

**Case Report**

# Surgical Management for Zuska's Disease: A Case Report and Review of Literature

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Zuska's disease is a rare chronic breast condition characterized by recurrent nonpuerperal abscesses and periareolar fistulas, often leading to persistent symptoms due to frequent misdiagnosis and inadequate treatment. We present the case of a 52-year-old perimenopausal woman with a history of recurrent breast infections that were mismanaged conservatively. Imaging revealed significant inflammatory changes, which, along with clinical findings, led to a diagnosis of Zuska's disease and prompted surgical intervention. A review of the literature supports our conclusion that surgical excision is the only definitive treatment for this condition. The surgery, involving targeted excision of the affected ducts and fistula, resulted in complete healing with excellent cosmetic and functional outcomes. This case highlights the critical importance of accurate diagnosis and definitive surgical treatment to prevent recurrence and ensure satisfactory cosmetic results.

**Keywords:** Breast Inflammation; Chronic Mastitis; Nonpuerperal Breast Abscess; Periareolar Fistula; Surgical Excision; Zuska's Disease**Introduction**

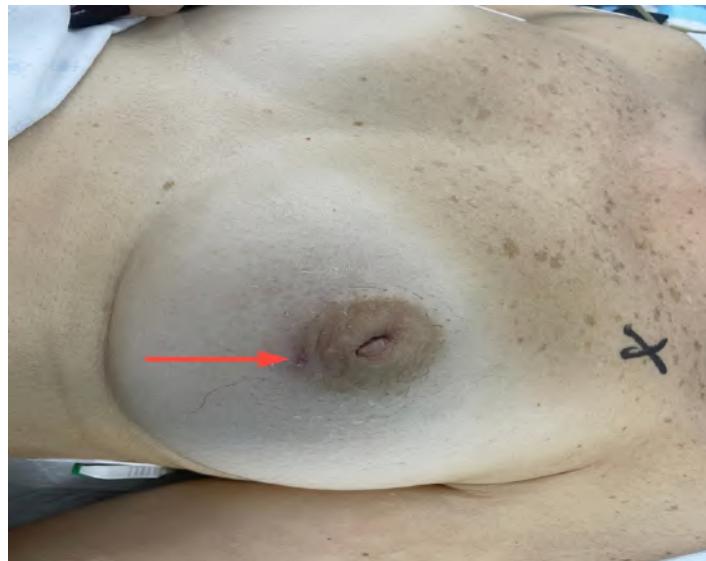
Representing only 1-2% of all symptomatic breast processes, Zuska's disease is a rare chronic condition characterized by recurrent nonpuerperal inflammation and abscess formation in the periareolar region [1,2]. First described by Zuska et al. in 1951, the association between nonpuerperal subareolar abscesses and lactiferous duct fistulas marked a significant advancement in understanding these breast conditions [3]. The pathophysiology of the disease is mainly a combination of ductal obstruction caused by squamous metaplasia and a resulting infection by gram positive or anaerobic bacteria [4]. Risk factors predominantly include perimenopausal age, smoking and hyperprolactinemia to a certain extent [5]. The challenge with diagnosing Zuska's disease lies in its frequent misdiagnosis as simple mastitis and subsequent antibiotic treatment, which, combined with poor patient follow-up, leads to recurrence, whereas definitive treatment requires

surgical intervention [6]. Herein, we present a case of a patient who suffered from undiagnosed Zuska's disease for several years and finally received a proper diagnosis and therapy.

**Case Presentation**

Our patient is a 52-year-old married peri-menopausal female, G0P0, smoker with a 20-pack year history and suffering from mild hyperprolactinemia treated with Dostinex<sup>©</sup> 0.5 mg once per week for the past 30 years. History goes back to 6 years ago when the patient had her first episode of infection and abscess formation in her left breast that was successfully treated with antibiotics. Since then, the patient has had a 2 episode per year recurrence of the infection and inflammation in the same breast that was every time treated conservatively with antibiotics. Her last yearly screening mammogram showed no abnormal masses, calcifications, or other radiographic signs suggestive of breast cancer, and a yearly screening bilateral breast ultrasound confirmed the absence of inflammatory-looking masses that would suggest malignancy. Since the imaging studies did not reveal any suspicious findings that would necessitate further invasive investigation, no biopsy

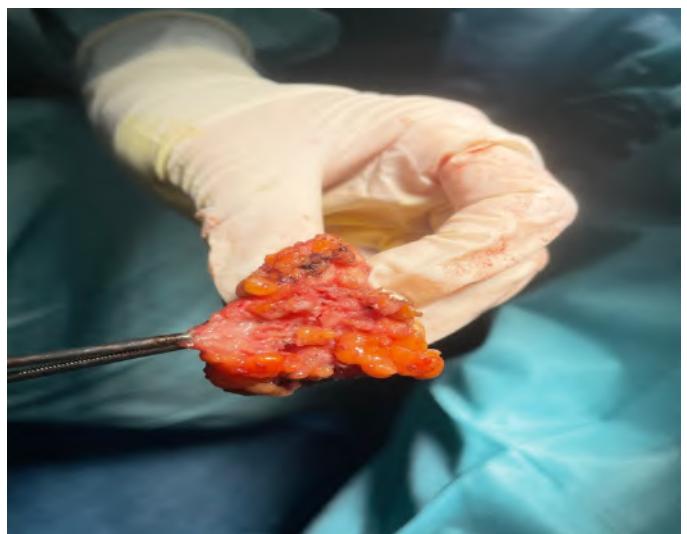
was indicated. Her last infectious episode prior to the current presentation was 7 months ago. Upon presentation to the clinic, the patient was in no acute distress and vitals were stable. She was suffering from a tender diffusely erythematous subareolar lesion towards 4 o'clock in her left breast (Figure 1). She had been taking Augmentin 1 g twice daily for 10 days before presenting for evaluation. A fistula opening can be noticed in the same location with no leakage of pus. A culture swab obtained from the fistula. There was an inversion of the left nipple. The exam of the right breast was unremarkable. No palpable lymph nodes were noted in either axilla. There was a white discharge from both nipples. The ultrasound evaluation of the left breast showed significant inflammation of the areolar skin towards 4 o'clock, the skin at this location diffusely edematous over a zone reaching 3 cm in span and having a maximal thickness of 0.52 cm; the skin does not harbor a clear fluid collection; the inflammatory skin at 4 o'clock is in close proximity with focally dilated lactiferous ducts that are obliterated; a round hyperechoic structure is seen in this location possibly a solidified sebaceous cyst, 0.64 x 0.53 cm in dimensions. There are no abscess collections containing fluid amenable to drainage.



**Figure 1:** Inflammation of the areolar skin towards 4 o'clock in the left breast

There are also no inflammatory looking masses that suggest malignancy. There are no solid inflammatory attenuating masses

typical of malignancy. The findings are highly suggestive of Zuska's disease. As a result, a decision of surgical intervention was made. Under general anesthesia, an attempt was made to inject methylene blue into the fistulous tract using a soft catheter, but it was obstructed. There was no need for guide wire localization of the inflamed breast mass since it was palpable. An incision was performed on the inferior border of areola between 3 et 9 o'clock. Undermining of the nipple-areola complex was done with cutting of the retro-areolar ducts. Skin undermining was performed in the lower pole of the breast. The inflamed palpable zone was then identified and resected in block until reaching the pectoralis fascia (Figure 2). The surgical site was washed using diluted Betadine®. To close the resulting defect, breast remodeling was done using advancement flaps. This targeted approach aims to remove only the affected ducts while preserving as much healthy tissue as possible, thereby minimizing cosmetic impact and preserving breast function. Post operation, the patient was transferred to a regular floor, and her hospital stay was not complicated. Laboratory results done after the operation showed a mildly elevated prolactin level of 47 ng/ml, which is not very significant, taking into consideration the patient's history of hyperprolactinemia. Complete healing was achieved, with a well-preserved breast shape, a favorable cosmetic outcome, and maintained symmetry between both breasts (Figure 3).



**Figure 2:** Excised part of the left breast including the abnormal lactiferous ducts and the fistula.



**Figure 3:** Complete healing of the scar with pleasing cosmetic outcome and maintained symmetry

## Discussion

Zuska's disease, a rare breast condition, accounts for only 1-2% of all symptomatic breast disorders [1]. Due to its rarity, it is often misdiagnosed and consequently mistreated, resulting in increased morbidity among patients [7]. Many physicians opt for conservative antibiotic treatment alone, which frequently

leads to disease recurrence and chronic patient discomfort [4,7]. Our aim is to emphasize that the definitive treatment for Zuska's disease requires surgical intervention to excise the affected tissue completely, thereby preventing further recurrence. Breast abscesses are generally divided into 2 categories: puerperal which are directly linked to lactation and caused usually by normal skin flora, and non-puerperal which are unrelated to lactation and mainly caused by either gram-positives or anaerobes [4]. Zuska's disease is characterized by the recurrence of nonpuerperal breast abscesses along with the presence of draining lactiferous fistulas into the superficial skin [4,8]. The exact etiology of Zuska's disease is not well established, yet the most acceptable origin is squamous metaplasia of the epithelial lining of the breast lactiferous ducts [6]. An obstructed lactiferous duct will create an adequate environment for bacterial growth which will result in chronic active inflammation and abscess formation around the nipple [4,5,9]. As the occlusion will prevent drainage of the duct, fistulous tracts often develop, typically opening at the border of the areola as was seen with our patient. If improperly treated without follow-up, the periareolar fistula will eventually become surrounded by keratinized epithelium, thus inhibiting its spontaneous healing and leading to chronic purulent discharge into the superficial skin [4]. According to the literature (Table 1), first, smoking appears to be the main risk factor present in most cases reported and second, the disease appears normally around the perimenopausal phase.

Author	Age/Sex	Smoker	Clinical history	Acute onset of symptoms	Ultrasound results	Treatment	Follow up
Yalcin et al.	39/F	No	9-year history of recurrent subareolar abscess and duct fistula	-	Multiple cystic masses in inferomedial quadrant of right breast	Drainage and surgical excision	-
Plaxco et al.	50/F	Yes	One episode 20 years ago treated with drainage	-	3.6 × 3.1 × 1.5 cm oval, hypoechoic, hypervascular periareolar mass in the right breast	Surgical excision	Symptoms largely resolved after 11 days
Serrano et al.	55/F	Yes	No history	1-week history of a right breast lump and green nipple discharge	Oval subareolar mass with heterogeneous echotexture	Drainage and antibiotics	Returned 8 months later for abscess persistence

	51/F	Yes	One episode 1.5 years prior to presentation	1-week history of draining right breast abscess	Retroareolar, hypoechoic, irregular, 3 × 1 cm abnormal area overlying mildly dilated ducts	Drainage and antibiotics	Patient kept returning for chronic breast abscess over a 30 months period
	36/F	No	4-year history of left breast pain, a palpable lump, and skin thickening in the areolar region	2-week history of worsening of the symptoms	A large hypoechoic irregular mass beneath the skin with marked skin thickening	Surgical excision	No recurrence on follow up
	46/F	No	History of mastitis after the birth of her first child	Increased nipple sensitivity in her right breast	2 × 1 cm oval mass with circumscribed margins and ductal dilatation	Surgical excision	Asymptomatic on follow up
Abboud et al.	56/F	Former smoker	History of bilateral breast abscesses requiring incision and drainage	10-day history of left breast pain, tenderness, and erythema	-	Drainage and antibiotics	Persistence of symptoms for several months treated each time with antibiotics

To the best of our knowledge, no cases in the literature reports presence of hyperprolactinemia in a patient having Zuska's disease. In our patient, hyperprolactinemia may have aggravated the condition by increasing lipid and protein levels in breast secretions as well as leading to mammary duct ectasia, thereby further promoting inflammation within and surrounding the ducts [10,11]. In a series of studies conducted by Peters et al., increased prolactin secretion was identified as a contributing factor to mammary duct ectasia [12]. Among 108 patients with nonpuerperal mastitis, 11 had asymptomatic hyperprolactinemia, with breast abscesses being the first clinical manifestation of the latter [12]. Therefore, hyperprolactinemia may be regarded as a risk factor for the development of mammary duct ectasia, which can subsequently lead to breast abscess formation. Our primary focus is to explain the significance of accurately diagnosing and adequately treating Zuska's disease. A key factor that can be used to prevent misdiagnosis is the presence of a history of previous breast manifestations. In fact, 6 out of the 7 patients in the literature had a history of breast inflammation, lump, recurrent abscess and/or ductal fistula [4,5,7,13]. Based on the history, presentation and extent of the disease, treatment can vary and include antibiotics

alone or in addition to ultrasound guided incision and drainage or surgical excision [2,9,19]. In the case series of Serrano et al. presenting 4 females with a similar presentation of Zuska's, the 2 patients who were treated with antibiotics and abscess drainage had either one or several abscess recurrence upon follow up, whereas the other 2 who underwent surgical excision were completely healed with no recurrence of symptoms [7]. Similar outcomes were reported in other cases where treatment options included abscess drainage followed by a complete excision of the diseased area [5,13]. In the complex case reported by Abboud et al., the patient consistently received conservative treatment but was lost to follow-up each time and subsequently returned with recurring symptoms [4]. Ultimately, surgical treatment was suggested as a definitive solution, but the patient declined [4]. Our case aligns with the outcomes reported previously, as our patient experienced years of unsuccessful antibiotic treatment before ultimately achieving a complete resolution of her condition through surgical intervention.

In a study performed on 62 patients with non-puerperal breast abscesses that received both oral antibiotics and ultrasound guided

drainage, only 50 had no sign of remaining infections, and the other 12 had to undergo a surgical excision of the abscess cavity in addition to the associated fistula and inflamed tissue [14]. This concludes that abscess drainage combined with antibiotic therapy can only be beneficial in cases of simple uncomplicated breast abscesses, whereas in resistant cases of Zuska's with fistula formation and tissue metaplasia, surgery is necessary for healing [14]. One final important aspect to address concerning the surgical treatment of Zuska's disease is the final aesthetic appearance of the breast. Giacalone et al. shares the details of his technique performed on 27 patients suffering from recurrent subareolar abscesses, and highlights the satisfying functional and cosmetic outcomes [9]. After tissue and fistula excision, glandular tissue and skin remodeling resulting in a breast lift is essential for to achieve symmetry of both breasts and guarantee a pleasing aesthetic look [9]. In our experience and in view of the currently available literature, the combination of surgical intervention and antibiotic therapy offers an optimal balance in terms of efficiency and satisfying results. This report on a 52-year-old female highlights the advantages of this approach, emphasizing the importance of not overlooking the aesthetic aspect of the final result.

## Conclusion

Zuska's disease, characterized by recurrent nonpuerperal abscesses and fistulae in the periareolar region, presents significant diagnostic and therapeutic challenges. This case underscores the critical importance of accurate diagnosis and the necessity for definitive surgical management. Our patient's history of recurrent infections, often misdiagnosed and managed conservatively, illustrates the limitations of abscess drainage coupled by antibiotic therapy. The focused excision of the affected ducts and fistula effectively addresses the underlying ductal obstruction and inflammation, significantly reducing the risk of recurrence. In addition to achieving adequate functional results, surgeons must employ specific surgical techniques to ensure breast symmetry and gratifying cosmetic outcomes.

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