

Modified Surgical Approach for Treating Zuska's Disease: Complete Resection of the Sinus Tract and Duct

Xiang Gao¹, Shang Ju², Tang-Shun Wang¹, Xue Feng¹, Xi-Meng Zuo¹, Shuang Gao¹, Xiao-Guang Shi¹

¹Department of General Surgery, Dongzhimen Hospital of Beijing University of Chinese Medicine, Beijing, 100700, People's Republic of China;

²Department of Peripheral Vascular Disease, Dongzhimen Hospital of Beijing University of Chinese Medicine, Beijing, 100700, People's Republic of China

Correspondence: Xiao-Guang Shi, Department of General Surgery, Dongzhimen Hospital of Beijing University of Chinese Medicine, No. 5, Haiyuncang Street, Dongcheng District, Beijing, 100700, People's Republic of China, Tel +86 (010)84013135, Fax +86(010)84013212, Email shixiaoguang221@163.com

Background: Mammary duct fistula is an abnormal tract formed between the wall of a breast duct and the adjacent tissue or skin, typically resulting from a rupture of the duct wall. This condition is characterized by inflammation due to the leakage of fluid from the surrounding breast tissue. Infection of the sinus tract can lead to the infection of adjacent lactiferous ducts or recurrence in the same sinus tract, ultimately resulting in abscess formation and a prolonged healing process. In severe cases, this condition may be accompanied by breast deformity and other complications.

Case Presentation: A patient with a mammary duct fistula from the General Surgery Department of Dongzhimen Hospital, affiliated with the Beijing University of Chinese Medicine, underwent a modified surgical procedure. Post-surgery, the patient's nipple inversion was fully corrected with no recurrence observed.

Conclusion: The novel modified surgical procedure demonstrates effectiveness in preventing local recurrence, shortening the clinical course, and addressing pathogenic factors such as nipple inversion. This approach has the potential to improve the cure rate of mammary duct fistulas and is worthy of clinical promotion.

Keywords: cure rate, modified surgical procedure, recurrence rate, shortening the diseased course, Zuska's disease

Background

Mammary duct fistula is a purulent condition occurring in the areolar or periareolar region during the non-lactation period. Predominantly affecting young women, it shows no significant correlation with breastfeeding or childbirth, although some patients may present with nipple inversion. The incidence in males is increasing, attributed to obstruction and bacterial infection resulting from underdeveloped lobules and pore systems, as well as squamous metaplasia of the ductal system.¹ Clinically, patients who experience disease recurrence following multiple incisions, drainage, and curettage, as well as those misdiagnosed with mammary tuberculosis or plasma cell mastitis, often undergo unnecessary extensive resections. Studies indicate that the disease varies in clinical manifestations across different stages, which can be divided into mammary duct ectasia, purulent nipple inflammation, periareolar abscess, and periareolar fistula.²

Effective treatment requires excision of the terminal segment of the ducts within and below the nipple while correcting the nipple deformity.³ Given this context, precise surgical treatment, grounded in a clear understanding of the pathological and anatomical mechanisms, is essential. In this study, we investigated the advantages of a modified surgical procedure for treating Zuska's disease, based on a retrospective analysis, with the aim of reducing the recurrence rate and shorten the disease course. The objective of this study is to provide a reference for improving the prognosis and restoring the normal structure of the nipple and areola.

Case Description

Patient Data

This study followed the Declaration of Helsinki and was approved by the Ethics Committee of Dongzhimen Hospital of Beijing University of Chinese Medicine to publish the case details included the images. Written informed consent was obtained from the patient for publication of the case details included publication of the images.

The patient, a 45-year-old female, was admitted to the hospital on May 5, 2023, due to recurrent redness, swelling, and ulceration in the left areolar area persisting for over one year. The patient first noticed redness, swelling, and pain on the medial side (the side closer to the body's midline) of the left areola without any obvious cause over a year ago. There was no nipple discharge or fever at that time. One week later, the skin ulcerated and pus was drained. The patient underwent ultrasonography at a local hospital, which revealed a 2.2×1.8 cm irregular hypoechoic area at the 9 o'clock position in the left areola, with visible fluid flow under pressure, suggesting inflammatory changes. The patient subsequently underwent incision and drainage, and after dressing changes, the wound gradually healed.

One month prior to her admission, the original inflammatory area became red and swollen, with the scar area ulcerating and discharging yellow pus. After a dressing treatment at a local medical facility, the patient sought care in our department. Her medical history was otherwise unremarkable. The patient was G2P1A1L1 (Gravida 2, Para 1, Abortus 1, and Living 1), having delivered one female child.

Physical examination findings included no abnormalities in the heart, lungs, and abdomen. A specialized examination (Figure 1) revealed symmetrical breasts, with the left nipple being reverted at a 2° angle. A 2×1 cm skin lesion was visible in the upper inner quadrant of the left areola, with a palpable subcutaneous lump measuring 2.5×2.0 cm. A small amount of pale-yellow fluid discharge expressed upon compression, which was accompanied by tenderness. There was no discharge from either nipple and no enlarged lymph nodes were palpable in either axilla.

Auxiliary examination included a routine blood test, which showed white blood cells (WBC) at a level of $9.8 \times 10^9/L$. Mamillary ultrasonography indicated: (1) heterogeneous echo texture of bilateral breast glands; (2) a 2.2×1.1 cm hypoechoic area was detected by ultrasound in the medial skin ulceration of the left areola with unclear boundaries and an irregular shape, containing a few blood flow signals within and around it (Figure 1B); and (3) no abnormal lymph nodes were observed in both axillae.

Surgical Approach and Procedure

- Preoperative photographs were taken to mark the extent of inflammation (Figure 2).
- Local anesthesia was administered in combination with intravenous anesthesia induction. Specifically, 0.5% lidocaine hydrochloride was infiltrated around the lesion, areola, and retromammary space for local anesthesia. This was combined with anesthetic induction using intravenous sedative drugs, which helped to reduce the patient's fear and pain during surgery and mitigated the discomfort of local pain at the incision site post-recovery from general anesthesia (Figure 3A).

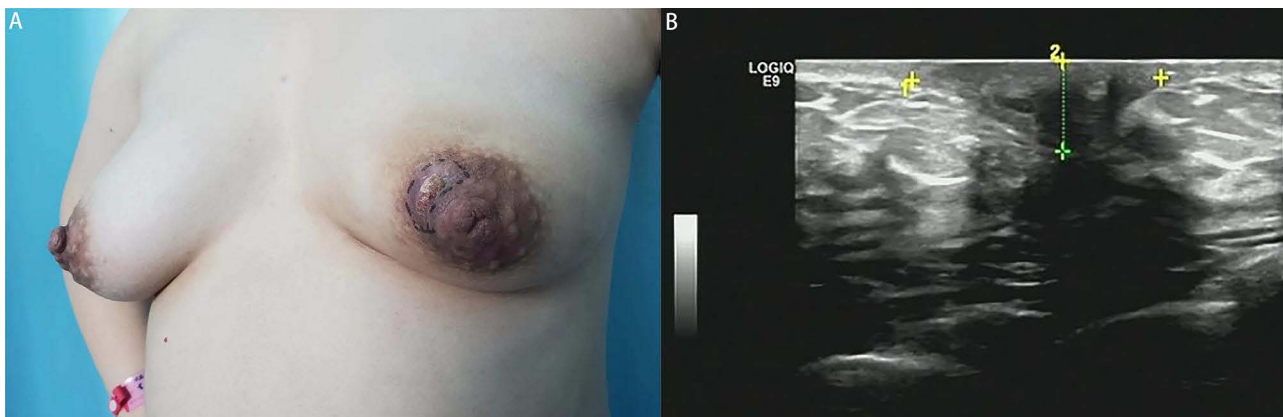


Figure 1 (A) Diagram depicting the affected area. (B) Ultrasound results of the breast.

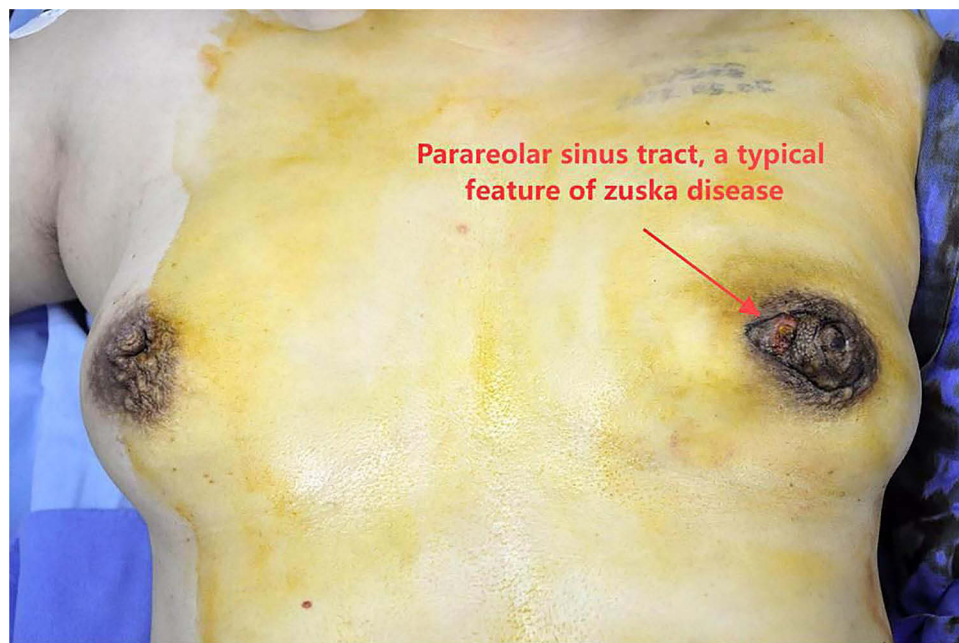


Figure 2 Preoperative photographs marking the extent of inflammation.

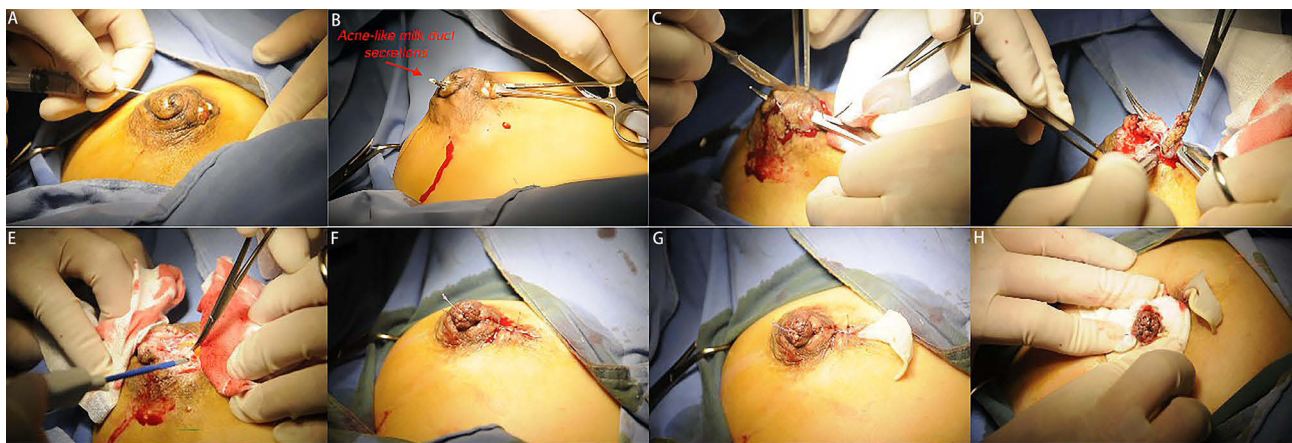


Figure 3 (A) Induction of local anesthesia combined with intravenous anesthesia. (B) Locating the skin sinus and the diseased lactiferous duct. (C) Detection of the duct. (D) Excision of the lesion. (E) Hemostasis and transposition of the glandular flap. (F) Nipple-areola plasty. (G) Wound drainage. (H) Circumferential pressure dressing applied to the areola area.

- c. The sinus tract opening on the skin and the affected lactiferous duct were located using sterile hemostatic forceps, which were inserted into the sinus tract opening external to the duct fistula and gradually advanced into the affected lactiferous duct. Once inside the lactiferous duct, the hemostatic forceps tip was carefully lifted to reach the nipple. Acne-like secretions were observed at the connections between the lesion and the nipple pore, with clear exposure of the hemostatic forceps. These secretions were cleaned using an alcohol cotton ball. The hemostatic forceps was used to lift the nipple and pass through the sinus tract, while preparing to make an incision (Figure 3B).
- d. Investigation into the duct (Figure 3C).

A radial fusiform incision was made centering on the hemostatic forceps that ran through the lesion area. The two ends of the fusiform incision extended to the diseased nipple pore and the skin sinus opening. The incision allowed access to the lactiferous sinus area, where the affected duct, indicated by the hemostatic forceps, was sharply separated. A peri-duct examination was performed to determine the absence or presence of abscesses, necrotic tissue, and previous inflammatory scars.

e. Excision of the lesion (Figure 3D)

The diseased tissue was removed in a 360° circular excision manner, centering on the affected duct and fistula. Notably, the opening of the sinus tract on the skin and a portion of the skin tissue surrounding the nipple were also excised concurrently.

f. Hemostasis and gland flap transposition (Figure 3E)

The surgical area was irrigated with povidone-iodine and hemostasis was performed using electrocautery. The glandular tissue under the areola was freed, transposed, and sutured to the remaining breast sinus area to fill the space and maintain a smooth, non-concave areola region.

g. Nipple-areola plasty (Figure 3F)

The neck of the nipple was first sutured, followed by closing the areola and nipple areas with interrupted sutures, layer by layer. The nipple was reshaped for aesthetical purposes.

h. Wound drainage (Figure 3G)

Since mammary duct fistula is an infectious disease, postoperative drainage is routine. Clinically, strip drainage is primarily used, and for patients experiencing significant exudate from the surgical area, negative pressure drainage may be implemented.

i. Circumferential pressure dressing for the areola (Figure 3H)

Postoperative restoration of blood supply to the areola is crucial. A circumferential pressure dressing was applied to encircle the areola skin with gauze, which exposed and encircled the nipple. This promotes the early establishment of collateral circulation in the capillaries beneath the areola and prevents postoperative nipple reversion.

Prevention and Management of Postoperative Complications

Proper cleaning and disinfection procedures were performed preoperatively to reduce the risk of postoperative infections. The surgical incision was properly cleaned, and dressings were changed as appropriate to keep the area clean and dry, thereby reducing the risk of infection. The patient was closely observed post-surgery to promptly identify and address any signs of infection. Given the diagnosis of a cutaneous sinus tract-type duct fistula, about one week of routine prophylactic antibiotic treatment was administered both during and after surgery.

Prognosis and Follow-Up Measures

Incisions were closely observed to ensure timely treatment of infections if any and to prevent the infection from worsening. Close observation was maintained to prevent excessive bleeding and provide timely treatment when necessary. It is recommended that patients with mammary duct fistula undergo professional examinations every six months. These examinations should include a physical examination of both breasts, ultrasonography, and X-ray examination. Patients with mammary duct fistula are advised to maintain a healthy lifestyle.

Treatment Outcomes

Post-surgery, wound dressings were changed beginning on the second postoperative day and continued until the wound healed and the sutures were removed, which took about 14 to 22 days. The patient was monitored for 6 months postoperatively, during which no recurrence was observed. With respect to nipple inversion, the nipples were completely corrected post-surgery, with no subsequent inversion.

Discussion and Conclusions

Zuska's disease is characterized by purulent discharge from duct fistulas, accompanied by nipple pain, erythema, edema, and other symptoms. Infection-induced sinus tracts can lead to further infection of adjacent lactiferous ducts or recurrence of the same sinus tract, ultimately resulting in abscess formation and a prolonged healing process. In severe cases, this condition can cause breast deformity and other complications. The primary pathological mechanism involves the metaplasia of ductal columnar epithelium into stratified squamous epithelium. Under normal circumstances, the keratinized squamous epithelium of the skin extends concavely 1 to 2 mm into the opening of the lactiferous duct.

However, if the squamous epithelium extends too deeply, it can cause keratin accumulation and blockage of the duct. If the duct ruptures, keratin debris can enter the interstitium, causing a foreign body-induced giant cell inflammatory reaction, followed by bacterial proliferation and infection. Guadagni and Nazzari stated that nipple reversion, recurrent erythema, and painful nodules beneath the areola in non-lactating women are suspicious findings, and the presence of lactiferous drainage sinuses within the areola is a characteristic feature.⁴ The diagnostic challenge lies in distinguishing these benign conditions from breast cancer.

A study concluded that Zuska's disease primarily results from three factors:⁵ (1) Proliferation caused by bacteria retrograding into the input tube. (2) Both Walker⁶ and Khoda⁷ confirmed that coagulase-negative staphylococci and *Peptostreptococcus* are the primary pathogens, with multiple bacterial genera contributing to the pathogenesis. (3) The aforementioned pathological mechanisms lead to the dilation of the lactiferous ducts, exacerbating bacterial infection severity and leading to recurrence.

Previous surgical studies have identified the main causes of recurrent Zuska's disease: (1) Misdiagnosis: Some physicians misclassify this disease as plasma cell mastitis, using needle aspiration for abscess drainage during the suppurative stage, followed by antibiotics or traditional Chinese medicine after relief. (2) Inadequate treatment and follow-up: After incision and drainage of the abscess, routine disinfection, dressing changes, and antibiotic treatment are performed without timely follow-up, causing patients to believe they had developed another breast disease. (3) Diversification of surgical procedures: Most doctors focus on the lesions of the first and second-level ducts at the root of the nipple and perform duct resections, neglecting to correct inverted nipples, resulting in diseased ducts with residual squamous epithelial metaplasia and duct openings, leading to recurrence.⁵ (4) The surgical wound is sutured too tightly without proper drainage, causing inflammatory exudate in the surgical area to not be promptly and effectively drained, thus prolonging the healing process.⁸

Based on existing literature, the current common treatment methods for Zuska's disease are as follows: (1) Single fistulotomy for drainage or local inflammatory tissue clearance: This method results in a recurrence rate up to 70–80% within 6 months.⁹ (2) Fistulectomy: This involves opening the nipple to remove the diseased lactiferous duct connected to the fistula. Simultaneously, thorough curettage is performed to remove inflammatory tissue, and a drainage strip is retained for adequate drainage. The recurrence rate following this surgical procedure is approximately 10–20% within six months.¹⁰ (3) Excision of distal dilated duct: Fistulography is performed before surgery, or methylene blue is injected during surgery to extensively excise the distal ducts of the affected mammary lactiferous ducts. This significantly reduces glandular secretion and resolves the problem of breast secretion accumulation in the sinus ampulla. The recurrence rate is significantly reduced after surgery, with a recurrence rate within six months of about 10% or less.¹¹

Although these surgical procedures aim to remove inflammatory tissue and address the issues of the lesion duct and lactiferous duct, they do not effectively resolve the disease in a short duration or may cause unnecessary removal of surrounding tissue, thereby affecting healing.

Therefore, it is of immense significance to investigate the previous surgical methods for treating Zuska's disease. The following are the three main surgical procedures:

Type A: Transverse nipple incision: The primary issue with Type A procedures is the longitudinal disruption of the nipple's blood supply, which can easily induce postoperative nipple ischemia and necrosis. This is especially true for patients with inverted nipples, leading to recurrent ischemia and even loss of the corrected nipple.

Type B: Areolar incision: This includes a circular areolar incision extending inward and outward in a radial pattern. While the incision is relatively concealed and allows a wide range of exploration, it can only address the lesions of the ducts below the nipple. It cannot remove the first-level ducts remaining in the nipple, posing a risk of recurrence.

Type C: Radial incisions centered on the nipple-areola complex: These incisions do not enter the areola. If sinus tract openings are present, they are excised en bloc in a fusiform manner. Incisions from the Type C procedure have the least impact on the blood supply to the nipple-areola complex. However, for lesions involving multiple ducts, this procedure can easily lead to missed lesions or incomplete removal of subareolar abscesses increasing the likelihood of postoperative complications such as wound infection.

Herein, the surgical approach for Zuska's disease was first described in 2004 where it was used in 26 similar cases,¹² which was fundamentally similar to that of skin sebaceous cyst excision. The core issue in the latter is the complete removal of the sebaceous cyst wall, which effectively prevents future cyst recurrence or infection. Similarly, the surgical strategy for Zuska's disease follows the same principle. Comprehensive excision of the mammary duct infected by the subcutaneous sinus tract (the external fistula opening at the areola) and the opening leading to the nipple are essential for effective prevention of recurrence. This technique is advantageous in effectively avoiding nipple ischemia caused by the Type A technique. In addition, it solves nipple reversion, with small suture tension and easy healing.

However, this technique has a limited detection scope compared to the Type B procedure with respect to other large mammary ducts that have not yet developed lesions. Nevertheless, the excision of the duct opening in the nipple area for patients with Zuska's disease is indeed an important factor in reducing postoperative recurrence. Some scholars suggest that in the mastectomy targeting Zuska's disease, preserving the nipple-areola complex, typically involves retaining 5 mm of tissue posterior to the nipple-areola complex.¹³ However, some ducts may be within this 5 mm range. If these ducts are not cleared, there remains a risk of recurrence. Furthermore, as the Types A and B procedures are highly likely to cause microvascular occlusion among diabetic patients, they are associated with a higher risk of nipple necrosis in this population. The Type C radial incision, on the other hand, is more advantageous for diabetic patients.¹⁴

Alternative treatment modalities for Zuska's disease include the use of stents for recurrent nipple inversion ductal fistula. Chen et al¹⁵ believe that, based on controlling the infection, the key to surgery is to establish a reliable and unobstructed drainage channel to prevent the accumulation of secretions. Therefore, identification of the lactiferous duct in the nipple communicating with the cavity, dilating and retaining the drainage stent, and simultaneously correcting the nipple inversion to establish an effective and unobstructed drainage channel can completely cure the disease. Clinically, 32 patients have been treated successfully with this method, with no recurrences observed after 16 months of follow-up.

Johnson et al¹⁶ suggest that in the treatment of Zuska's disease in men, any male patient with a subareolar abscess should be suspected of having a ductal fistula and treated appropriately. Failure to recognize this important feature may result in unnecessary recurrences, necessitating further intervention. Although there is a limited amount of literature on male lactiferous duct fistulas, complete excision of the lactiferous duct fistula appears to be the most successful treatment. In our practice, we treated two cases through excision of the abscess and lactiferous duct fistula, achieving successful outcomes.

However, the limitations for a case report, and with other diseases, include potential biases such as selective biases by using this patient in their case report and the type of pre- and post-operative care given to this particular patient against conventional methods. Further case series and/or retrospective/prospective studies are needed to demonstrate that this new approach to treating Zuska's disease has a lower recurrence rate compared to conventional methods.

In summary, surgical intervention is currently the preferred method for curing Zuska's disease. Three key points contribute to a low recurrence rate following the surgical procedure at our hospital:

(1) After locating the fistula, splitting the nipple along the fistula allows for complete excision of the lesion without leaving any residual lactiferous ducts in the nipple. (2) To prevent residual squamous epithelium and granulation, it is imperative to excise the range from the proximal duct segment to the opening at the nipple and remove scar tissue and granulation tissue. (3) Draining the wound and applying a pressure dressing is critical. Changing the dressing on the 2nd to 3rd day after compression prevents obstructed drainage and premature closure of both ends of the fistula, which can lead to insufficient treatment and recurrence. Allowing the wound to fill with granulation tissue reduces the likelihood of recurrence.

This method has been clinically validated and constitutes an effective strategy worth promoting for treating recurrent mammary duct fistula.

Disclosure

The authors declare that they have no competing interests.

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