Double purse-string suture surgical wound closure after excision of nipple adenoma of the breast: a case report.

Kanelina Bimpa [1], Theodosia Charitou[1], Apostolos Ziogas[2], Konstantinos Kantounis[3], Emmanouil Xydias[2], Mattheos Bobos[4], and Elias Tsakos[1]

[1]Embryoclinic, IVF Unit
[2]University of Thessaly School of Health Sciences
[3]Private Clinic
[4]School of Health Sciences, International Hellenic University, Alexandrian Campus

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CASE REPORT

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Kanelina Bimpa[1], Theodosia Charitou[1], Apostolos C. Ziogas[2], Konstantinos Kantounis[3], Emmanouil Xydias[4], Mattheos Bobos[5,6], Elias Tsakos[1]

[1]Department of Breast Surgery, Embryoclinic, IVF Unit, Thessaloniki, Greece

[2]University of Thessaly, School of Health Sciences, Department of Medicine

[3]Department of Dermatology, Private Clinic, Thessaloniki, Greece.

[4]University of Thessaly, School of Health Sciences, Department of Medicine

[5]Department of Biomedical Sciences, School of Health Sciences, International Hellenic University, Alexandrian Campus, Thessaloniki, Greece.

[6] Microdiagnostics Ltd Pathology Lab, Thessaloniki, Greece.

Correspondence to:

Apostolos C. Ziogas, University of Thessaly, School of Health Sciences, Department of Medicine

Rousvelt Street, 24

41222

Larissa, Greece

+30 241 400 8018

ziogasapo@hotmail.com

Declarations of interest: none

Abstract: We present a rare case of nipple adenoma in a 53-year-old Caucasian woman. The lesion presented with a hemorrhagic nipple surface and was treated with wide surgical excision of the nipple and part of the surrounding areola and with a double purse-string surgical closure of the remaining areolar area. This technique was considered safe and effective and aimed to reconstruct the nipple area, thus providing the patient with an acceptable aesthetic result. Double purse-string surgical closure is proposed as a unique and straightforward, oncologically safe surgical procedure. This technique combines complete removal of the nipple adenoma, preservation of the remaining areola, minimization of skin flattening at the reconstructed area, improvement of the long-term aesthetic result and provision of a satisfactory surgical option for the patient.

Keywords: Breast, Nipple, Nipple adenoma, Nipple and Areaola reconstruction, Case report

Key clinical message: Nipple adenoma is a rare, benign pathology that requires adequate excision and satisfactory surgical reconstruction. Our excision and double purse-string suture method covered both requirements, forgoing necessitation of special training of the surgeon or expensive equipment, thus rendering it a simple, safe and effective treatment option.

Introduction

Nipple Adenoma (NA), also referred to as erosive adenoma or florid papillomatosis of the breast [1], is a rare benign breast disease affecting the nipple; it is considered a generally under-recognized condition and it usually affects middle-aged women with an average age of 43-45 years [2]. Exact incidence rate is not yet known due to its rarity, however certain studies estimated that the pathology was present in one out of every 8,000-8,500 skin biopsies or surgical specimens [3], implying a greater incidence in the general population. Male and adolescent patients have been reported, however they are the exception, accounting for under 5% of recorded cases [3].

NA presents clinically with nipple enlargement, nipple discharge (serous or hematic) and the presence of palpable lesion or erosion of the nipple [4, 5], with other clinical manifestations including benign developmental variations, inversion, retraction, or enlargement of the nipple, which may be of either benign or malignant nature, skin changes in and around the nipple, infections with resultant nipple changes or the presence of subareolar mass [6].

Several diagnostic and other examination tools are being used to assess NA, including mammography, breast ultrasonography, galactography, magnetic resonance imaging, cytology examination and core biopsy and histo-pathology examination [3]. Accurate clinical evaluation and management of NA usually requires a multi-disciplinary approach, involving primary care physicians, dermatologists, breast specialists and histopathologists [7]. This thorough diagnostic approach is necessary, as NA may clinically mimic malignant

conditions such as Paget's disease, carcinoma of the breast or nipple eczema [8] and adequate histological assessment is vital in the differentiation of the pseudo invasive pattern that often characterizes NA, a benign tumor, from breast cancer precursors and aggressive carcinoma [9-11].

Treatment of NA is surgical, with various techniques having been described, however no single approach has, yet, been proposed as the gold standard. This report presents the case of NA in the nipple areolar complex (NAC) area treated with the Double Purse-String (DPS) technique, a novel surgical excision and reconstruction method. The following case is presented in accordance with the CARE reporting checklist.

Case Presentation

A Caucasian woman, aged 53, presented to the breast surgery department with a small nodule on her left nipple, self-diagnosed 6 months prior to her visit. The patient reported that the nodule had slightly increased in size, had formed a traumatic surface and that a mildly hemorrhagic discharge was produced. The patient had a negative personal and family history for cancer, no breast cancer risk factors, no other comorbidities or administered medications. She did not recall any trauma on her left breast and reported no other symptoms, such as pruritus of the area. Upon physical examination the nodule was soft, fragile and bled easily. Physical examination, ultrasound scan and mammography of the breasts were negative for any associated pathology. Cytology examination of the nipple discharge was negative for cancerous cells and scanty presence of red cells was reported.

The hypothesis of NA was proposed and the patient was referred to a dermatologist, who performed a punch biopsy in order to confirm the diagnosis. Histopathology examination revealed benign nodular glandular proliferation on the nipple area embedded in a fibrotic stroma. Immunohistochemical evaluation using the p63 / h-caldesmon cocktail, revealed the presence of myo-epithelial cells. Cytokeratin 5/6 identified features of usual ductal hyperplasia, whereas the estrogen receptor expression was low. The diagnosis of nipple adenoma was confirmed with the typical histological and immunohistochemical features. The excision of the lesion confirmed the initial biopsy diagnosis **(Figure 1)**.

The patient underwent surgical excision of the nipple adenoma under local anesthesia. Prior to the procedure, the patient was marked twice pre-operatively, with two concentric circles around the areola region **(Figure 2)**, the distance between the two aiming to be similar to the protrusion of the right nipple. After the complete excision of the nipple, which was completely covered by the adenoma, two purse string sutures were placed at the remaining areola; one at the edge of the incision (inner marked circle), and one at the periphery (outer marked circle), at a suitable distance so as to allow for adequate projection of the areola skin, thus mimicking the contra-lateral nipple. A coated vicryl 910 4.0 thread was used, due to its slow absorption and its being braided, thus more likely to hold its position and maintain the projection for longer. Tightening of the two sutures was applied with caution in order to avoid any tension during healing process. **(Figure 2)**. The patient fully recovered with no complications and was discharged one hour after the procedure. A comprehensive, step-by-step description of the DPS technique has been illustrated in detail **(Figure 3)**.

The patient was examined at 10 days and 4 months post-operatively and underwent follow-up assessment every 6 months. There was minimal flattening of the area but enough projection to mimic a nipple. The patient reported satisfactory aesthetic result and had no intention of further aesthetic interventions. There were no

adverse and/or unanticipated events observed. The patient signed an informed consent form according to the institutional regulations for this publication.

Discussion

Several surgical approaches and reconstruction options post NA excision have been proposed in the available literature. Such techniques are purse-string for nipple adenoma, nipple elevation and nipple-areola reconstruction [7, 12-14], in which NAC creation is the last step in the breast reconstruction process and is vital as it greatly affects patients psychologically. The main challenge is to maintain the projection of the reconstructed nipple over time when the original was removed. Local graft reconstruction combined with tattooing is the most popular technique, with individualized treatment options accounting for patient anatomical characteristics and aesthetic preferences, with results unfortunately not always proving satisfactory. To avoid this challenging reconstruction step, other, more conservative methods have also been proposed. Lee et al[15] utilized Moh's Micrographic Surgery (MMS) and proposed that, if applied early, it could facilitate the excision of the tumor with the preservation of the nipple. Similarly, Bae et al [16] performed cryosurgery in their patient, a technique that is gaining ground as a minimally invasive treatment of NA as well, with remarkable outcomes as far as aesthetics are concerned.

In this report, we presented a Double Purse-String (DPS) technique, which allows for the complete surgical excision of the nipple while preserving the surrounding relatively large areola using a DPS suture on the wound. Subsequently the suture can be used to reshape the areola, create a skin projection (reconstructed nipple), adjust the size, avoid tension, prevent flattening or dissatisfying defects and offer adequate projection; thus adapting to the morphology of the contralateral healthy nipple and preserving breast symmetry. Additionally, patients maintain sensation in the reconstructed area, which promotes an overall feeling of post-operative satisfaction and offers psychological benefits, with complementary options, such as 3D tattooing of the areola following recovery being available as well.

To our knowledge this is the first case of the DPS technique being used in the treatment of NA. The main strength of our proposed method is that a symmetric nipple-areola complex was created that matched the contralateral nipple in size, color, position and projection, giving it a pleasing and natural appearance. It achieved a sustained projection of the areola without complications, such as irregularities and hypertrophic scar. Another advantage is that the DPS technique can be performed in cases of larger or even aggressive NA as well, since surgical reconstruction only requires a small remnant of the surrounding areola, thus facilitating the removal of larger tumors. In contrast, other, less invasive techniques can only be applied in benign and localized lesions, since no reconstruction is performed. Such techniques also require specialized training and expertise on the part of the surgeon in order to be effectively performed, whereas our proposed DPS method is significantly simpler and requires minimal additional training. This advantage of DPS is given even greater emphasis when the rarity of NA is considered, as more complex techniques require more cases to overcome the learning curve and attain expertise. The less invasive techniques also carry the admittedly low, but plausible risk of recurrence due to incomplete resection, a phenomenon observed by Perzin et al [17], who noted 7% recurrence rate in cases treated via local excision, as well as of malignancy arising from NA, a rare but documented occurrence in eight cases [3].

There are certain minor concerns regarding the DPS technique and its practical application that should be addressed. One such concern is the use of the particular suture thread, as it has been reported to increase propensity to infection and local tissue reaction. Since this is a single case, no other sutures have been tested

for more favorable results, however, it is our belief that with proper management pre-, intra- and post-operatively, the possibility of future nipple flattening can be minimized, as we observed in the reported case. Another is the fact that for an aesthetically satisfactory result, adequate residual areola area is necessary to achieve sufficient projection and an overall appearance similar to the contra-lateral nipple, thus it would be more sensible to utilize the DPS technique in cases with adequate residual NAC size post-excision for optimal aesthetic results. Additionally, breastfeeding mothers might face difficulties due to the architectural distortion of the nipple and underlying ducts area, however no evidence-based conclusions can be reported yet, as our patient was post-menopausal. Besides, most cases of NA arise in 43 to 45-year-old, women, who have very likely already completed childbearing.

In our point of view, treatment of NA by the DPS technique is an effective therapeutic intervention, not only due to the assured complete resection of the tumor, but also because of the aesthetically pleasing result that can be achieved; a result comparable to the more advanced, less invasive surgical techniques, though without the higher risk for post-operational residual tumor that these techniques confer.

Consent

Written informed consent was obtained from the patient for publication of this Case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Footnote:

Reporting Checklist: The authors have completed the CARE reporting checklist.

Conflicts of Interest

The authors have no conflicts of interest to declare.

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Ethical Statement

The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

Author contributions:

1) Kanelina Bimpa: conception and design, administrative support, provision of study materials or patients, collection and assembly of data, data analysis and interpretation, manuscript writing, final approval of manuscript

2) Theodosia Charitou: conception and design, provision of study materials or patients, collection and assembly of data, manuscript writing, final approval of manuscript

3) Apostolos C. Ziogas: collection and assembly of data, data analysis and interpretation, manuscript writing, final approval of manuscript, manuscript submission

4) Konstantinos Kantounis: conception and design, administrative support, provision of study materials or patients, collection and assembly of data, final approval of manuscript

5) Emmanouil Xydias: collection and assembly of data, data analysis and interpretation, manuscript writing, final approval of manuscript

6) Mattheos Bobos: conception and design, provision of study materials or patients, collection and assembly of data, manuscript writing, final approval of manuscript

7) Elias Tsakos: administrative support, provision of study materials or patients, collection and assembly of data, data analysis and interpretation, final approval of manuscript

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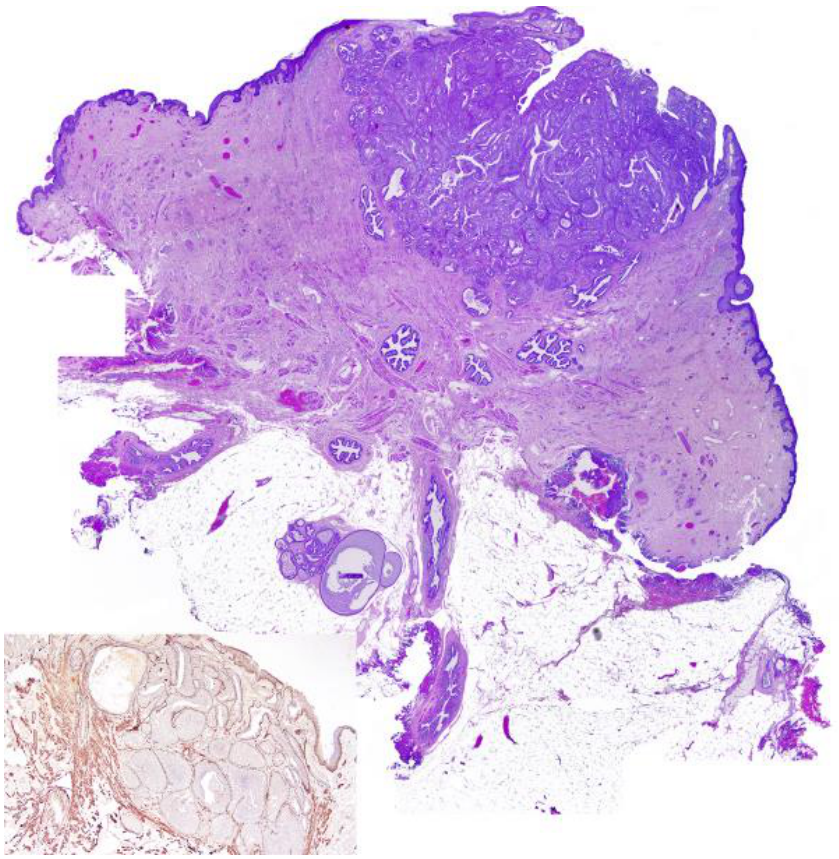

Figure 1: Large panel. Whole scanned microphotograph from the resection specimen stained with hematoxylin & eosin showed benign proliferative epithelial lesion on the nipple area with nodular architecture, consisted of papillary, solid and cribriform structures. Small panel. Immunohistochemistry using the cocktail p63 / h-Caldesmon document the presence of myoepithelial component of the lesion, whereas smooth muscle bundles on the stroma are highlighted with the h-Caldesmon marker. Papillary carcinomas lack myoepithelial cells.

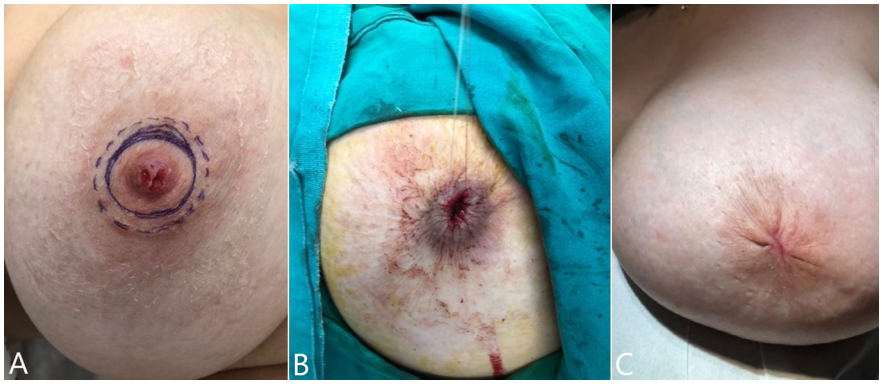

Figure 2: Complete resection of the nipple before and after double purse-string closure. A: Preoperative photograph of patient's breast showing nipple adenoma and marks. B: Application of DPS after complete removal of the left nipple. C: Follow-up check, 4 months after the DPS procedure.

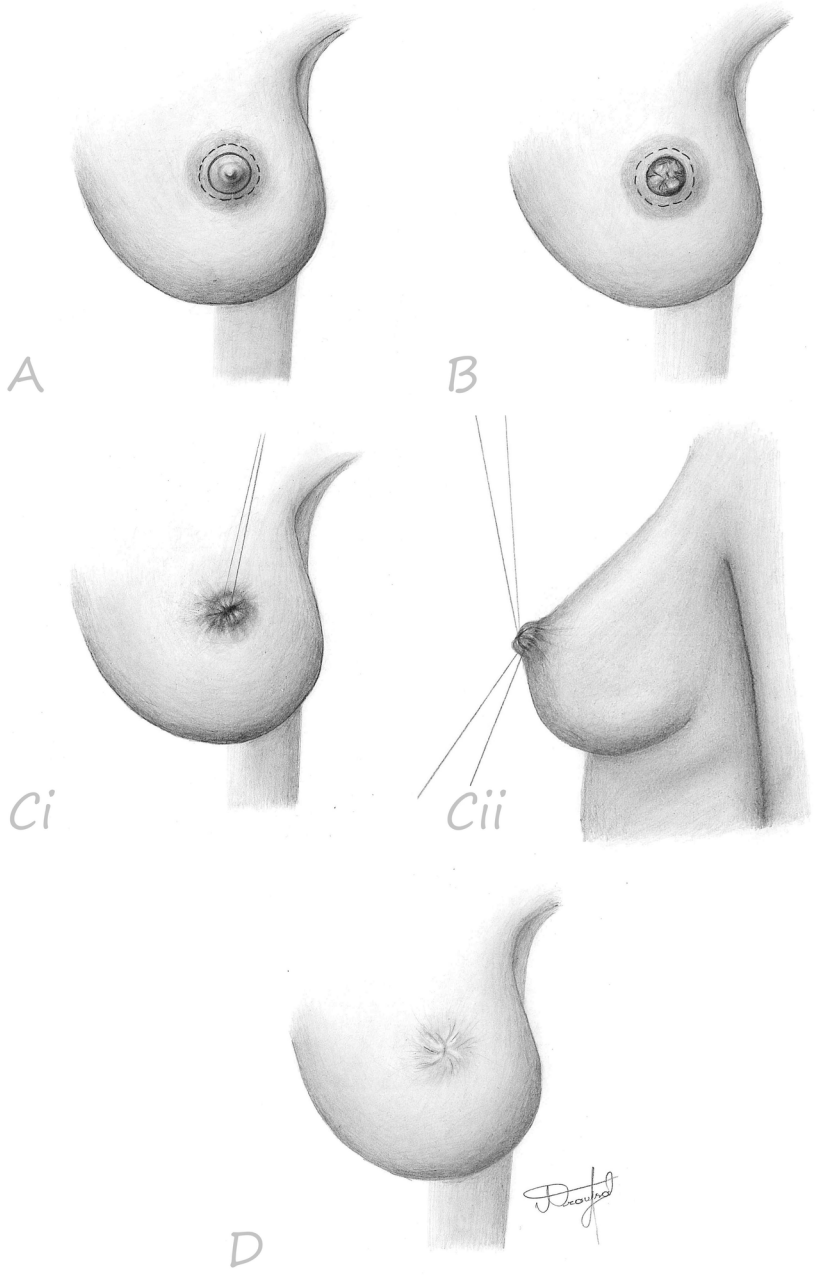

Figure 3: A step-by-step illustration of the DPS technique. A: Marking of the area to be excised (inner, continuous circle), this margin will close with the upper suture of the DPS. Marking of a second area at a distance equal to the desired projection of the new nipple (outer, broken circle), this will form the basis of the new nipple and will close with the lower/second suture. B: Surgical excision of the marked area, containing the adenoma and adequate surrounding tissues. Ci (anterior view) and Cii (side view): Closure of the wound. Inner circle is closed completely by a purse-string suture and forms the top of the nipple. The outer circle is closed by a second purse-string suture lower than the first one. The distance between the two creates a projection resembling the normal, contra-lateral nipple. D: Final reconstructed nipple at follow-up. Despite some minimal flattening, there was adequate projection, along with the radial appearance of the area to maintain an aesthetically pleasing result.