

## CASE REPORT

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# Management of Nipple Loss Caused by Excision of a Nipple Adenoma: A Reconstructive Case

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**ABSTRACT** Nipple adenomas are rare benign tumors that present as a palpable nodule, often accompanied by serous or bloody discharge, ulceration, hyperkeratosis, nipple deformities, and/or erosion. The standard treatment for nipple adenoma is total surgical excision; however, this procedure may result in the loss of the nipple. Although nipple reconstruction is an option, there is a paucity of literature describing cases wherein nipple reconstruction was performed after the total excision of the nipple for adenoma. We describe herein a rare case of a 29-year-old woman who underwent total nipple excision and subsequent nipple reconstruction with a C-V flap. After a year of follow-up, the patient was still satisfied with the cosmetic outcomes of her surgical reconstruction, which we have summarized along with our follow-up strategies.

**Keywords:** Erosive adenomatosis of the nipple; mammoplasty; follow-up studies

Nipple adenomas are rare benign tumors, described as a proliferation of lactiferous ducts within the nipple, which present as a palpable nodule in the nipple, accompanied by serous or bloody discharge, ulceration, hyperkeratosis, nipple deformities, and/or erosion.<sup>1-5</sup> Symptoms are typically present for several months before treatment, as the rarity of nipple adenomas often results in diagnostic difficulties and treatment delays.<sup>3</sup>

These benign lesions, due to aggressive clinical findings such as bloody discharge and ulceration, may mimic Paget's disease of the breast and even squamous cell carcinoma of the nipple. Therefore, cytopathological examination is recommended before treatment to avoid aggressive excision.<sup>2,5</sup> Total excision is the standard treatment for nipple adenoma; however, it may result in nipple loss, depending on the extent of tumor.<sup>1-5</sup> Tumor recurrence is rare once a tumor has been completely excised.<sup>6</sup>

Available case reports primarily describe partial or total nipple excision without subsequent nipple reconstruction.<sup>1-5</sup>

Herein, we describe a patient who underwent total nipple excision and subsequent nipple reconstruction in the treatment of nipple adenoma, along with our follow-up algorithm, which is supported by data from the literature.

## CASE REPORT

A 29-year-old woman with no family history of breast cancer was referred to us by the oncological surgery department, with a 1.5-year history of an erosive nodule that completely covered the nipple and was exacerbated by bloody discharge (Figure 1). Ultrasonography and contrast-enhanced breast magnetic resonance imaging showed a 2.5-cm nodular, non-

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FIGURE 1: Full coverage erosive nodule on the right nipple.



FIGURE 2: One-year post-nipple reconstruction with a C-V flap from the existing areola.

contrasting mass in the right nipple. Cytopathological testing confirmed nipple adenoma. Surgery was planned, and the lesion was completely excised under local anesthesia and sent for pathological testing. Following a report of clean surgical margins, the patient's nipple was reconstructed under local anesthesia using a C-V flap. She healed without complications and was satisfied with the results. She has been monitored for 1 year postoperatively and no recurrence has been observed (Figure 2).

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

## DISCUSSION

Nipple adenomas are rare benign tumors, accounting for approximately 0.45% of breast pathologies; therefore, it is difficult to gather data regarding their clin-

icopathological characteristics.<sup>6</sup> Although the World Health Organization defines nipple adenomas as benign, there are cases wherein breast cancer was simultaneously detected or developed during the follow-up period.<sup>1</sup> Owing to the limited data available on nipple adenoma, its potential associations with breast cancer remain unknown.

Although nipple adenomas most commonly present in middle aged women, isolated cases have been reported in adolescent and even children.<sup>7,8</sup> Despite aggressive findings such as bloody discharge and ulceration, months often pass between symptom onset and diagnosis.<sup>1,5,6</sup> The present patient had the lesion for 1.5 years, and although she initially consulted the dermatology, general surgery, and oncological surgery departments, no one recognized the lesion as a nipple adenoma.

Total excision is the standard treatment for nipple adenomas. Mosh micrographic surgery, nipple splitting enucleation of the nipple adenoma and cryotherapy are alternate treatment options and should be considered in young women to preserve breastfeeding.<sup>1</sup>

In cases where adenoma completely infiltrate the nipple and total excision leads to nipple loss, there is no clear consensus on the parameters for reconstruction. In most of the relevant literature, nipple reconstruction was either not performed or not recommended.<sup>1-5</sup> Bimpa et al. described a single case which involved reconstruction performed using the double string suture technique; however, the desired nipple height was not achieved and the patient declined further surgery.<sup>9</sup>

In the case presented herein, we reconstructed the nipple using a C-V flap. Given that recurrence rates are minimal in patients with negative surgical margins, there are no contraindications for reconstruction. Additionally, nipple loss negatively effects self-worth in women of all age groups. To ensure the best outcomes, therefore, we recommend nipple reconstruction in all patients once negative surgical margins are confirmed.

Local flaps, such as the C-V flap, which have the advantages of not damaging the contralateral nipple and being autologous and technically simple to per-

form, are one of the most commonly used nipple reconstruction techniques. However, a common disadvantage is that the post-operative nipple height decreases over time. Planning these flaps using the original areolar tissue, rather than a skin flap, as in total nipple-areolar complex reconstruction, contributes to better long-term retention of the nipple projection. In the present case, the patient was completely satisfied with the cosmetic appearance of the nipple reconstructed with a C-V flap using the original areolar tissue 1 year after surgery.

While an algorithm for determining follow-up frequency has yet to be defined, two main factors have been highlighted in relevant literature: (1) complete excision of the lesion, and (2) risk factors for breast cancer. Although Suster et al. suggested that no follow-up is required for completely excised lesions, breast cancer guidelines indicate that it is necessary for patients with risk factors for breast cancer or concomitant lesions.<sup>6,10</sup> Considering the age, absence of risk factors for breast cancer, and negative surgical margins in the patient presented herein, self-examination was recommended without subsequent imaging.

In conclusion, nipple adenomas are rare benign lesions for which the standard treatment is complete tumor excision. Subsequent nipple reconstruction can

be planned once negative surgical margins are confirmed, and the frequency of follow-up is based on patient-specific factors.

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### Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

### Authorship Contributions

**Idea/Concept:** Bilgen Can, Ali Duran; **Design:** Bilgen Can; **Control/Supervision:** Bilgen Can, Ali Duran; **Data Collection and/or Processing:** Bilgen Can, Ali Duran; **Analysis and/or Interpretation:** Bilgen Can; **Literature Review:** Bilgen Can, Ali Duran; **Writing the Article:** Bilgen Can, Ali Duran; **Critical Review:** Bilgen Can; **References and Findings:** Bilgen Can, Ali Duran; **Materials:** Bilgen Can.

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