Int J Clin Exp Pathol 2021;14(6):782-785
www.ijcep.com /ISSN:1936-2625/IJCEP0129417

Case Report

Sequential occurrence of different subtypes of nonpuerperal mastitis in contralateral breasts: a report of two cases

Yun Ren, Jianzhong Xu, Xiaoxia Li, Bo Zhao, Jiao Zhang, Jindan Zhang

Department of Breast Surgery, People's Hospital, Changzhi 046000, Shanxi, China

Received January 1, 2021; Accepted April 14, 2021; Epub June 15, 2021; Published June 30, 2021

Abstract: Background: Nonpuerperal mastitis (NPM) is a nonspecific chronic inflammation in the breast. The two most common pathologic subtypes are idiopathic granulomatous mastitis (IGM) and plasma cell mastitis (PCM). The cause of NPM is unknown, and the recurrence rate is high; however, the sequential development of two different pathologic subtypes of NPM in contralateral breasts of the same patient is rare. Case summary: Two women who were in their 30 s and with a history of childbearing had an inflammatory mass in one breast, which was identified as PCM by core needle biopsy. However, 1 year after treatment, IGM occurred in the contralateral breast. Histopathologic results after the surgery confirmed both diagnoses. New-onset PCM in both patients was successfully treated with local extended surgical resection and antibiotics combined with incision and drainage. The IGM that developed in the contralateral breast after 1 year was treated with local extended surgical resection and oral steroids. No signs of NPM recurrence were observed in either breast after a follow-up period of more than 1 year. Conclusion: Because recurrence after NPM treatment often occurs in the affected breast, the contralateral breast, which does not often exhibit morbidity factors, is often ignored during treatment follow-up. However, our cases suggest that patients with the occurrence of NPM in one breast should undergo long-term follow-up of the contralateral breast by close monitoring. NPM recurrence should also be considered when there is pain, redness, and swelling in the contralateral breast.

Keywords: Nonpuerperal mastitis, plasma cell mastitis, idiopathic granulomatous mastitis, breast, case report

Introduction

Nonpuerperal mastitis (NPM) is a type of nonspecific chronic inflammation in the breast. It has multiple pathologic subtypes, and the two most common of these subtypes are idiopathic granulomatous mastitis (IGM) and plasma cell mastitis (PCM) [1, 2]. NPM predominantly occurs in women who are in their 30 s and 40 s and with a history of childbearing. The main clinical manifestations are inflammatory masses and abscesses. The cause of NPM is unknown, and patients frequently experience relapse [1-4]. Currently, the most common clinical treatment is local extended surgical resection, which can significantly reduce the recurrence rate [3, 4].

In clinical practice, the occurrence of both IGM and PCM in different breasts of the same patient is rare. Herein, we report two cases of PCM and IGM that occurred sequentially in contralateral breasts of the same patient. The patient was completely cured of the disease after a series of treatments, including incision and drainage as well as surgery, and after more than 1 year of follow-up, neither of the two patients experienced relapse of NPM in either breast.

Case report

The study protocol was approved by the Ethics Committee of the Changzhi City People's Hospital and relevant institutions (serial number: 202002) on February 10, 2020. Written informed consent was obtained from both patients in this study.

Case 1

A 31-year-old woman with two children aged 3 years and 7 years had congenital inversion of the right nipple. Her left nipple developed nor-

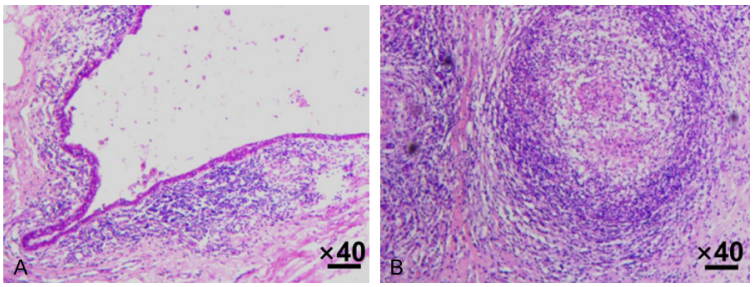

Figure 1. Case 1 images. A. Histopathology of the right breast mass confirming plasma cell mastitis: plasma cells, neutrophils, and lymphocytes are seen around the ectatic duct of the breast (H&E stain; magnification: ×40). H&E = hematoxylin and eosin. B. Histopathology of the left breast mass confirming idiopathic granulomatous mastitis: non-caseating granulomas composed of Langerhans giant cells accompanied by lymphocytes, plasma cells, and epithelioid histiocytes are found within and around the lobules (H&E stain; magnification: ×40).

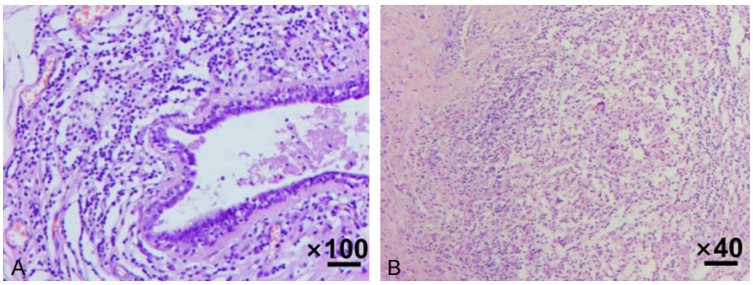

Figure 2. Case 2 images. A. Histopathology of the left breast mass confirming plasma cell mastitis (H&E stain; magnification: ×100). B. Histopathology of the right breast mass confirming idiopathic granulomatous mastitis (H&E stain; magnification: ×40).

mally. In July 2017, she found abscess-like lesions in her right breast under the areola. Within a week, the mass gradually increased to a size of approximately 8×7×5 cm, accompanied by swelling of the skin and pain. The mass was identified as PCM by core needle biopsy, and histologic examination showed obvious chronic inflammation, with predominance of plasma cells, neutrophils, and lymphocytes, as well as focal steatonecrosis (**Figure 1**). After incision and drainage, the breast inflammation significantly decreased, and the tumor size was reduced to 5×4×3 cm. Subsequently, extended resection and nipple retraction correction were performed to remove the residual inflammation under the nipple, which consisted of lumps and diseased breast ducts that made the nipple protrude outward, and to restore its original configuration. Postoperative specimens showed typical inflammatory infiltrates around the lesion's ducts in the right retroareolar region, as well as abscess-like lesions, which confirmed the diagnosis of PCM. The lesion did not recur after the surgery.

In July 2018, the patient experienced mild intermittent pain in the medial quadrant of the left breast, but we did not find any associated mass. To treat the patient for the symptoms, the patient took only ibuprofen sustained-release capsules for 1 week. However, a 6.5×5×5 cm inflammatory mass suddenly developed in this location. Color Doppler ultrasound showed abscess formation in the mass, and it was identified as IGM on core needle biopsy. After treatment with oral methylprednisolone tablets (0.5 mg/kg/day) for 1 month, the mass gradually decreased to a size of 4.5×3.5×3 cm. The patient then underwent extensive resection to remove the residual lesion. In the subsequent surgical resection specimens, non-caseating granulomatous inflammation was observed in the breast lobules, which again confirmed the pathologic diagnosis of IGM. Pathologic examination did not reveal infection with *Mycobacterium tuberculosis*, and laboratory testing ruled out the presence of bacterial infection. After 24 months of follow-up, there was no disease recurrence in either breast.

Case 2

In July 2017, a 34-year-old woman with congenital inversion of both nipples, who had a 2-year-old child, found an inflammatory mass under the left areola with obvious skin redness and pain. The patient's breast sonogram revealed a 5.5×5×4 cm inflammatory mass with internal abscess formation. Core needle biopsy confirmed the mass as PCM, and the lesion was successfully treated with multiple incision and drainage treatments, as well as 5 days of antibiotics (levofloxacin at 400 mg/day by intravenous infusion). Several old incision and drainage scars remained visible on the skin of the left breast (**Figure 2**).

In September 2018, a "peanut"-sized mass suddenly developed in the patient's right breast (at the 6 o'clock position, 3 cm distal to the nipple-areolar complex), accompanied by mild pain. The patient was again treated with antibiotics (levofloxacin at 400 mg/day by intravenous infusion) for 5 days. However, the treatment was ineffective, and the right breast mass increased to a size of 6.3×5.5×3 cm, accompanied by local skin redness and obvious pain. The mass was confirmed to be IGM on core needle biopsy. After 1 month of oral methylprednisolone treatment (0.5 mg/kg), the breast inflammation gradually diminished, and the tumor size gradually decreased to 4.7×3.5×2 cm. Extensive resection was then performed. Postoperative pathological results again confirmed that the right breast mass was IGM. After 18 months of follow-up, there was no recurrence of breast disease.

Discussion

This study showed a similar pathogenesis of NPM in the two patients. First, PCM occurred in breasts with an inverted nipple. The mass was successfully treated with surgery or incision and drainage, but 1 year later, IGM occurred in the contralateral breast. The IGM was also successfully treated with oral steroids combined with local extended surgical resection. After more than 1 year of follow-up, there was no recurrence of NPM in either breast. It is worth noting that after PCM was treated in the affected breast of both patients, the possibility of NPM recurrence in the contralateral breast was ignored. The early symptoms of NPM and possible trigger factors of recurrence, such as the presence of an inverted nipple, were also ignored.

NPM is a benign disease, but it can severely deform the breast. Some studies proposed that the main cause of PCM is the blockage and infection of the large retroareolar ducts due to an inverted nipple. However, some studies have reported that PCM may be related to autoimmune diseases [5, 6]. PCM mainly involves the large retroareolar mammary ducts. Apart from breast pain and subareolar masses, an important clinical manifestation of PCM is inverted nipples [3, 5], which was seen in the two PCM patients in this study. The most common treatments for early-stage PCM with subareolar abscess are antibiotics and incision and drainage [7, 8]; these treatments were used in our second case. However, the most effective methods to treat PCM, namely, extensive lesion resection and inverted nipple correction [3, 8], were used in our first case.

Granulomatous mastitis often manifests clinically as masses, abscesses, and sinus formation in the breast. There are diverse treatment modalities for IGM, including antibiotics, steroids, surgery, and immunosuppressants [9]. However, there has been no consensus on a standard treatment. Presently, a combination of steroids and surgery is acknowledged as the most effective treatment regimen [10, 11]. In the two patients reported in this case study, the lesion was effectively treated using this combination therapy.

Distinguishing between the two subtypes of NPM requires accurate pathologic examination. Histologically, PCM is characterized by large numbers of plasma cells, lymphocytes, and macrophages infiltrating the large retroareolar mammary ducts, while IGM is a type of non-caseous granulomatous inflammation centered on the terminal ductal-lobular unit of the mammary gland [3, 6].

One of the clinical features of NPM that confounds physicians is its high recurrence rate after treatment [7, 12]. NPM normally recurs as a lesion of the same pathologic subtype immediately beside the original lesion in the affected breast. It is rare that two different subtypes of NPM occur sequentially in contralateral breasts, as observed in this study. Therefore, any occurrence of NPM in one breast should be followed by close monitoring and long-term follow-up of the contralateral breast. The possibility of NPM recurrence should also be considered when there is pain, redness, and swelling in the contralateral breast. It remains unclear whether autoimmune processes affect the pathogenesis of both IGM and PCM; thus, further research in this area is warranted.

Acknowledgements

This work was supported by the Soft Science Research Project of the Shanxi Province (No. 2018041032-2).

Written informed consent was obtained from both patients in this study.

Disclosure of conflict of interest

None.

Address correspondence to: Jindan Zhang, Department of Breast Surgery, Changzhi People's Hospital, No. 502 Changxing Middle Road, Changzhi 046000, Shanxi Province, China. E-mail: zjd330903008@163.com

References

[1] Gopalakrishnan Nair C, Hiran, Jacob P, Menon RR and Misha. Inflammatory diseases of the non-lactating female breasts. Int J Surg 2015; 13: 8-11.

[2] Geshon-Cohen J and Ingleby H. Secretory disease and plasma cell mastitis in the female breast; roentgenologic and pathologic studies. Surg Gynecol Obstet 1952; 95: 497-504.

[3] Ming J, Meng G, Yuan Q, Zhong L, Tang P, Zhang K, Chen Q, Fan L and Jiang J. Clinical characteristics and surgical modality of plasma cell mastitis: analysis of 91 cases. Am Surg 2013; 79: 54-60.

[4] Co M, Cheng VCC, Wei J, Wong SC, Chan SM, Shek T and Kwong A. Idiopatq1hic granulomatous mastitis: a 10-year study from a multicentre clinical database. Pathology 2018; 50: 742-747.

[5] Al-Masad JK. Mammary duct ectasia and periductal mastitis in males. Saudi Med J 2001; 22: 1030-1033.

[6] Martinez-Ramos D, Simon-Monterde L, Suelves-Piqueres C, Queralt-Martin R, Granel-Villach L, Laguna-Sastre JM, Nicolau MJ and Escrig-Sos J. Idiopathic granulomatous mastitis: a systematic review of 3060 patients. Breast J 2019; 25: 1245-1250.

[7] Versluijs-Ossewaarde FN, Roumen RM and Goris RJ. Subareolar breast abscesses: characteristics and results of surgical treatment. Breast J 2005; 11: 179-182.

[8] Lannin DR. Twenty-two year experience with recurring subareolar abscess and lactiferous duct fistula treated by a single breast surgeon. Am J Surg 2004; 188: 407-410.

[9] Farouk O, Abdelkhalek M, Abdallah A, Shata A, Senbel A, Attia E, Abd Elghaffar M, Mesbah M, Soliman N, Amin M and El-Tantawy D. Rifampicin for idiopathic granulomatous lobular mastitis: a promising alternative for treatment. World J Surg 2017; 41: 1313-1321.

[10] Deng JQ, Yu L, Yang Y, Feng XJ, Sun J, Liu J, Fan FS and Liao LQ. Steroids administered after vacuum-assisted biopsy in the management of idiopathic granulomatous mastitis. J Clin Pathol. 2017; 70: 827-831.

[11] Shin YD, Park SS, Song YJ, Son SM and Choi YJ. Is surgical excision necessary for the treatment of granulomatous lobular mastitis? BMC Womens Health 2017; 17: 49.

[12] Yilmaz TU, Gürel B, Güler SA, Baran MA, Erşan B, Duman S and Utkan Z. Scoring idiopathic granulomatous mastitis: an effective system for predicting recurrence? Eur J Breast Health 2018; 14: 112-116.