Mammary Fistula: Treatment and Outcome

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Abstract
Background: The aim of the present study was to review the clinicopathological findings associated with mammary fistula and compare the surgical methods used in the treatment of mammary duct fistula in our perspective.

Methods: This is a retrospective study within the period of 2003 to 2015 carried on 52 patients presented with a periareolar skin opening. The study included data of 57 cases (considering five cases of bilateral involvement) consists of nipple inversion, history of fistula formation and drainage of abscess, operative technique, recurrence and histopathological findings.

Mainly two operative technique were followed with Fistulectomy with primary repair were selected for patients < 30 years of age, wants cosmetically better looking scar, No history of previous operation around nipple areola complex other than incision and drainage of abscess. Single fistulous communication.

Fistulectomy with packing were undertaken in all the other patients.

Except the two procedure other procedure according to need was undertaken and included in this study as a part of management of the total 57 fistula handled during this period.

Conclusion: Mammary fistula is not recognised and treated properly in our country. Thus resulted in repeated operations and prolong suffering from the benign condition. As the disease is uncommon, adoption of a definite and uniform policy for management is difficult.

Timed intervention even by fistulectomy reduces the need for total duct excision, or other more extensive surgery which may be the last resort for the young patients who want to breast feed her child.

As cosmesis is not a problem in our patients fistulectomy and packing can be done confidently to get rid of the miserable condition.

Keywords: Mammary Fistula; Nipple Inversion; Fistulectomy

Introduction
An abnormal tract communicating with an infected mammary duct with the periareolar skin is called an Mammary fistula. Subareolar abscess resulting from duct ectasia and periductal mastitis are the major predisposing factor [1].

Recurring subareolar abscess also known as Zuska’s disease, is a bacterial infection of the breast where the abscess burst open through the periareolar skin leading to formation of a fistula from the duct of the affected nipple to the subareolar tissue. The thick creamy dis-
charge passes from the affected nipple resulting from Squamous metaplasia of the lactiferous ducts which subsequently leads to formation of multiple recurrent abscess [2,3].

Though the pathogenesis was described by Zuska [4] but Atkins [5] was proposed and introduced the term mammillary fistula [6,7]. Subareolar abscess is mostly associated with smoking [3,4,8].

These fistulae are different in their aetiology, pathogenesis, treatment and prognosis. Smoking is a major factor for fistula formation [8].

Atkins first suggested that nipple inversion might have a causal relationship but cause and effect are difficult to separate, particularly as nipple retraction is a common sign of the early stages of this disease.

For the management of mammillary fistulae many types of surgery were suggested but due to lack of randomised trial and also as it is comparatively rare it is difficult to conclude which one is better. Among many options that have been mentioned in the literature include fistulotomy as suggested by Atkins. Patey suggested fistulectomy alone for fistula management, Dixon suggested fistulectomy with excision of the involved duct. Hadfield introduced radical duct excision for management [5-7,9].

Aim of the Study

The aim of the present study was to review the clinicopathological findings associated with mammary fistula and compare the surgical methods used in the treatment of mammary duct fistula in our perspective.

Methodology

Methods

This is a retrospective study carried on within the period of 2003 to 2015. The aim of the present study was to review the clinicopathological findings associated with recurrence of mammary fistula and compare the two surgical methods Fistulectomy with primary repair versus Fistulectomy with packing used in the treatment of mammary duct fistula in our perspective.

Study includes 52 patients presented with 57 cases of periareolar skin opening. The study included data consists of nipple inversion, history of fistula formation and drainage of abscess, operative technique, recurrence and histopathological findings.

Mainly two operation technique were followed in 50 cases, with Fistulectomy with primary repair were selected for patients < 30 years of age, wants cosmetically better looking scar, No history of previous operation around nipple areola complex other than incision and drainage of abscess with single fistulous communication in 15 cases. Fistulectomy with packing were undertaken in 35 cases.

Except the two procedure other procedure according to need was undertaken and included in this study as a part of management of the total 57 fistula handled during this period.

Result

Fifty two women presented with mammary duct fistula included in this study. All of them had persistent discharge from the fistula opening. Five (9.6%) of them had bilateral fistulae. Demography of the patients presented in table 1.

<table>
<thead>
<tr>
<th>Study duration</th>
<th>2003 to 2015</th>
</tr>
</thead>
<tbody>
<tr>
<td>Study population</td>
<td>52</td>
</tr>
<tr>
<td>Age, y (mean)</td>
<td>17 - 65 (34)</td>
</tr>
<tr>
<td>Breast involved, right left/bilateral</td>
<td>38%/52%/9.6%</td>
</tr>
<tr>
<td>Inverted nipple</td>
<td>44.2%</td>
</tr>
<tr>
<td>Congenital/recent</td>
<td>28.8%/ 15.4%</td>
</tr>
<tr>
<td>Right/left/both</td>
<td>13.5%/21.2%/9.6%</td>
</tr>
</tbody>
</table>

Table 1: Demography of the patients.
Mammary Fistula: Treatment and Outcome

The mean age of the study group was 34.03 years with age range 17 to 65 years. Most of the patient in the study belonged to age range 25 - 34 years 23 (44.2%) (Table 2).

<table>
<thead>
<tr>
<th>Age in years</th>
<th>No</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>15 - 24</td>
<td>9</td>
<td>17.3</td>
</tr>
<tr>
<td>25 - 34</td>
<td>23</td>
<td>44.2</td>
</tr>
<tr>
<td>35 - 44</td>
<td>8</td>
<td>15.4</td>
</tr>
<tr>
<td>45 - 54</td>
<td>7</td>
<td>13.5</td>
</tr>
<tr>
<td>55 - 64</td>
<td>4</td>
<td>07.7</td>
</tr>
<tr>
<td>&gt; 64</td>
<td>1</td>
<td>01.9</td>
</tr>
</tbody>
</table>

Table 2: Age in years.

Most of the women in our series were married accounting about 90.4%, as there is a trend for early marriage in this country. During the time of presentation only two woman were lactating her child. In 14 cases no history of breast feeding during lactational period from the side of the breast where subsequently fistula was formed. History of diabetes was present in 4 cases.

Patients were suffering from repeated abscess with spontaneous discharge 24 (42.1%) or previous incision and drainage of abscess 18 (31.6%), sometimes several times on the same or opposite breast. Patients apply traditional medicine for abscess rupture in 12 cases, with the use of herbs only in 8 cases and herbs with I/D done by Quack/village doctor in 2 cases. Two cases of breast biopsy were also done after herbal application (Table 3).

<table>
<thead>
<tr>
<th>Formatio</th>
<th>No</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Spontaneous</td>
<td>24</td>
<td>46.2%</td>
</tr>
<tr>
<td>By I/D</td>
<td>16</td>
<td>30.8%</td>
</tr>
<tr>
<td>Breast biopsy</td>
<td>04</td>
<td>07.7%</td>
</tr>
<tr>
<td>Traditional medicine</td>
<td>08</td>
<td>15.4%</td>
</tr>
</tbody>
</table>

Table 3: Events Leading to the Development of Mammary Fistula.

Treatment

Patients presenting with abscess were 36 (63.2%) initially treated conservatively by second generation cephalosporin (cephradine or cefuroxime) and metronidazole for 7 - 10 days. An ultrasound was performed after the course of antibiotics was completed. Eighteen (31%) patients underwent incision and drainage either outside or in our hospital for control of abscess.

To establish mammary fistula, history of discharge from the side of nipple with fistulous opening was identified clinically. In the operation theatre under general anaesthesia fistula and its related ducts were identified by inserting a probe. Every patient received antibiotic.

Most of the cases were operated several times before attending to us. Out of these cases were operated minimum 2 times in 14 cases, 3 times in 7 cases, four and five times in 3 and 2 cases respectively. On one case 5 times operation done 4 times I/D and one time excision and biopsy on the same lady. Lastly it was proved to be tuberculosis. On one patients single time fistula on right side but operated two times on left. Another case two times fistula operated on right but one time on left.

Patients operated single time in 31 cases (54.4%). Recurrence occurred in 26 (45.6%) cases (including incision and drainage of abscess). Primarily I/D of abscess was undertaken in 18 cases and all of them recurred.
Fistulectomy with packing were undertaken in 31 patients and fistulectomy with primary repair in 14 cases (Table 4).

<table>
<thead>
<tr>
<th>Report</th>
<th>No</th>
<th>%</th>
</tr>
</thead>
</table>
| Duct ectasia              | 16 | 30.8%
| Chronic abscess           | 04 | 07.7%
| Nonspecific               | 26 | 50.0%
| Foreign body granuloma    | 02 | 03.8%
| Tuberculosis              | 04 | 07.7%

*Table 4: Histopathology report.*

Most of the fistula after incision and drainage managed by fistulectomy and packing. In one case wall of abscess cavity proved to be tuberculosis on histopathological exam. Only one young female with fistula after incision and drainage was underwent fistulectomy and primary closure according to her wish for cosmetic region without any recurrence.

There was recurrence in four cases after fistulectomy and primary closure. Two cases of recurrence occur after fistulectomy and primary closure managed by fistulectomy and packing subsequently and another two cases by radical duct excision.

Out of three cases of recurrence following excision biopsy, in two cases radical duct excision done. One cases of traditional medicine followed by excision biopsy and another case of radical duct excision after excision biopsy proved to be tuberculosis on final histopathology report after chronic discharge for long time. One case of recurrence after fistulectomy and packing was tuberculosis. Antitubercular chemotherapy were needed in 4 cases (7.7%).

Pathology

Histopathological data were obtained either from the wall of the abscess cavity or from the excised ductal system. Histopathological results revealed periductal mastitis in 18 (31.6%) patients but most of the cases report was nonspecific inflammation 28 (49.1%). Histopathology reported tuberculous mastitis in four (7.0%) patients (Table 5).

<table>
<thead>
<tr>
<th>Recurrence</th>
<th>Yes</th>
<th>No</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Inverted nipple</td>
<td>Yes</td>
<td>15</td>
<td>08</td>
</tr>
<tr>
<td></td>
<td>No</td>
<td>09</td>
<td>20</td>
</tr>
<tr>
<td>Histopathology</td>
<td>Duct ectasia</td>
<td>08</td>
<td>08</td>
</tr>
<tr>
<td></td>
<td>Nonspecific</td>
<td>06</td>
<td>20</td>
</tr>
<tr>
<td></td>
<td>Tuberculosis</td>
<td>04</td>
<td>00</td>
</tr>
<tr>
<td></td>
<td>Chronic abscess</td>
<td>04</td>
<td>00</td>
</tr>
<tr>
<td></td>
<td>Foreign body granuloma</td>
<td>02</td>
<td>00</td>
</tr>
</tbody>
</table>

*Table 5: Relationship of recurrence of fistula with clinicopathological parameters.*

* Chi square test done to detect significance at .05 significance level

Relationship of recurrence of fistula with clinicopathological parameters was presented in table 6, which revealed that recurrence of mammary fistula was significantly associated with nipple inversion (p = 0.006), and the histopathology report (p = .001).

**Table 6:** Choice for Surgical Treatment for established mammary Fistula.

Outcome of treatment in our study group showed that patient fistulectomy and primary closure resulted in recurrence in five cases. In one case there was recurrence due to fistulectomy with packing. So in our series fistulectomy with packing better than Fistulectomy with primary closure in relation to recurrence (p = .027) (Table 7).

<table>
<thead>
<tr>
<th>Treatment</th>
<th>Recurrence</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Fistulectomy and packing</td>
<td>31</td>
<td></td>
</tr>
<tr>
<td>Fistulectomy and primary closure</td>
<td>14</td>
<td></td>
</tr>
<tr>
<td>Excision and Biopsy</td>
<td>05</td>
<td></td>
</tr>
<tr>
<td>Radical duct excision</td>
<td>07</td>
<td></td>
</tr>
</tbody>
</table>

**Table 7:** Outcome of two surgical treatment undertaken for mammary fistula.

*Fisher Exact test done to detect significance at .05 significance level

**Cosmesis**

All Patients are counselled about the distortion of the nipple earlier before performing fistulectomy and packing. Usually the patients belong to elder age group or suffering long time from repeated or recurrent abscess and discharge. Usually they prefer to get relief from suffering more than cosmesis. So they are satisfied with the outcome of operation with some degree of nipple abnormality. Though scoring system were not done formally among the patients.
Recurrence after fistulectomy and primary closure.

Fistulectomy after recurrence.
Mammillary fistula is a relatively uncommon condition. It produce long-term distress for patients and cause prolonged morbidity. Multiple surgery sometimes fails to achieve relief of symptoms.

One of the major pre-disposing factors is duct ectasia and Other specific causes of inflammation of the breast particularly granulomatous inflammation and tuberculosis. But other nonspecific inflammation along with mammary duct ectasia, are seen more commonly [10].

Among the aetiology many factors including mechanical factors, abnormalities of the nipple, local growth of such as tumors, cysts etc. Profuse Secretion and hormonal imbalance may also responsible for fistula formation [10].

Mammary fistula is a highly recurrent condition commonly associated with a nonlactating subareolar abscess and affecting women aged between 20 and 50 years [11]. According to Atkins [6] the lesion was found more in younger and lactating women.

In this study 52 women with mammary fistula were included Most of the patients were pre-menopausal in these studies. The mean age of the study group was 34.03 years with age range 17 to 65 years.

Similar mean ages (30 - 35 years) was reported by some other studies [1,8,12,13]. Higher mean ages (> 35 years) was also recorded [14-16]. Much higher mean ages also presented by Rees BL [17] 72 years, by Demirel AH 43.9 years. Age range varies from 15- 75 years [1,8,12-18].

Initially duct obstruction secondary to nipple inversion was considered as a main factor in pathogenesis of fistula formation [5].

The patients with duct ectasia had a nipple abnormality in the form of retraction or a fish mouth appearance. This indicates that nipple retraction could be a predisposing factor in the development of periductal mastitis and fistula formation [16].

In our study 45.6% patients had nipple retraction, 42% cases nipple retraction was seen by Havandi S., et al [15]. A lower rate 30 - 32% of nipple retraction was also noted [13,16]. A much higher rate (60%) of women had indrawn nipples by NJ Bundred., et al. [1] and (71%) patients by Li S., et al [13].

Discussion

Deformity after fistulectomy.
In the present study, we did not have enough data on the causative microorganisms to make comparison with the other study. The present study has shown that tuberculous mastitis could be a cause of mammary duct fistulae. Four (7.0%) patients had tuberculosis in our study. In the study by Almasat JK [16] one of the patients out of sixteen patients one patient had a fistula resulting from tuberculous mastitis.

Although fistulas can form spontaneously, in two thirds of patients they form after aspiration or an incision and drainage procedure [3,4], it has a remarkable tendency to recur repeatedly [11,19].

In our study Patients presented with repeated abscess and spontaneous discharge in 24 (42.1%) cases They consult and treated surgically for the first time with the presentation of fistula.

In our series there was previous incision and drainage of abscess in 18 (31.6%) patients sometimes several times on the same or opposite breast. On the contrary previous incision drainage of abscess was more frequently found in other study.

As reported by N Beechey-Newman., et al [13] some surgical procedure had been performed in 66% before the patients were seen More than half the previous operations 75% were incision and drainage of an abscess. Only 10 (17%) were treated at the time of first presentation with a fistula [13].

Ten patients out of 35 cases presented with Spontaneous rupture of an inflammatory mass in the study by Hanavadis., et al [15]. Fourteen patients out of thirty five previously had incision and drainage of subareolar breast abscesses before presenting with a fistula.

In our study there was found a trend to apply traditional medicine for abscess rupture. A total 12 cases came after herbal application, with the use of herbs only in 8 cases and herbs with I/D done in 2 cases. Excision was done by the Quack/village doctor after application of herbs in 2 cases, no histopathology were done at that moment in those cases.

Though no report of application of traditional medicine was available but 49 (83%) presented after being treated elsewhere for long term recurrent sepsis [13].

According to Li S., et al [14], involvement of the left breast was noted in 35 (50%), the right breast in 30 (43%), and bilateral breasts in 5 (7%) patients. In our series right, left and bilateral breast involvement were found in 38%/52%/9.6% respectively.

In our study histopathological results revealed periductal mastitis in 18 (31.6%) patients but most of the cases report was nonspecific inflammation 28 (49.1%).

Reviewing the pathology by other studies periductal mastitis were demonstrated in majority of the patients [1,10,13,15,16] suggested that periductal mastitis and duct ectasia as the underlying cause for fistula development.

In the present series histopathology reported tuberculous mastitis in four (7.0%) patients. One patients out of 16 with tubercular mastitis reported by Almasad JK [16] and idiopathic granulomatous mastitis in five patients. Out of the 40 patients two patients had granulomatous Mastitis by NJ Bundred., et al [1].

No specific inflammation was recorded in other study [13-15].

On the other hand Li S [14] identified active infection in 41%, chronic inflammation in 50% cases.

Abrahamson [10] concluded that varying histologic features, included overlapping early and late changes can be found in a single specimen.

Mammary fistula usually started with a small subareolar abscess, which either drains spontaneously or by incision and drainage of abscess. Usually it heals after drainage but recurs again [6].
In the study group recurrence of mammary fistula was significantly associated with nipple inversion (P = .006), formation of fistula (P = .000) and the histopathology report (P = .001).

Mammary duct obstruction resulting from primary nipple inversion was a major predisposing factor in the development of periductal mastitis, which may lead to periareolar abscess and fistula formation or it may be a consequence of the disease itself [1,5,16,17].

On the other hand no predictive factors for recurrence could be identified by the study [13] in terms of prior operations, and presence of nipple inversion.

Mammary fistulas are quite uncommon and the appropriate management is not well established. The acute process usually treated with a course of antibiotics or incision and drainage, but the subareolar abscess will continue to recur unless these ducts are excised. Because the underlying cause of recurrent infections is obstructed lactiferous ducts by keratin plugs [8]. So all the technique for mammary fistula involve excision of the involved duct or radical excision of the duct.

Various options for the management of mammary fistulae have been mentioned in the literature including Atkins [6] proposed saucerizing the fistulous track and allowing the wound to granulate. Patey [7] suggested that the track be excised completely. Dixon [4] used to do fistulectomy with excision of the involved duct only. But Hadfield recommended fistulectomy with total excision of the major ductal system as. The major goal of all the surgical procedures is isolation and resection of the affected ducts and fistulas [3].

The type of excisional technique depends upon the findings at operation. When a simple sinus tract communicates with the nipple, excision of the sinus tract and reconstruction of the nipple is necessary [10].

Most patients in the series underwent excision of the fistulous tract followed by primary closure or packing after fistulectomy.

Similar to our study duct excision involving the affected ducts and saucerization was also suggested by other studies with no recurrences [1,8,12,15,20].

Fistulectomy and saucerization with healing by secondary intention has been shown to achieve long term control in 92% of cases. It proved to be an effective treatment when carried out by surgeons of varying levels of skill and experience [8].

Fistulectomy was suggested as a more appropriate procedure for simple fistula and total duct excision should be reserved for complex fistulas [1,15].

Even in case of retroareolar abscess which usually turn to mammary fistula, successful definitive treatment was excision of the central nipple, including the obstructed ducts [2,14,19].

Some study also differ in their view about the efficacy of the most effective operative technique. Hadfield [9] recommended fistulectomy with total excision of the major ductal system. Also reported by Almasat JK [16] that fistulae were effectively treated by total excision of the ductal system in conjunction with the fistulous tract with no recurrence and minimum complications with cosmetically acceptable.

But Havandi S [15] stated that mammillary fistulas were also caused by total duct excision for duct ectasia. They suggested restriction of total duct excision for severe forms of periductal mastitis with discharge from multiple ducts.

In our series fistulectomy with packing was better than fistulectomy with primary closure in relation to recurrence compared by Fisher’s exact test (P = .027). Study by N J Bundred, et al. [1] compared three treatment groups by Fisher’s exact test. Success rate of packing of the wound under antibiotic cover were significantly better than primary closure alone (P = 0.036 and P = 0.027 respectively).

In our setting we radical duct excision were not preferred because in our women cosmesis was not a major factor. More over during reproductive period radical duct excision was not an acceptable choice as most patients wanted to have child and wanted to breast feed
Mammary Fistula: Treatment and Outcome

their baby. So in our setting this option of treatment was rarely applied. Similar to our study Dixon [6] stated that though in some series a high proportion of patients undergoing total duct excision, the operation is less desirable in younger women, who wish to breastfeed subsequently [6].

All patients in our study were counselled about the distortion of the nipple earlier before performing fistulectomy and packing. Usually the patients belong to elder age group or suffering long time from repeated or recurrent abscess and discharge. Usually they prefer to get relief from suffering more than cosmesis. So they are satisfied with the outcome of operation with some degree of nipple abnormality. Though scoring system were not done formally among these patients.

Similar to our study N Beechey-Newman, et al. [13] stated that two-thirds of the patients were either pleased or satisfied with the final cosmetic result of the surgery, but more than 90% said that it left them with some distortion of the nipple. Fistulectomy and saucerization achieves long-term cure in the majority of patients with mammary duct fistula, but it results in some degree of distortion of the nipple.

For better cosmetic outcome excision of the fistula tract through radial incisions was advised to access both affected major duct and fistula tract which also resulted in low recurrence rate [12]. Another approach was fistulectomy followed by reapproximation of the nipple areola complex [21]. Fistulectomy followed by reconstruction of nipple areola complex was suggested by Hartley MN [22].

Conclusion

Mammary fistula is not recognised and treated properly in our country. Thus resulted in repeated operations and prolong suffering from the benign condition. As the disease is uncommon, adoption of a definite and uniform policy for management is difficult.

Mammary duct fistula should be suspected for all women with peri-areolar breast abscesses. Early recognition of the condition and prompt treatment reduces the prolong suffering and morbidity of the patients. Timed intervention even by fistulectomy reduces the need for total duct excision, or other more extensive surgery which may be used as last resort specially for the young patients who want to breastfeed her child.

As cosmesis is not a problem in our patients fistulectomy and packing can be done confidently in any case specially in elderly and in case of recurrence to get rid of the miserable condition.

Bibliography

Mammary Fistula: Treatment and Outcome


