



Case Report

Post-operative Ayurvedic management of non-healing idiopathic granulomatous mastitis - A case report

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ABSTRACT

Idiopathic granulomatous mastitis (IGM) is a chronic inflammatory disease of the breast which may present as painful, unilateral, discrete, breast masses with a tendency to recur. The pathogenesis is still unclear. In Ayurvedic literature, clinical features similar to IGM is seen in *Stanavidradhi* (breast abscess). We present the case of a 25-year old uniparous woman who had previously undergone excision biopsy for a breast lump and was diagnosed with IGM by histopathology examination. After one month, she developed pain, redness, and swelling at the operated site. She was treated for two months with corticosteroids and antibiotics, but the symptoms did not subside. After Ayurvedic treatment, the condition started to respond in a week and was completely resolved within three months. She was followed-up with medicines for three months and without medicines for another six months during which period, no recurrence was seen. The non-healing nature and high rate of recurrence of IGM necessitates prolonged treatment with steroids and antibiotics that have long-lasting adverse effects. In this context, Ayurveda may be able to offer an effective option with significantly fewer adverse effects, not only in the management of abscess and sinus, but also in prevention of recurrence.

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1. Introduction

Idiopathic granulomatous mastitis (IGM) or granulomatous lobular mastitis is a chronic inflammatory disease of the breast which was first reported by Kessler and Wollock in 1972 [1]. This entity is commonly reported in women of childbearing age. Rarely, this has been reported in young girls, post-menopausal women, and men [2]. Till lately, IGM was considered to be a rare entity; however, more recent reviews suggest that this may not be the case. Emerging reports of a large number of cases especially in developing countries point to an increased prevalence in these nations [3]. IGM may present as painful, unilateral, discrete breast masses with a tendency to recur [4]. There may also be inflammation and skin ulceration with fistula formation and axillary lymphadenopathy. Nipple retraction, ulceration, and secretions have been documented but occur rarely. The pathogenesis of IGM is still unclear. Multiple factors such as hormonal imbalance, autoimmunity, trauma, Corynebacterium infection, alpha-1-antitrypsin deficiency, and smoking have been implicated as causes or

triggering factors [5]. One hypothesis regarding the pathogenesis of IGM suggests that extravasation of secretions from the breast lobules causes an inflammatory response in the connective tissue ultimately leading to a local granulomatous response [6]. The differential diagnosis of IGM includes mastitis, carcinoma, systemic diseases such as tuberculosis, sarcoidosis, erythema nodosum, and Wegener granulomatosis. It is differentiated from mastitis especially when the post-excisional infection does not respond to antibiotics [7]. Radiographic findings of IGM manifests a variety of non-specific appearances and is often mistaken for malignancy resulting in unnecessary mastectomies [8]. A definitive diagnosis requires biopsy and histopathology examination. Excisional biopsy has both a diagnostic and therapeutic role [9]. The lack of a non-invasive diagnostic tool adds to the limitation of diagnosis and hence, IGM is often under or misdiagnosed. Treatment options explored depend on the extent of the lesion and degree of severity. Optimal treatment for IGM is still under contention and includes corticosteroids, wide excision, antibiotics, etc [10]. The role of steroids is debated, as in some patients, IGM is reported to relapse quickly on stopping steroids [11]. IGM has a high rate of recurrence and is to be suspected if there is reappearance of fistula, abscess, or mass on the same breast. Recurrence may occur years after initial treatment and requires prolonged follow-up.

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In Ayurvedic literature, clinical features, similar to IGM are reported as *Stanavidradhi*. *Vidradhi* is a round or elongated swelling of the connective tissues associated with severe pain and burning sensation. *Vidradhi* is broadly classified as *bahyavidradhi* and *antarvidradhi*. *Bahyavidradhi* appears as visible external swellings or superficial abscesses. *Antarvidradhi* is a deep abscess affecting internal structures and is much more painful and fatal compared to *bahyavidradhi*. *Stanavidradhi* is elaborated by Acharyas Susrutha, Vagbhata, and Charaka. Acharya Susrutha explains that vitiated *vata* and *doshas* vitiates *rakta* and *mamsa* and produce *stanaroga* (disease of breast) which exhibits the features of *bahya vidradhi* [12]. Unhealthy diet and lifestyle practices are implicated in the pathogenesis of *stanavidradhi*. While treating *stanavidradhi* or breast abscess, Acharya Vagbhata advises the treatment of *sopha* (inflammation) in the initial stage and that of *vrana* (wound/ulcer) after rupture of abscess [13]. As the present case presented after two months of onset with pain, redness, and oozing from sinus, it was considered as *dushtavrana* (chronic/infected ulcer) and treated likewise. A wide range of treatment options that can be used to treat *vrana* are elaborated in the classical texts and are decided upon by taking into consideration the predominance of *doshas* and other physiological and pathological factors involved.

2. Patient information

We present here, the case of a 25-year old uniparous female who underwent excision biopsy for a breast lump and was diagnosed with IGM by histopathology examination. The lump (6 cm × 5 cm) was in the lower inner quadrant of the left breast at 7–9 o'clock position. Mammogram reported BI-RADS 4a. Ultrasound-guided left breast biopsy revealed marked inflammation around the lobules with destruction and presence of histiocytes and occasional giant cells with numerous neutrophils. The inflammation extended

to the interlobular stroma and also into the surrounding fat. Pockets of abscess formation were observed. Histopathologic diagnosis was lobulocentric granulomatous inflammation with abscess formation probably, idiopathic. The patient had no history of tuberculosis, sarcoidosis, or other infectious or granulomatous disease. No relevant family history was found. After one month of excision, she developed pain, redness, and swelling at the operated site. She was treated for two months with corticosteroids and antibiotics, but the symptoms did not subside and the pain increased. Anxious about the adverse effects of long-term steroid use, the patient decided to seek alternate options and consulted us at the Prasutitantra and Streeroga Department, Govt. Ayurveda College, Tripunithura, Kerala. Past medical history with signs and symptoms, date of hospital visit, investigation reports, and intervention is shown in Table 1.

3. Clinical findings

The patient presented with severe pain, redness, and oozing from a sinus in the left breast on 1/5/2020. She was para one, her last childbirth was four years back and she had breastfed her baby for two and half years. She was afebrile with normal rhythmic pulse 78/min and respiratory rate 22/min. Blood pressure was 110/70 mm Hg and weight was 55 kg. On examination her right breast appeared normal. The left breast was tender with redness and oozing from a sinus at 4-5 o'clock position. There was no retraction of either nipples. Small axillary lymph nodes were palpable on both sides.

4. Therapeutic intervention

The prognosis of this case was *krichrasadhya* as the site of *vrana* was the breast [14]. Moreover, the condition had become chronic and was not responding to high doses of steroids and antibiotics.

Table 1
Showing past medical history with symptoms and signs, date of hospital visit, investigation reports and intervention.

Symptoms and signs	Date of hospital visit	Investigations	Interventions
Breast lump with pain O/E- lump of 6 cm × 5 cm in the lower inner quadrant of the left breast at 7–9 o'clock position.	28/2/2020	Mammogram - BI-RADS 4a	Excision biopsy
Pain in the operated site O/E-no redness, no fever, minimal induration lateral to scar	21/3/2020		Tab Wysolone (Prednisolone) 10 mg OD and Tab Eterocoxib 90 mg, advice to come for review if pain persisted
Pain along with pus discharge	26/3/2020	Pus aspirated for culture and sensitivity.	Bactrim DS (Sulfamethoxazole) and Prednisolone 10 mg OD continued.
Pain with discharge O/E-minimal discharge with small gap	30/3/2020	C&S- no growth	Continued on Sulfamethoxazole Prednisolone reduced to 5 mg – 1 week
Pain and redness O/E-Redness- more in the outer central quadrant, induration over the scar, but no fever	4/4/2020	Frank pus aspirated. Sent for C&S	Recurrence suspected. Prednisolone raised to 30 mg OD for four days to be tapered to 20 mg OD for two weeks along with Azithral (Azithromycin) 500 mg for the first five days.
O/E-larger area of induration in lateral quadrant with some area of softening. No evidence of fever or <i>s/o</i> inflammation	9/4/2020	C&S - sterile	Aspirated some amount of blood stained fluid. Steroid same dose continued (20 mg)
Symptoms have subsided but large area of induration with small sinus.	15/4/2020	Ultrasonogram - hypochoic liquefied collection in the left breast at 5 o'clock position about 5 cm from nipple, tracking along the subcutaneous tissue towards the skin surface measuring 3.6 × 1 × 1.6 cm (approximate volume 3 ml) suggestive of a liquefied abscess	Prednisolone continued in same dose for 2 weeks as she seems to respond to it.
Pain and redness persisting	28/4/2020		Tried to aspirate granuloma, small collection obtained. Prednisolone 20 mg for two weeks, plan for surgery if needed

The patient was very stressed and anxious, as she was not getting any relief even after excision of lump and prolonged use of steroids. We advised her to taper off the steroids after consulting her previous doctor, but this was ignored and unknown to us, she stopped steroids within two days of starting Ayurvedic treatment. This came to our notice only by the next review but no adverse event was reported by her. The details of internal and external medications prescribed are shown in Table 2.

Classical reference, ingredients, and therapeutic effects of prescribed medicines are given in Supplementary Table 1.

5. Follow-up and outcomes

Periodic follow-up was done with necessary changes in medicine. Timeline of follow-up is depicted in the following table (Table 3).

Ultrasound scan on 18/11/2020 reported nil focal lesions in both breasts and post-operative changes in the left breast. All medicines were stopped by end of November 2020 and as of 31/05/2021, she has had no recurrence.

6. Discussion

The clinical picture showed *aamavastha* and involvement of all three *doshas*, with *sopha* and *dushtavrana*. The initial line of approach was *deepana*, *ama pachana*, *srotoshodhana*, *sophahara* and *kleda hara*. This was achieved with *Punarnavady kashaya*, *Suryaprabhagulika*, *Pushyanuga choorna* along with *Triphala choorna lepa* and *Nalpamara kashaya dhara* externally. *Punarnavady kashaya* [15] was prescribed as it is *deepana*, *pachana*, *raktaprasadana*, and *sopha hara*, in action. The *deepana* effect of drugs increases *jataragni* and *dhatwagni* and being *pachana*, it digests and removes the toxic

metabolites in the tissues. Uninterrupted functioning of *jataragni* and *dhatwagni* is essential for proper *dhatuparinama* that ensures tissue health which is a vital factor in the healing process. *Raktaprasadana* or purification of the vitiated blood is essential to restore the normal functioning of blood and the immune system. *Raktadushti* plays a major role in the pathogenesis of both *vidradhi* and *vrana*. *Suryaprabhagutika* [16] was selected as it has powerful antibiotic and anti-inflammatory activity. It is also *deepana*, *pachana*, *srothosodhana*, and *soolaprasamana* (analgesic). *Srothoshodhana* or cleansing of blockages in circulatory channels in the body ensures free flow of body fluids without any *sanga* or stagnation which may slow down the healing process. *Pushyanuga choorna* [17] was selected because of its *deepana*, *pachana*, *vedana sthapana*, and *rakthadushti hara* properties. *Triphala choorna* [18] is *deepana*, *ropana*, and *twak gata kledasoshana*. Its *kashaya rasa* and *lekhana guna* removes *kleda* and stops oozing from wounds. It has established digestive, immunomodulating, antimicrobial, antioxidant, anti-inflammatory, and anti-neoplastic actions. *Nalpamara kashaya* [19] is effectively used for cleansing *vrana* as it is *kledahara*, *vrana shodhana* and *vrana prasadana* in action.

This was followed by administration of medicines that were more *vrana ropana* and *rakta prasadana* in action like *Mahatiktaka kashaya*, *Sudarshana gutika*, *Vilwadi gutika* and *Avipathy choorna*, that would prevent recurrence of inflammation. *Mahatiktaka kashaya* [20] is *tiktha rasa pradhana*, *sheeta veerya* and *pitta samana* which makes it ideal for *vrana ropana*. It is also *raktha prasadana*, *srotho sodhana*, *sopha hara*, *kleda hara*, and *vrana shodhana ropana* in action. *Vilwadi gutika* [21] is widely used in Kerala as an effective anti-toxic formulation and is *kapha vata samana*, *kleda visoshana*, *deepana*, *pachana*, *lekhana*, and *srotho sodhana*. *Sudarshana gutika* [22] is *amapachana*, *soola hara*, and *raktha dooshya hara*. *Avipathy choorna* [23] is *deepana* and *srotoshodhana*. It is beneficial in all *pitta*

Table 2

List of internal and external medications with dose, adjuvant, and duration.

Sl No	Formulation	Dose, frequency and time	Adjuvant/method of use	Duration
1	<i>Punarnavady kashaya</i>	20 ml of <i>kashaya</i> , twice daily on empty stomach	40 ml of lukewarm water	01/05/2020 to 09/06/2020
2	<i>Suryaprabha gulika</i>	1 tablet twice daily	<i>Punarnavady kashaya</i> .	
3	<i>Pushyanuga churna</i>	5 gms twice daily, before food	100 ml <i>tandulodaka</i> (rice wash) and one teaspoon of honey.	
4	<i>Triphala churna</i>	Twice a day, Topical application	As paste with water	
5	<i>Nalpamara kashaya seka</i>	Topical use (Slow washing)	50 gm powder boiled in 2 L of water and cooled	
6	<i>Avipathichoorna - virechana</i>	15 gm in the morning for one day, 5 g, evening after food, daily from next day of <i>virechana</i>	Lukewarm water	09/06/2020 to 15/7/2020
7	<i>Mahatiktakakashaya</i>	20 ml <i>kashaya</i> , twice daily on empty stomach	40 ml of lukewarm water	
8	<i>Vilwadi gulika</i>	One each twice daily, after food	Ginger juice and honey	
9	<i>Sudarshan gulika</i>	One each twice daily	<i>Mahatiktaka kashaya</i>	
10	<i>Nalpamara kashaya</i> <i>Mahatikhthaka kashaya</i>	<i>Seka</i>	25 gm <i>Nalpamara choorna</i> and 25 gm <i>Mahatiktaka kashaya choorna</i> boiled in 2 L water	
	1-stopped 2,3 and 4 continued 2 - one afternoon for ten days 3, 4 - as before			
11	<i>Gulgguluthikthaka kashaya</i>	20 ml of <i>kashaya</i> , twice daily on empty stomach	40 ml lukewarm water	15/7/2020 to 03/08/2020
	2, 7-stopped 3,4,6,8,9 and 10 continued as before			
12	<i>Gulgulupanchapala choorna</i>	10 gm, twice daily before food	Honey	03/08/2020 to 30/11/2020
13	<i>Mahatikhthaka ghrta</i>	15 gm, twice daily before food		
	3,4,9, 10, 11 stopped 6 continued, 20 gm, morning once in a month 8 continued, 1 daily, after food			

Table 3
Dates of follow-up with changes in signs and symptoms.

Dates	Periodic clinical outcomes
01/05/2020 (First visit)	Pain, redness and pus discharge from sinus at 4–5 o'clock position
09/06/2020 (Follow-up 1)	Pain and redness subsided but slight oozing from the sinus
15/7/2020 (Follow-up 2)	Oozing stopped, mild pain over previously operated site at 7–9 o'clock position
03/08/2020 (Follow-up 3)	Pain had subsided in one week and abscess resolved completely
18/11/2020 (Follow-up 4)	Breasts and nipples normal with scars in the left breast at 7–9 o'clock and 4–5 o'clock positions. No tenderness or discharge from either nipples.

disorders especially *saama pitta*. It is purgative and has antioxidant, anti-inflammatory and anti-ulcer activity.

When recurrence was suspected, *Guggulutikthaka kashaya*, [24] that is especially effective in deep seated abscesses was started. It is *deepana*, *raktha prasadana*, *srothosodhana*, *lekhana*, *chedana*, *kush-tahara*, *vrana ropana*, *sophahara*, *vrana sodhana*, *kleda hara*, and *paakahara*. It is indicated in *naadi vrana*, *vidradi*, *kushta*, etc.

After complete healing was attained, in *niraama avastha* for prevention of recurrence, *Mahatiktaka ghrita* [20] which is *deepana*, *srothosodhana*, *rasayana* with immunomodulatory action and *Guggulu panchapala choorna* [25] which is very effective in prevention of abscess, sinus, and fistula were prescribed for three months. It is *kapha vata hara*, *lekhana*, *srotho sodhana*, *deepana*, *pachana*, *soolahara*, *vrana sodhana*, *ropana*, and *sopha hara*. It is indicated in *naadi vrana*, *kushta*, *bhagandara*, and *gulma*. Along with this, *mridu virechana* by *avipathy choorna* once in a month to prevent vitiation of *pitta* and *rakta* was advised.

7. Conclusion

There is no definitive treatment for IGM till date. The natural history of IGM is complicated with periods of exacerbations causing considerable distress to the patient. The non-healing nature and high recurrence of IGM necessitates prolonged treatment with steroids and antibiotics that have long-lasting adverse effects. There is considerable debate regarding the benefit of these and newer treatment options with lesser adverse effects are being explored. In this context, Ayurveda may be able to offer an effective treatment option with significantly fewer adverse effects than conventional medicines and surgery, not only in the management of abscess and sinus, but also in preventing recurrence.

8. Patient perspective

The patient was very happy with the outcome of treatment. On her first visit to us, she and her husband had expressed fear about the wound not healing and having to undergo surgery once again. They were concerned about the consequences of long-term steroid use. All this had affected their quality of life. After treatment, as the symptoms were relieved and the condition has not recurred, their fears have been allayed.

9. Informed consent

Informed consent was taken from the patient for this study.

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Conflict of interest

None.

Author contributions

Dr. Maya Balakrishnan: Conceptualisation, Methodology, Validation, Investigation, Resources, Writing original draft, review and editing. **Dr Shibila K:** Data curation, Visualisation.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.jaim.2021.06.023>.

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