

Zuska's disease: Just a woman's illness

Sofia Estevinho, Gunes Karakus, José Fiel, Madalena Nogueira

ABSTRACT

Introduction: Subareolar breast abscess or fistula is a rare benign condition affecting the breast, especially in man. This disorder constitutes a significant surgical challenge because it is associated with significant morbidity. The patient has discomfort and a tendency for recurrence [1]. Literature on mamillary fistulas in men is scarce, therefore standardization of treatment does not exist although complete surgical excision provides better results. **Case Report:** We report the case of male recurrent subareolar fistula. A 36-year-old male, smoker, was seen in our department because of episodes of purulent drainage from his left periareolar area (two drainage orifices) with discomfort for the last two years. During this time, medical therapy was unsuccessful. We performed a surgical excision consisting of wedge resection, as a whole, of the fistulous tract and orifice. Pathology revealed dilated subareolar ducts lined by squamous metaplastic cells and surrounded by granulomatous inflammation, compatible with Zuska's disease. So far, he is free from recurrence. **Conclusion:** Although a rare disease, Zuska's disease must be considered in differential diagnosis of recurrent breast diseases.

Keywords: Breast disease, Periareolar area, Purulent drainage, Recurrent subareolar abscess or fistula, Zuska's disease.

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INTRODUCTION

Zuska's disease (recurrent subareolar abscess or fistula) is characterized by repetitive inflammatory processes, subareolar abscesses, without apparent cause, typically drains at the margin of the areola. In such cases, squamous metaplasia occurs in one or more lactiferous ducts, in their passage through the nipple lactiferous duct, leading to inflammation, ductal dilatation and obstruction [1]. Zuska et al. were the first to describe this disease in women, in 1952 and is a rare benign condition affecting the breast. These processes usually affect young women of childbearing age, and cigarette smoking is the most important risk factor for development and severity of the metaplasia [2]. Its management can be very frustrating for the treating physician because of its often chronic nature and tendency to recur. The definitive treatment of subareolar breast abscesses, especially when recur, consists of adequate excisional therapy [3].

Literature on mamillary fistula in man is scarce, therefore, its surgical approach is based on the practical experience with female cases.

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The purpose of this case report is to share our experience, because it is a rare disorder (especially in man) and awareness of the condition facilitates correct diagnosis and treatment.

CASE REPORT

A 36-year-old man, smoking 10 cigarettes per day, was seen in our department for lesions in the left breast. He described episodes of purulent drainage from his left periareolar area (two drainage orifices) with discomfort during the previous two years. During this time, he undertook several courses of antibiotics which were unsuccessful. The patient had no further relevant medical history.

Cutaneous examination showed a normal nipple in the left breast with two ulcerated lesions in the areola's external border (Figure 1A–C). Purulent material drained from the periareolar lesion upon lateral pressure. A recent ultrasound revealed an echogenic mass with a central hypoechogenicity and a diameter of 18 mm. We performed a surgical excision consisting of a complete wedge resection of the fistulous tract and orifice (a radial incision was used, which included an ellipse of skin, the entire duct and the fistula tract). The inflammatory area was completely resected until normal tissue was reached. The tissue was closed in layers so that no cavity remained (Figure 2). Histopathological analysis revealed dilated subareolar ducts lined with squamous metaplastic cells and surrounded with granulomatous inflammation compatible with Zuska's disease (Figure 3).

DISCUSSION

Recurrent subareolar breast abscesses occur due to the formation of a keratin plug at the terminal part of a segment of the lactiferous duct. This is facilitated by a process of squamous metaplasia or epidermization, whereby the transition between the columnar epithelium of the lactiferous duct and the squamous stratified epithelium of the skin is displaced to the origin of the lactiferous duct, leading to duct ectasia and a chronic inflammatory process [2]. The obstruction caused by the keratin plug causes dilation and exposure of periductal tissue to keratin debris originated from rupture of the ductal epithelium and leads to a local inflammatory



Figure 1: (A, B, C): Normal nipple in the left breast with two ulcerated lesions in the areola's external border.



Figure 2: Immediate post-surgical appearance.

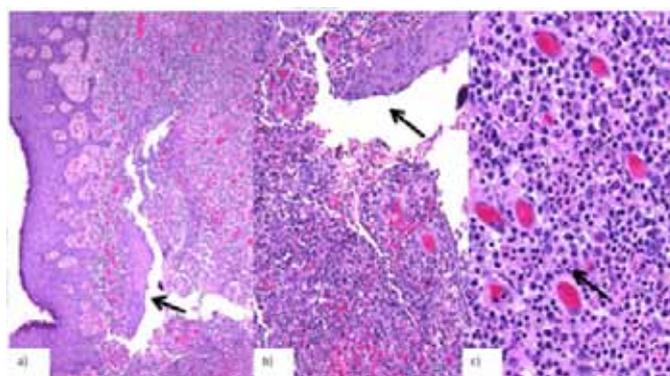


Figure 3: Aspects are compatible with acute inflammatory process, with fistulous tract formation: (A) Marked inflammatory process, with abscess formation, in the dermis (arrow), (B) Higher magnification detail of the fistulous tract, with a squamous lining (arrow), (C) Higher magnification detail of the mixed inflammatory process, with abundant neutrophils (arrow).



Figure 4: Cosmetic result six months after surgery.

reaction which is subsequently contaminated by germs of the normal local flora and results in abscess or fistula formation.

From recent anatomical evidence it is known that the nipple consists of 16–20 orifices of which 6–9 are duct systems and the remaining are openings of sebaceous glands. Thus, the plugged ducts may correspond to the nipple's sebaceous glands, which would explain their cutaneous or subcutaneous location, without reaching the whole depth of the gland (as in a puerperal abscess) and their drainage orifice at the edge of the areola [4].

The occurrence of this pathology in men favors this pathogenic theory. The most important risk factor for development and severity of the metaplasia is cigarette smoking. However, there are other risk factors involved such as age and nipple piercing [1].

In two retrospective studies, one with 68 and another with 89 patients with the diagnosis of breast abscess, smoking was the only factor significantly associated recurrence and multiple surgical interventions [5, 6].

The diagnosis is based on clinical manifestations (purulent drainage) and presentation (recurrence) and ultrasonography. Ultrasound imaging can provide additional information such as the size, depth and anatomical location of the lesion and can help in guided-aspiration of the collection [7]. The definitive diagnosis is made with fine needle aspiration or surgical excision [3] and cytological examination.

Despite being a long known pathology there is some controversy in its pathogenesis and consequently in the most appropriate approach and treatment.

Multiple recurrences of a subareolar abscess in the same location or occurrence of chronic fistula at the areola are frequently a frustrating problem for the patient and the physician [3].

The literature on recurrent subareolar breast abscesses in men is scarce, so its approach is based on the clinical experience described in women. Many studies have shown a high rate of recurrence up to 80% in those treated without excision of the lactiferous ducts compared to those in which all diseased ducts were excised [8].

Often, the initial management in some of these cases is only antibiotic therapy with or without incision and drainage of the abscess.

Several surgical techniques and therapeutic approaches have been proposed by various authors, such as open drainage of the abscess cavity and healing through secondary intention [9]; needle aspiration or incision and drainage together with antibiotics [3]; retroareolar resection with circumareolar approach [10]; two elliptical incisions in a line radially through the diseased portion of the periareolar region where the fistulous tract was present; a radial incision including the skin (with one large elliptical incision) [3]; and more recently, oncoplastic techniques have been applied to treat chronic subareolar abscesses [10] but these patients require many procedures to achieve symmetry.

In our case, we used the Prats' technique [4] which consists in a complete wedge resection of the fistulous tract and orifice to a depth of 1–1.5 cm followed by closure in layers.

CONCLUSION

Although a rare disease, Zuska's disease must be considered in differential diagnosis of recurrent breast diseases. The most important risk factor for the development and severity of metaplasia is cigarette smoking. The definitive therapy for Zuska's breast disease consists in adequate surgical excision. To prevent recurrence and morbidity it is important to perform a complete excision of the fistulous tract including the fistula, all of the retroareolar fibroglandular tissue and any ductal tissue within the nipple.

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Author Contributions

Sofia Estevinho – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Gunes Karakus – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

José Fiel – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

Madalena Nogueira – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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