DOI: https://doi.org/10.2298/SARH200914006R
UDC: 618.19-002

CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Idiopathic granulomatous mastitis – new approach in operative treatment

Milan Ranisavljević, Nenad Šolajić, Ferenc Vicko

Oncology Institute of Vojvodina, Sremska Kamenica, Serbia;
University of Novi Sad, Faculty of Medicine, Novi Sad, Serbia

SUMMARY

Introduction Idiopathic granulomatous mastitis (GM) is described as a very rare, non-lactating, chronic mastitis that occurs primarily in women of childbearing age. Significant clinical problem related to GM is the diagnostic differentiation from breast cancer. Less advanced forms of GM can be successfully treated with limited surgical excisions and radical treatment is recommended only for the most extensive forms.

Case report First examination of the patient, by the surgeon at Oncology Institute of Vojvodina was in December 2018, when initial suspicion of breast cancer was set up. Core needle biopsy was performed and after histopathological (HP) analysis, confirmation of GM was obtained. The patient was initially offered Prednisone and Methotrexate therapy, which she refused and accepted only surgical treatment. Surgical treatment was performed few weeks after needle biopsy and consisted of performing a nipple spearing mastectomy with excision of the orifices of all fistulous ducts and their primary sutures. The HP findings of the operative specimen confirmed the diagnosis of GM. While there were no signs of dieses relapse, patient was suggested secondary reconstruction of the left breast. Twelve months after the primary operation, secondary breast reconstruction was performed with the interposition of a contoured silicone implant into a muscle pocket in a standard manner.

Conclusion Nipple spearing mastectomy with secondary breast reconstruction is esthetically satisfactory treatment for patients with locally advanced GM.

Keywords: idiopathic granulomatous mastitis; nipple spearing mastectomy; secondary breast reconstruction

INTRODUCTION

Idiopathic granulomatous mastitis (GM) is described as non-lactating chronic mastitis that occurs primarily in women of childbearing age, but could be found in patients many years after breast feeding. Kessler and Wolloch [1] have first described this rare illness in 1972. Since the clinical and radiological imaging of GM are similar to the breast cancer, it could lead to misdiagnosis before the final histopathological (HP) diagnosis.

Many authors suggested that possible etiology of GM include breast infection with microbes, autoimmune disorders and hyperprolactinemia [2]. Uncertain etiology leads to non-optimal treatment for GM and included watch and wait strategy, antibiotics, steroids and surgery as earliest and most widely used therapy [3–8]. In cases when GM is localized in only one part of the breast, local excision is used and radical treatment is recommended only for the most extensive forms, when all breast tissue is involved [2, 3, 5].

CASE REPORT

First examination of the patient by the surgeon at the Oncology Institute of Vojvodina was in December 2018, when initial suspicion of breast cancer was set up. In history patient has one child (nine years old), non-smoker, with a negative family history of breast cancer. Ultrasound findings supplied by the patient are characterized as BI RADS 5 in the left breast. On clinical examination in the left breast, at the border of the lateral quadrants a tumor mass, about 5 cm in diameter, was palpated, which protrudes and deforms the skin of the breast with three active fistulous ducts (two located in upper medial quadrant and one periareolar in the same quadrant) from which the clear secretion is conducted (Figure 1).

Core needle biopsy was performed a few days after the clinical examination and HP analysis of the specimen, which confirmed the GM diagnosis (Figure 2).

After a core needle biopsy, a few more fistulas were produced (in upper lateral quadrant), with pronounced inflammation of the left breast skin, and *per os* antibiotics (augmentin + metronidazole) were included in the therapy. The inflammatory process affected almost all of the breast tissue except the area just below the nipple areola complex.

After the inflammatory process calming down and spontaneous closing of the fistulous ducts in upper lateral quadrant, the patient performed tests for tuberculosis, sarcoidosis and Wegener's granulomatosis, and all of them were negative. Smears taken from fistulous ducts

Received • Примљено: September 14, 2020
Revised • Ревизија: November 22, 2021
Accepted • Прихваћено: December 28, 2021
Online first: January 14, 2022

Correspondence to:
Milan RANISAVLJEVIĆ
University of Novi Sad
Faculty of Medicine
Hajduk Veljkova 3
21000 Novi Sad
Serbia
milan.ranisavljevic@mf.uns.ac.rs

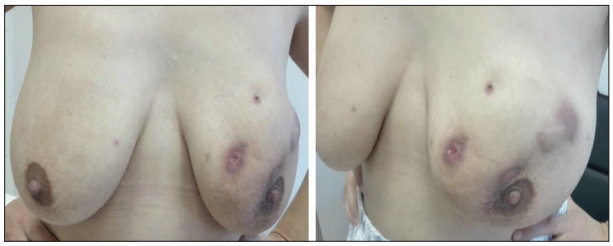

Figure 1. Clinical findings one day before the operation; three active fistulous ducts; palpable tumor masses in lateral quadrants of the left breast (approximately 5 cm in diameter)

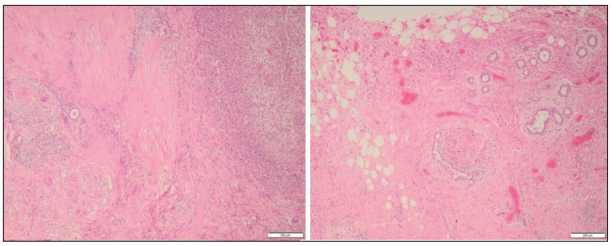

Figure 2. Histopathological findings of granulomatous mastitis; A – preserved lobular architecture of the mammary gland (lower left) and an abscess rimmed by granulomatous inflammatory infiltrate (upper right); hematoxylin and eosin, 20 ×; B – a granuloma containing numerous giant cells protruding into a duct; hematoxylin and eosin, 20 ×

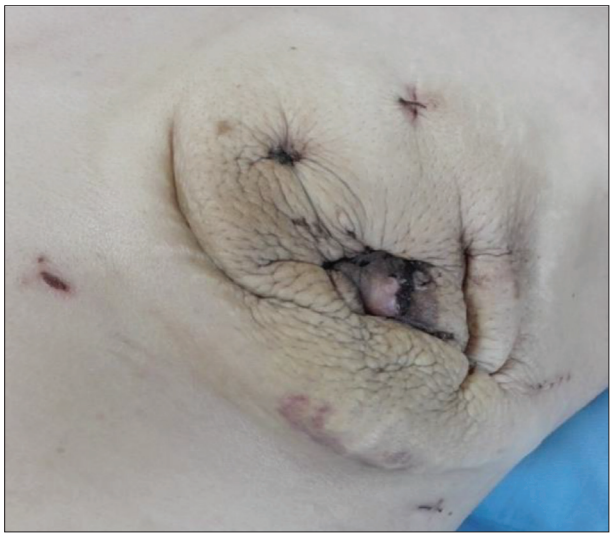

Figure 3. Clinical findings after the surgery and removing the drains; stitches on the sites from excided fistulous canals; two incisions on the border of the breast and abdominal skin are from drains; vital skin flap, after performing nipple spearing mastectomy with nipple areola complex preservation

were negative on bacteria and fungi. The patient was initially offered Prednisone and Methotrexate therapy, with the presentation of all possible positive and negative effects of the proposed therapy, which she refused and accepted only surgical treatment.

Surgical treatment was performed few weeks after the HP conformation of the diagnosis and consisted of performing a nipple areola complex spearing mastectomy with excision of the orifices of all fistulous ducts and their primary closing with adsorptive stitches (Vicryl 5-0, Ethicon, Ethicon Inc., Raritan, NJ, USA). Intraoperative, many clear collections of 2–5 centimeters in diameter were found within the breast tissue, with a pronounced inflammatory component, which blended with the surrounding healthy breast tissue. The operative wound was drained with two 21 Gauge drains. Operative wound (radial incision on the border of lateral quadrants) is closed with adsorptive stitches (Vicryl 5-0).

Antibiotic was not used in postoperative treatment while there were no signs of operative wound infection.

After reducing the secretion on the drain bags, and removing the drains, the patient was discharged from hospital for further home treatment. Local findings after the operation are shown on Figure 3. Definitive HP analysis of operative specimen confirmed GM (Figure 2).

After the operation, the patient was regularly monitored by the surgeon on a monthly basis.

Since there were no signs of disease relapse (12 months after primary operation), the patient was suggested secondary reconstruction of the left breast with silicone breast implants. Postoperative breast ultrasound finding was without signs of disease in the left breast.

Secondary breast reconstruction was performed with the interposition of a contoured 530 cc silicone breast implant (Mentor Medical Systems B.V, Leiden, Netherlands) into a muscle pocket made of a large pectoralis mayor and serratus anterior muscle in a standard manner with liberation of breast skin from muscles.

One year after the secondary operation there are no signs of local relapses and patient is very satisfied with esthetical results (Figure 4).

This case report was approved by the institutional ethics committee, and written consent was obtained from the patient for the publication of this case report and any accompanying images.

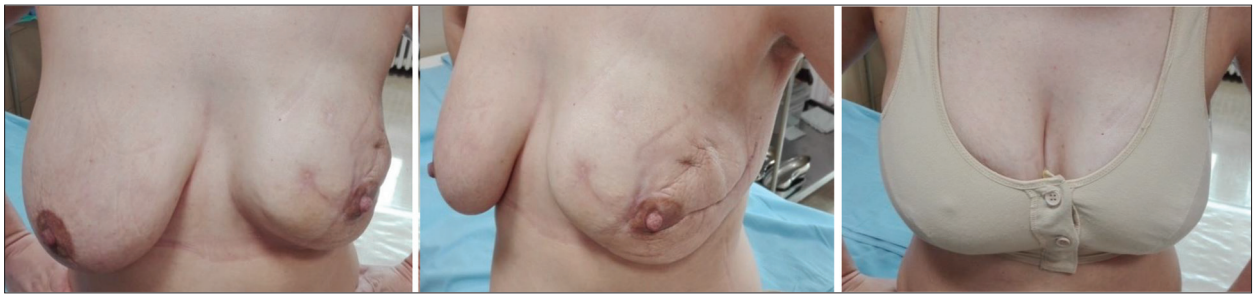

Figure 4. Clinical findings after the secondary reconstruction using silicone breast implants; postoperative clinical finding one month after the operation; reconstructed breast is smaller than the healthy one; in this operative treatment, we have not made contralateral breast reduction since the patient has shown no interest in performing that procedure

DISCUSSION

According to our findings, this was the first case of secondary breast reconstruction using contoured silicone implants in GM treatment.

Both surgical and nonsurgical (antibiotic therapy, oral steroids, observation) treatments have been advocated as the first-line treatments of GM [3–8]. Because the clinical and imaging features of GM are very similar to those of breast carcinoma, tissue biopsy remains the gold standard to confirm the diagnosis [9].

Li [10] believes that GM is self-limiting disease with good prognosis and suggests that conservative management with close surveillance would be the best treatment modality. In his study, 50% of the patients had spontaneous complete resolution of disease after 14.5 months and did not relapse. When compared to other therapeutically modalities, observation therapy has the longest recovery time causing physical and emotional pain [8, 10].

GM is mostly represented like breast infection with large abscesses and antibiotics (a combination of amoxicillin and metronidazole) are usually used, but many studies have shown no benefit of this therapeutic model [6, 9, 11]. Many authors have found that GM is in some cases related to anaerobic Corynebacterium infection [12. 13, 14].

Some researchers believe that the pathogenesis of GM may be an autoimmune response to the secretion of mammary ductal proteins, so they use steroids to treat GM and some positive results have been achieved [10, 15]. Complete clinical and radiological regression was observed in 63% of the patients when using methylprednisolone (0.5 mg/kg/day for four weeks) but with high recurrence rate (31%) and longer recovery time [15]. Steroid therapy also has a notable problem that it may have side effects at high doses. Immunosuppressive therapy is recommended for patients who have relapsed after steroid therapy and have steroid resistance or unbearable side effects, but the therapeutic effect of methotrexate remains unclear [6]. Otherwise, best recovery time is shown after the use of methylprednisolone and surgery (one month) versus using only steroid therapy (six months) ($p = 0.001$) [16, 17].

In terms of recurrence and post-treatment recovery, surgery has been one of the main treatments since GM was first reported, and studies including surgical resection as a first-line treatment showed significantly superior results compared with steroid therapy alone [3, 5, 6, 16, 17]. Possible surgical treatments include breast conserving surgery and mastectomy (depending on the size of involved breast tissue with GM [3, 5, 6, 16, 17]. Like nonsurgical treatment options, surgery, as well, has some disadvantages, primarily bad postoperative aesthetic effect and loss of breastfeeding. After the breast conservative surgery, recurrence rate of GM is 10%, and after the mastectomy 0% [6].

Performing nipple spearing mastectomy with secondary breast reconstruction using silicone breast implants is aesthetically satisfactory treatment for patients with locally advanced GM. Further studies are needed to confirm this hypothesis.

Conflict of interest: None declared.

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Идиопатски грануломатозни маститис – нов приступ хируршком лечењу

Милан Ранисављевић, Ненад Шолајић, Ференц Вицко

Институт за онкологију Војводине, Сремска Каменица, Србија;
Универзитет у Новом Саду, Медицински факултет, Нови Сад, Србија

САЖЕТАК

Увод Идиопатски грануломатозни маститис (ГМ) изузетно је ретка врста хроничне упале дојке која се обично јавља код жена након порођаја. Значајан диференцијално дијагностички проблем код ГМ је то што клинички имитира карцином дојке. Мање форме ГМ успешно се могу третирати ограниченим ексцизијама, а радикални третман препоручен је само у случајевима екстензивних форми које захватају већи део ткива дојке.

Приказ болесника Први преглед болеснице од стране хирурга на Институту за онкологију Војводине био је у децембру 2018. године, када је постављена иницијална сумња на постојање карцинома леве дојке, с обзиром на клинички налаз. Урађена је иглена биопсија туморске масе, а патохистолошки налаз говорио је у прилог ГМ. Болесници је иницијално понуђена терапија преднизоном и метотрексатом, коју је она одбила и прихватила једино оперативни третман. Операција је учињена неколико недеља након иглене биопсије, када је изведена супкутана мастектомија са презервацијом комплекса ареоле и мамиле уз ексцизију свих отвора фистулозних канала и њихову примарну сутуру. Патохистолошки налаз оперативног материјала потврдио је дијагнозу ГМ. С обзиром на то да није дошло до релапса болести након 12 месеци од примарне операције, болесници је предложено да се изведе секундарна реконструкција леве дојке. Изведена је контурираним силиконским имплантом који је пласиран у мишићни џеп на стандардан начин.

Закључак Супкутана мастектомија уз секундарну реконструкцију дојке естетски је прихватљив третман код болесница са локално узнапредовалим обликом ГМ.

Кључне речи: идиопатски грануломатозни маститис; супкутана мастектомија; секундарна реконструкција дојке