern of intraductal papilloma or other malignancy, the patient was referred for surgery resection. We performed an excisional biopsy of the mass through a subareolar incision. Intraoperatively, the mass appeared well encapsulated, and extended up to the nipple-areolar complex. This lesion was easily dissected free from normal breast tissue and was removed *in toto*. Recovery was uncomplicated, and there is no evidence of tumor recurrence at 12 months.

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*Pathologic findings*

The breast mass measured 1.5 × 0.6 × 0.4 cm. The external surface was yellow-tan. Bisection revealed multiple small cysts containing serosanguineous fluid. Microscopic sections shoved dilated ducts with occasional hyperplastic epithelium, apocrine metaplasia, and foamy macrophages. Intraductal hemosiderin-laden macrophages were also noted. Structurally there was ductal ectasia with epithelial hyperplasia and hemorrhage, consistent with a diagnosis of juvenile papillomatosis. The cyst lining stained positive for cytokeratin and epithelial membrane antigen by immunostains.

## Discussion

Juvenile papillomatosis of the breast is a benign localized proliferative condition that generally occurs in women younger over age 30 years. This disease was first described by Rosen in 1980 (5), and since that time a number of papers have been published which describe the particular pathologic and epidemiologic characteristics of this unique disease.

In young females, juvenile papillomatosis has been demonstrated to be a marker for the increased risk of breast cancer in the patient's immediate family (7). The significance of a positive family history of breast cancer among first degree relatives of a patient with a borderline or "moderate risk" lesion of atypical or lobular hyperplasia is well established (7). In 1990, Rosen and Kimmel further assessed the long-term risk for a patient with juvenile papillomatosis, and found that patients with a positive family history for breast cancer and recurrent bilateral papillomatosis may be at increased risk for breast cancer themselves (6). In contrast to papillomatosis breast lesions in young women, juvenile papillomatosis of the male breast is an exceedingly rare condition. By extensive review of the literature, we found only four cases of similar epithelial proliferation of the male breast, and all in boys greater than 11 years of age (1±3) (8). Similarly, papillomatosis is generally found in girls over age 10 years, and to the best of our knowledge, this condition has never been reported in a young infant. Proliferative breast conditions in infants are rare lesions (4). In general, early proliferative breast lesions are secondary to the effects of maternal hormonal stimulation, and generally resolve over time without surgical resection. Proliferative breast lesions of infancy almost always represent benign growths which resolve without further intervention, although breast lesions in infants may represent metastases from other sites of cancer. In general, surgery on the prepubertal female breast should be discouraged because of the risk of damage to the breast bud and subsequent breast development. However, breast enlargement which does not decrease after several months post-natally should be closely followed for lesions such as papillomatosis which may require surgical resection. If resection is required, a resection which preserves as much normal breast tissue as possible should be performed.

Our case of young male represent one of reported cases to date with juvenile papillomatosis of the breast. This presentation of a breast epithelial proliferative disorder in a young male represent a novel and previously undescribed clinical variant of juvenile papillomatosis. The effect of this condition on this patient's risk of subsequent breast tumor development is unclear, as is the risk to this patient's family for the development of breast cancer. We recommend that this patient and all other male or female children with juvenile papillomatosis should be followed clinically to detect further lesions which may require surgery.

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