Original Article JCMCTA 2025 ; 36 (1) : 119-123

Recurrence of Idiopathic Granulomatous Mastitis Following Treatment With Oral Prednisolone

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Abstract

Background: Idiopathic Granulomatous Mastitis (IGM) isn't very uncommon persistent inflammatory disorder of the breast. IGM treatment is still a debate. presently, no universally accepted treatment method is available. The complete remission/resolution and recurrence rates of prednisolone treatment reported in currently available studies varied. In this context, We have conducted this prospective study to evaluate the recurrence rates of prednisolone therapy for the patients with IGM. To investigate the impact of oral Prednisolone on recurrence of IGM.

Materials and methods: This hospital based prospective observational study was conducted at Department of Surgery, Chittagong Medical College Hospital (CMCH) for one year period from January 2019 to December 2019. A total 150 case of IGM with histological diagnosis were selected purposively from Surgery Department of CMCH .Treatment protocol included oral Prednisolone at a dose of 1mg/kg/day for 1 month followed by tapering of the doses over next 1 month. Patients were assessed 2 months and 6 months after treatment completion for reappearance of lump, change in pain and skin conditions from baseline.

Results: Mean of age 29.67 (± 5.67) years and ranges between 20 and 48 years. The maximum not unusual presenting signs and symptoms have been a mass in the breast and ache. Majority of the lump (74%, 111/one hundred fifty) had been positioned within the top and outer quadrant of the breast. Skin ulcer and sinus was present in only 4% and 5.3% cases respectively. Out of 150 cases 47 (31.3%) had recurrence. Size and extent of the breast lump of the recurrent cases were smaller than the initial breast lump. Pain intensity varies in nature during initial and recurrent presentation. There was no axillary lymph node involvement or skin ulcer in the recurrent cases.

Conclusion: Prednisolone therapy was effective in the treatment of IGM as evident by low recurrence rate.

Key words: Breast lump; Idiopathic granulomatous Mastitis; Prednisolone; Recurrence; Skin Ulcer; Sinus.

Introduction

Idiopathic Granulomatous Mastitis (IGM) is a chronic, invasive breast disease with the best treatment options. IGM treats 24% of inflammatory breast diseases.[1] The annual incidence of IGM is reported as 2.4% and 0.37% per 100,000 women.[2,3] This disease usually affects women of childbearing age or women with a history of oral contraceptive use.[4] Environmental stimuli, exposure to inflammation, diabetes, primary or secondary hyperprolactinemia, smoking, alpha 1 antitrypsin deficiency and autoimmunity have also been implicated as etiological factors.[5,6] The differential diagnosis of IGM includes infectious diseases such as bacteria, mycobacteria and fungi, sarcoidosis, actino-mycosis, Wegener granulomatosis, foreign body, concern for fat necrosis, cyst rupture, ductal dilatation, plasma cellular mastitis (Non-granulo). Most autoimmune hypothesis support the autoimmune hypothesis and suggest that the disease responds to steroid therapy, although serologic tests are often negative.[6,7] Although IGM is a benign condition, it clinically and radiologically resembles most breast cancers.[8] Misdiagnosis is likely because IGM has the same radiological features as breast cancer.[9] Specific analysis is most clearly demonstrated by histopathology in IGM.[10,11] There is currently no consensus or guidelines regarding the optimal treatment for patients with IGM. Other treatments include antibiotics, topical or local steroid therapy, anti-inflammatory therapy including Methotrexate

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Submitted on : 23.09.2024
Accepted on : 29.11.2024

(MTX) total excision, mastectomy and local assessment.[3] Although the best treatment has not yet been determined, Steroid therapy has been shown to be effective in reducing granulomatous masses.[12,13] Steroid therapy was first proposed by De Hertogh et al. who investigated the effectiveness of different treatments and concluded that steroids were the best option for treating this disease.[14] Su et al. also found that steroids produced a green color in 87% of patients following this medication protocol without recurrence, while Maffini et al. showed that the disease improved with the administration of necessary antibiotics and oral steroids.[15,16] So, in this context it was rational to design a study to see the recurrence of IGM patients treated with Prednisolone. Study results would be helpful for the clinician to provide an evidence based better treatment for the patients of IGM. Moreover, an appropriate treatment protocol of IGM can be sort out.

Materials and methods

After getting approval from the ethical committee both verbal and written consent were obtained from the participants. Only diagnosed IGM patients were included in this Hospital based prospective observational study. The investigator collected the data.Data recorded in general surgery ward of CMCH.The study was conducted in Department of Surgery of CMCH, among 150 patients with IGM. Only histopathologically proved IGM patients, Who completed treatment with oral Prednisolone were taken as sample. Patients with IGM who treated with Combined Steroid and Methotrexate or treated surgically were excluded. After getting informed written consent, A Patients received a two months course of oral prednisolone. Two months after completion of treatment patients were reevaluated to assess recurrence. The findings of the study are summarized in the Tables and graphs. Follow up protocol consists of clinical, radiological and Histopathological examination of patients. Patients were assessed clinically supervised by consultant with the following parameters for recurrence, 2 months and 6 months after treatment completion -

- Reappearance of Lump
- Pain.
- Skin changes (Ulcer, sinus).

Radiological assessment was done by USG of both breasts with axilla and Histopathological assessment was done by core cut biopsy or incisional biopsy. All the records of the patient including history, clinical examination findings on admission, histopathology report were assessed. Then recurrence of IGM following treatment was recorded. By using case record form, a detailed review of the sample was done for once. Previous documented records of patient's clinical findings, histopathology report and prescribed treatment paper were collected and recorded. Then patients present condition was also be assessed supervised by consultant and was recorded.After data collection, the data were compiled in a Microsoft Office Excel spreadsheet. The data were then entered into SPSS (Statistical Analysis Program) for Windows version 23 software to process and analyze the data. Qualitative data such as skin changes were presented as frequency and percentage using the chi-square test. Quantitative data (Such as breast size) were presented as mean and SD (Standard Deviation) and analyzed by t-test. Statistical significance was defined as $p<0.05$ and the confidence level was set at 95%.

Results

Table I Age distribution of the 150 patients with IGM

Age (Years)		
Age groups	20-30 years	81 (54.0%)
	31-40 years	59 (39.3%)
	40 years and above	10 (6.7%)
Mean (±SD) age		29.67 (± 5.67)
Range		20-48

SD: Standard Deviation.

Table I shows the mean age of these patients was 29.67 (± 5.67) years and their age range was 20-48 years. Majority of the patients were below 30 years of age and only 10 (6.7%) of the patients were 40 years and above.

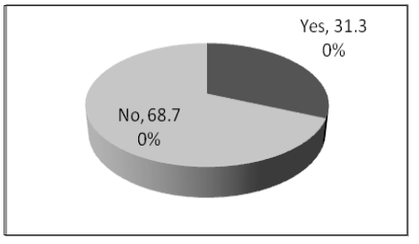

Figure 1 Recurrence of disease among 150 patients with IGM after stopping prednisolone

Figure 1 shows that, out of 150 patients who were treated with prednisolone the incidence of recurrence was 31.3% (47 patients).

Table II Symptom, physical examination and lesion localization of the 47 patients of IGM had recurrence

Variables	Levels	Frequency (Percentage)
Symptoms	Mass and pain	47 (100.0%)
Incomplete resolution		1 (2.1%)
Pain characteristics	Same as baseline	45 (95.7%)
	Less than baseline	1 (2.1%)
	More than baseline	1 (2.1%)
Physical examination	Palpable mass	47 (100.0.0%)
	Palpable axillary LN	0 (0%)
	Skin ulcer	0 (0%)
	Sinus	1 (2.1%)
Lesion localization	Upper outer quadrant	45 (95.7%)
	Upper inner quadrant	1 (2.1%)
	Lower outer quadrant	1 (2.1%)

LN: Lymph Node.

Table II shows that Fourty Seven patients who had recurrence had breast lump and pain. Only one patient had increased pain intensity than baseline and in others pain was same as before or reduced. On physical examination, there was no axillary LN involvement in any patient and in one patient there was skin sinus. Similar to baseline findings most of the recurrent lump was located at upper outer quadrant (95.7%, 45/47).

Discussion

IGM is a chronic benign disease, constituting 24% of all breast inflammatory disease.[17] It needs a prolonged course of treatment and follow-up as it is commonly associated with recurrence. This prospective observational study was conducted to identify rate of recurrence of IGM after treating with oral prednisolone. For this purpose, a cohort of 150 patients of IGM followed up for 2 months and 6 months after a 2 months course of oral prednisolone.

Definitive diagnosis of IGM can easily be made by histopathological examination of the breast lesion. In the present series core biopsy or incisional biopsy was performed and obtained a appropriate diagnosis of GM in all cases. The recurrent cases were also confirmed by the same investigations.

The age ranges from 20 and 48 years. Mean age was 29.67 (± 5.67) years. Young women between 17 and 42 years of age are generally affected with IGM. The mean age observed in the present study was compatible with the available literature.[18,19]

Many studies have summarized that IGM is caused by an autoimmune disease and steroid therapy is a very common treatment. Corticosteroids for the treatment of IGM was first proposed by De Hertogh et al.[14] However, the optimal dose and duration of steroid administration has not been established yet. Previous studies suggested that an initial treatment dose of 30–60 mg/day of prednisone was slowly tapered over several weeks. More recent studies have showed favorable outcomes using short-term, low-dose steroid therapy.[3] Although a wide excision of the mass in IGM was traditionally practiced in the past, this treatment strategy was recognized to have high rates of recurrence, skin ulceration, abscess formation, stulae, and wound infection.[20, 14] An initial dose of 1 mg/kg/day of prednisolone was selected in the present study, based on previous reports.[12,16] It was continued for one month, after which the dose was gradually tapered in the next month. The dosage and length of duration of administration of corticosteroid require further evaluation.

In the available literature the recurrence rate of IGM is reported to be 0.0% to 46.2% even if complete resolution is obtained following corticosteroid therapy.[12,21] In the present study 47 cases of total 150 cases relapsed forming a recurrence rate of 31.3%. The study of Cetin et al. reveals the overall recurrence rate with steroid treatment was 24.7%.[22] This also consistent with our study.

The present study indicated that IGM often located at upper and outer quadrant of breast both in initial and recurrent cases. This finding was also in agreement with the findings of Uysal et al.[23]

Patients with IGM most commonly present with a breast mass which may extend into the skin or the underlying muscle, usually associated with retraction of the nipple and axillary lymphadenopathy.[23] In the present series 96.7% of the patients had both breast lump and pain as the presenting symptoms. On physical examination, the entire group had palpable mass. However, skin ulcer and sinus was present in only 4% and 5.3% of the patients respectively. Axillary lymph mode involvement was found 12% of the patients. Corresponding figure was 7% in the study of Uysal et al.[23]

Another study of Yuan et al. shows administration of corticosteroids for large lesions prior to surgery may help to gain better cosmesis and treatment time needs to be adjusted according to the progression of disease condition.[24]

In the present study, the steroid treatment outcomes were evaluated by palpation and ultrasonography and IGM were followed up for maximum of 6 months post treatment due to time limitation. Based on the literature and personal experiences, recurrences may present several months to years after initial treatment.[3] Further study with a longer follow-up schedule is warranted to determine the persistence of effect of prednisolone in the resolution of IGM.

Limitations

There are few limitations in this study. These are:

- Sample size was small and collected purposively.
- This study was conducted in single tertiary level hospital of Bangladesh. So, the result of the study could not be generalized to entire IGM population of Bangladesh.
- Duration of follow-up was short.

Conclusion

Idiopathic granulomatous mastitis is not a rare disease in our country. Our study shows that prednisolone treatment is effective for IGM patient as recurrence rate is low. Although the duration of prednisolone treatment is long, it has a good healing effect, a good cosmetic effect and a low return rate. Therefore, prednisolone can be the first choice in the treatment of IGM because it is a noninvasive treatment. However, more research is needed with a larger number of patients to create an effective treatment that will reduce the recurrence rate and achieve good results. This study evaluated the recurrence of IGM after prednisolone treatment. The results of the study show that prednisolone treatment is effective in IGM. IGM recurrence was observed in 47 out of 150 patients (31.3%) two months after stopping prednisolone treatment.

Recommendations

In the light of the findings of the present study, the large scale multicenter randomize control study is needed to establish the study findings.

Acknowledgement

The authors would like to express deepest regards and profound gratitude to their honorable teacher Ex-Professor & Head, Department of Surgery, Chittagong Medical College for his kind help, sincere co-operation, valuable advice, active guidance and constructive criticism.

Contribution of authors

SA-Conception, design, acquisition of data, data interpretation, manuscript writing and Final approval.

SA-Analysis, critical revision and final approval.

FM-Conception, design, analysis, data interpretation, drafting and final approval.

MAA-Data interpretation, analysis, critical revision and final approval.

LP- Analysis, data interpretation, critical revision and final approval.

BP-Data analysis, critical revision and final approval.

Disclosure

All the authors declared no conflict of interest.

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