A Novel Surgical Technique to Correct Intrareolar Polythelia

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ABSTRACT
Polythelia is a rare congenital malformation that occurs in 1-2% of the population. Intra-areolar polythelia is the presence of one or more supernumerary nipples located within the areola. This is extremely rare. This article presents 3 cases of intra-areolar polythelia treated at our Department. These cases did not present other associated malformation. Surgical correction was performed for psychological and cosmetic reasons using advancement flaps. The aesthetic and functional results were satisfactory.

Key words: Polythelia, breast deformities, supernumerary nipples

Introduction
Accessory nipples are relatively common in clinical medicine. Furthermore, there is a great incidence of misdiagnoses. The supernumerary nipples are located anywhere from the armpit to the inguinal fold, along the original milk line. Clinically, the patient can either be asymptomatic or have symptoms like discomfort during menstruation, pain, or discharge.

In 1915, Kajava classified the expression of accessory breast tissue in eight categories that is still being used:

TYPE 1: Complete supernumerary nipple - Nipple and areola and glandular breast tissue.
TYPE 2: Supernumerary nipple - Nipple and glandular tissue (no areola).
TYPE 3: Supernumerary nipple - Areola and glandular tissue (no nipple).
TYPE 4: Aberrant glandular tissue only.
TYPE 5: Supernumerary nipple - Nipple and areola and pseudomamma (fat tissue that replaces the glandular tissue).
TYPE 6: Supernumerary nipple - Nipple only (the most common supernumerary nipple).
TYPE 7: Supernumerary nipple - Areola only (polythelia areolaris).
TYPE 8: Patch of hair only (polythelia pilosa).

It must be differentiated from intra-areolar polythelia that is an extremely rare congenital malformation in which one or more supernumerary nipples are located within the areola.

Intrauterine malformation causing a dichotomy of the nipple is believed to be the main etiologic issue. Association with congenital urogenital abnormalities was described. Literature shows only a few cases of intra-areolar polythelia.

The 3 cases reported of dichotomized nipple were not associated with any other breast malformation. The surgical correction methods and the results are herein presented.

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Case Report

The first case was a 46-year-old woman, who had an intra-areolar polythelia of the right nipple since birth (Figure 1). Her menarche occurred at the age of 12 years, and had a normal breast development during puberty. She did not have any other supernumerary nipples. The right areola had a diameter of 43.2 mm and the left areola was 40.2 mm. The right areola had nipple duplication, near the principal nipple. There were no pathologic secretions or other abnormalities related to the breast. No lactiferous ductal system was detected in the supernumerary nipples by echography. She desired surgical correction of her condition mainly for both aesthetic and psychological reasons.

The second patient was a 16-year-old man who had an intraareolar polythelia of the left nipple and a supernumerary nipple in the right thoracic region, along the milk line (Figure 2). There was no association with other malformation or syndromes. No pathologic secretion was observed. Local anomalies of the breast and the two lactiferous ductal systems were displayed by echography. The right areola had a diameter of 37.5 mm and the left was 38 mm. The intraareolar polythelia was located infero-medially, at 1.7 mm distance from the principal nipple. He desired a surgical correction for both aesthetic and psychological reasons.

The third case was a 18-year-old woman who had a vertical duplication of left nipple since birth (Figure 3). Her menarche occurred at 13 years and she had a normal breast development. She did not have any other supernumerary nipple or any other congenital malformations. No pathologic secretion was found. The echographic investigation revealed two lactiferous ductal systems in the left nipple. The two left nipples were well developed, but each was only half size of the contra-lateral nipple. The diameter of the right areola was 42.5 mm and the diameter of the left was 43 mm. She wanted a surgical correction for aesthetic and psychological reasons.

All surgical procedures were performed under local anaesthesia, in the Outpatient Surgery Unit, after hematologic tests and echography of the breast.

In the first case, we treated the patient with elevation of a simple flap that was advanced, shaped and sutured in the correct position in the center of the nipple (Figure 1). The preoperative echographic investigation did not show any lactiferous ductal system within the supernumerary nipples, therefore we were able to use this simple technique without damaging the lactiferous ducts.

In the second case of a male patient, we used the same technique considering the patient’s gender and the simplicity of the technique (Figure 2).

We cauterized the lactiferous ducts that were detected by echographic investigation, in the flap. The flap was then advanced, shaped and sutured in the correct position.

In this way, we were able to reduce scarring and optimize the aesthetic result.

In the third case we had a duplication of the left nipple and two lactiferous ductal systems were seen by echographic investigation (Figure 3).

In this case, we reconstructed the nipple by using Z-plasty transposition flaps, described by MG Onesti et al. (1) for the correction of bilateral intra-areolar polythelia.

We drew two opposite flaps, one including the principal portion of the supernumerary nipple, the flap in the middle of areola and the other one including the portion of areola between principal and accessory nipple. The flap that included the nipple was elevated and transposed to the center of the areola, and sutured to the principal nipple, with preservation of lactiferous ducts. The flap between the two nipples was transposed infero-laterally to rebuild the normal shape of the areola.
Follow up visits were performed at 6 months and 1 year after surgery. The anatomic result of surgical correction was satisfactory concerning the shape, projection and sensation of nipples in all three cases.

**Discussion and Conclusion**

Accessory breast tissue is a relatively common situation that has a high incidence of being misdiagnosed (2). It may indicate underlying congenital anomalies (3).

The dichotomy of nipple is an extremely rare congenital malformation and is often not recognizable. It must be differentiated from supernumerary nipples or polythelia. In 1915, Kajava classified the expression of accessory breast tissue in eight categories that is still being used today. Intraareolar polythelia is classified in the group of supernumerary nipple with only one nipple. Only 10 cases have been presented in literature.

Arranz Lopez et al. (4) described in 2006 a technique for reconstruction of unilateral dichotomy of the nipple. They described a case of dichotomy that affected not only the nipple, but also the areola, like in case number 3. The reconstruction of areola-nipple complex was performed by a Z plasty transposition flap. Two opposed flaps, that included the principal portion of nipples, were elevated and transposed in the center of the areola. The purpose of this technique is to minimize any tissue damage in order to avoid injury to the lactiferous ducts that are of paramount importance for breast-feeding in fertile patients.

The surgical technique described by MG Onesti in 2010 (1), is based on two opposing flaps, one including the main portion of the supernumerary nipple and the other including the portion of areola between the nipples. The flap involving the nipple is positioned in the center of the areola and sutured to the principal nipple.

In the presented cases, we used a simple technique based on advancement flaps. This technique can only be used in male or female patients who do not have lactiferous ductal system within the supernumerary nipples.

For clinical management, an ecography during preoperative period is advisable and should be repeated after surgery. In fact, the goal of this procedure is to avoid damage to lactiferous duct, for preservation of nursing.

Patients afflicted by breast abnormalities may experience depression and a poor self-esteem and this may influence their social relationship. Hence, surgical treatment is advisable for functional, psychological and aesthetic reasons.

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**Informed Consent:** Written informed consent was obtained from patients who participated in this study.

**Peer-review:** Externally peer-reviewed.


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**References**