

Intraductal Papilloma with Atypical Ductal Hyperplasia of The Male Breast: A Case Report and A Brief Review

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Abstract

Intraductal papilloma (IP) is a benign tumor, which is very rare in male breast. In this report, we report a 50-year-old Chinese man who presented with a painless mass in his right breast accompanied by occasional small amount of bloody discharge. Mammography and MRI showed a subareolar nodule graded as the Breast Imaging-Reporting and Data System category 4. The patient underwent mammary gland excision through an incision along the lower edge of areola. Histopathologic examination demonstrated multiple intraductal papilloma with atypical ductal hyperplasia (ADH). There are nine cases of IP with ADH have been reported in PubMed (English language) literature until now. The most common presenting symptom is duct blood discharge. It is difficult to distinguish benign and malignant papillary lesions from invasive cancer by radiologically, should have a histopathological evaluation.

Keywords: intraductal papilloma; atypical ductal hyperplasia; male; breast

Introduction

Intraductal papilloma and atypical ductal hyperplasia are very rare conditions in male breast pathology and are rarely reported in the literature. We report a case of a 50-year-old male with a tumor that proved to be multiple intraductal papilloma with atypical ductal hyperplasia and performed a literature review.

Case Report

A 50-year-old male patient was admitted to the hospital with a painless mass of the right breast associated with occasional small amount of bloody discharge for more than 3 months. He had no history of gynecomastia due to local trauma, recent weight loss, use of anabolic steroids, or other medications. Gonadotropin levels were as follows: FSH=5.80IU/L, LH=3.82IU/L, and testosterone=5.63ng/mL. Physical examination

showed a single-duct bloody discharge with tough irregular area on the outside of the right breast areola.

Ultrasound imaging of the breast revealed a 3.1cm×1.2cm×3.3cm hypoecho region with unclear boundary, irregular shape, and abundant blood flow signal(BI-RADS 4b) (**Figure. 1**).Mammography showed an blurred edges, uneven density, irregular dense shadows at 10 and 11 points in the right breast(BI-RADS 0) (**Figure. 2**).Additional magnetic resonance imaging (MRI) showed the lesions in the outer and upper quadrant of the right breast were non-mass enhancement, with segment-like distribution and unclear boundary, and the time signal intensity curve (TIC) curve was inflow type (BI-RADS 4a) (**Figure. 3**).



A



B

Figure 1: (A) Ultrasonographic showing a unclear boundary, irregular shape hypoechoic. (B) PSV: 16.7cm/S.

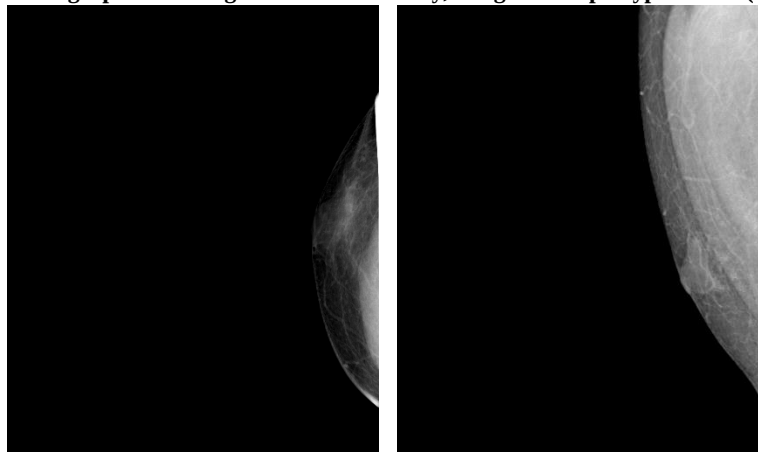
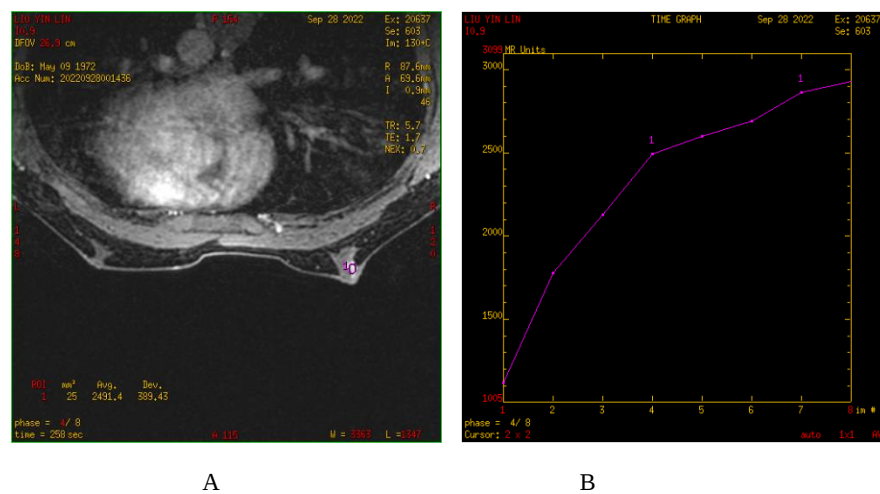


Figure 2: Mammography showing asymmetric dense shadow of right breast.



A

B

Figure 3: (A) MRI showing non mass enhancement, segmental distribution, unclear boundary. (B) TIC: Inflow type.

Preoperative core biopsy showed ductal epithelial hyperplasia without malignancy (**Figure. 4**). Subsequently, mammary gland excision through an incision along the lower edge of areola was performed under general anesthesia. Frozen section analysis confirmed atypical hyperplasia of duct

and Nipple bottom margin negative. Preserve nipple areola complex for patients during operation. The histologic examination demonstrated multiple intraductal papilloma with atypical hyperplasia (**Figures 5,6**).

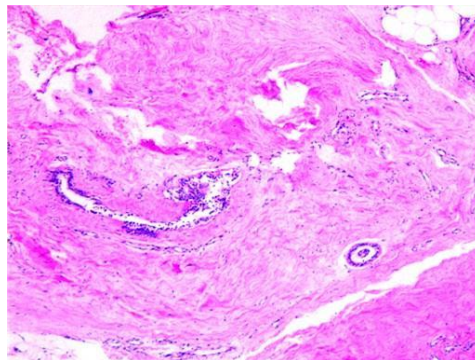


Figure 4: Partially dilated duct epithelial hyperplasia, interstitial degeneration, and hemorrhage. (H + E, ×40)

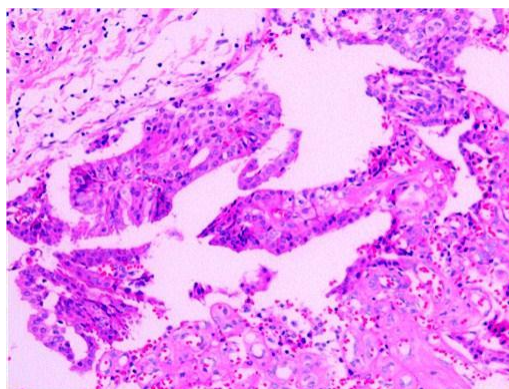


Figure 5: Multiple intraductal papilloma with atypical hyperplasia, individual ductal epithelial hyperplasia. (H + E, ×100)

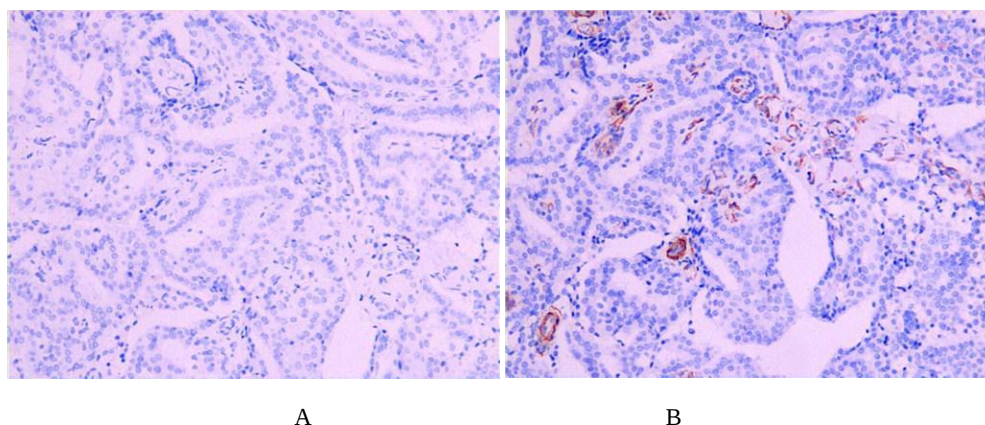


Figure 6: Immunohistochemically showing positivity of P63 (A) and SMA (B) .

The patient underwent mammary gland excision through an incision along the lower edge of areola. There were no postoperative complications, a good cosmetic result was achieved, and the patient was discharged home after surgery. At 3-month follow-up examination, there was no evidence of recurrence.

Discussion

Intraductal papilloma of the breast is commonly seen in 40 to 45 years old women in labor, and rarely in men¹. The actual causes of male IPs are

unclear. Although there is no clear evidence, misuse of anabolic steroids, elevated serum prolactin levels, phenothiazine treatment, and prolonged phenothiazine treatment are all triggers for the development of IP in the male breast². Male nipple discharge is an important sign and should be evaluated with caution. [3] Breast duct endoscopy, ultrasound, mammography and MRI can assist in clinical diagnosis, but the final diagnosis depends on pathology [4].

A systematic search in PubMed revealed only 9 cases of IP in men. The clinical characteristics of patients are presented in **Table 1**.

Report	Age	bloody discharge(Y/N)	ultrasonography	Mammography	MRI	Preoperative biopsy(Y/N)	Histopathology
Sara et al. ^[5]	71	Y	-	-	-	N	IP
Tsilimigras et al. ^[3]	11	Y	BI-RADS 3	-	-	Y(FNAB)	IP
De Vries et al. ^[6]	29	N	BI-RADS 2	BI-RADS 3	-	Y	IP
Szabo et al. ^[7]	42	Y	BI-RADS 3	BI-RADS 3	-	N	IP
IliasVagios et al. ^[2]	55	Y	BI-RADS 4	-	-	N	IP
Kartik Viswanathan et al. ^[8]	45	N	-	-	-	Y	IP
Sushma Bharti ^[9]	60	N	-	BI-RADS 4b	-	Y(FNAB)	IP with ADH
Durkin et al. ^[10]	14	Y	BI-RADS 4a	-	-	N	IP
Gaurav Anand BLA ^[11]	52	Y	BI-RADS 4	BI-RADS 4a	-	Y	IP

Table 1: Reported cases of intraductal papilloma in men

-: unknown.

IP: intraductal papilloma

FNAB: fine needle aspiration biopsy

ADH: Atypical ductal hyperplasia

BI-RADS: Breast Imaging-Reporting and Data System

The median age of IP patients is 45. 66.7% (6/9) of the patients had clinical signs of bloody nipple discharge, 66.7% (6/9) of the patients underwent ultrasound examination, of which 50.0% (3/6) were assessed as BI-RADS 4, 44.4% (4/9) were assessed by mammography, and 50.0% (2/4) of the patients were classified as BI-RADS 4. 55.6% (5/9) patients were diagnosed by aspiration biopsy preoperatively, including 2 cases of fine needle aspiration biopsy (FNAB) and 3 cases of core-needle biopsy. The role of FNAB in the diagnosis of benign and malignant papillary lesions remains controversial. Study of Aylin Simsir et al [12]. found that lesions displaying a papillary pattern on FNAB are nonpapillary on follow-up. Among benign processes, fibrocystic degeneration and fibroadenoma may be cytologically similar to papilloma.

Sydney et al. found that the accuracy of core biopsy in diagnosing benign papilloma was 97% [13]. In this case, we performed core-needle biopsy before operation, and 3 specimens were obtained. The pathological results failed to directly diagnose intraductal papilloma, only indicating that no malignant tumor cells were found. The reason may be insufficient specimen were obtained by preoperative aspiration biopsy.

There is no previous report on the application of MRI in male IP. The study on MRI of papillary breast diseases by Sarica et al. concluded that the main benefits of post areola imaging are its ability to show the relationship between ducts and the extension of enhanced areas¹⁴. In this case, we evaluated the lesion with MRI. A slightly longer T1 and slightly longer T2 signal lesion was seen in the upper quadrant of the right outer breast. DWI sequence showed slightly higher signal intensity, and ADC image signal was slightly reduced; After 15ml of Gd-DTPA was injected intravenously, the scanning showed that the lesions in the upper quadrant outside the right breast were non mass enhanced, distributed in segments, with unclear boundaries, and the larger slice area was about $1.4 \times 1.8 \times 1.2$ cm, TIC curve is inflow type.

Based on the patient's imaging evaluation and preoperative pathological results, we finally prepared the mammary gland excision for the patient. At the same time, according to the frozen section during the operation, we ensured that the bottom cutting edge of the nipple and areola was negative while retaining the patient's nipple and areola complex. There were no complications and the cosmetic effect was good.

Conclusion

Male IP is extremely rare, and bloody nipple discharge is the most common clinical presentation. Core-needle biopsy is helpful for preoperative diagnosis, and breast MRI evaluation can be used to determine surgical treatment regimen.

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