

VIRGINAL HYPERTROPHY*

Case Report

*Mustafa Küçükaydın MD***, *Selim Kurtoğlu MD****, *Hamit Okur MD*****
*Tahir E. Patıroğlu MD******, *Muzaffer Zorlu MD******
*Zübeyde Gündüz MD******, *Ahmet Kazez MD******

SUMMARY: Küçükaydın M, Kurtoğlu S, Okur H, Patıroğlu TE, Zorlu M, Gündüz Z, Kazez A. (Departments of Pediatric Surgery, Pediatrics and Pathology, Erciyes University Faculty of Medicine, Kayseri, Turkey). Virginal Hypertrophy: a case report. Turk J Pediatr 1994; 36: 243-248.

A 13-year-old girl with virginal hypertrophy (bilateral extensive juvenile hypertrophy) of the breasts is presented. Her breasts began to grow rapidly after puberty and reached an enormous size within a year. On examination, both breasts were greatly enlarged. Routine blood chemistry and the endocrinological investigations were normal. The computerized tomography scan of the sella was unremarkable. A bilateral reduction mammoplasty was performed, and histological analysis of the breast tissue revealed the diagnosis of virginal hypertrophy. After four months her breasts began to grow again, and a second mammoplasty was performed. After this operation, tamoxifen citrate was given to prevent recurrence for four months, and during the follow-up period of 20 months, no recurrence was noted. *Key words:* virginal hypertrophy, mammoplasty, tamoxifen.

Virginal hypertrophy (infantile breast hyperplasia, juvenile hypertrophy) is an uncommon condition of massive breast hypertrophy. Histologically, it is characterised by an enormous increase in the fibrous connective tissue of the breast, and there is only moderate ductal proliferation¹. Virginal hypertrophy is not associated with hormonal abnormalities or remote tumors and is not a premalignant lesion. Reduction mammoplasty is technically difficult because of the large size of the breast and may be followed by recurrence. In severe cases, subcutaneous mastectomy, free nipple transplant and insertion of prosthesis has been suggested^{2,3}. A review of the literature revealed only a few cases in which girls required a breast reduction plasty at age thirteen. For this reason we want to present this case and stress the importance of tamoxifen to prevent recurrence.

* From the Departments of Pediatric Surgery, Pediatrics and Pathology, Erciyes University Faculty of Medicine, Kayseri.

** Associate Professor of Pediatric Surgery, Erciyes University Faculty of Medicine.

*** Professor of Pediatrics, Erciyes University Faculty of Medicine.

**** Research Assistant In Pediatric Surgery, Erciyes University Faculty of Medicine.

***** Professor of Pathology, Erciyes University Faculty of Medicine.

***** Research Assistant In Pediatrics, Erciyes University Faculty of Medicine.

Case Report

A 13-year-old girl was admitted to the Pediatric Surgery Department because of extensive bilateral juvenile hypertrophy of the breasts. The patient's menarche was at age 12 with irregular periods. Her breasts began to grow rapidly after puberty and continued to grow. The patient complained of increased back pain when sitting for a long time. Socially the child was completely integrated into the family but suffered greatly because of her physical appearance and had become depressed. She was the second child in the family and had a brother and two sisters. There was no evidence of drug or medication use.

On examination, the girl weighed 46 kg (25th percentile) and was 155 cm tall (25th percentile). Her breasts were greatly enlarged and extended to the umbilicus (Fig. 1). Capraro's technique was used when measuring the breasts. The right breast was 40x32 cm and 1280 Capraro units; the left breast was 38x31 cm and 1178 Capraro units. (Breast measurements in a normal 13 year-old-girl according to Capraro's original technique are 15x11 cm and 165 units on each side). The diameter of the areola was 9 cm on the right and 8 cm on the left side. The skin was very taut and slightly red with many prominent superficial veins. Neither galactorrhea nor a mass was found on physical examination⁴. Pubescent development was stage V according to Tanner scales. On general physical examination, there were no abnormal findings except for kyphosis.

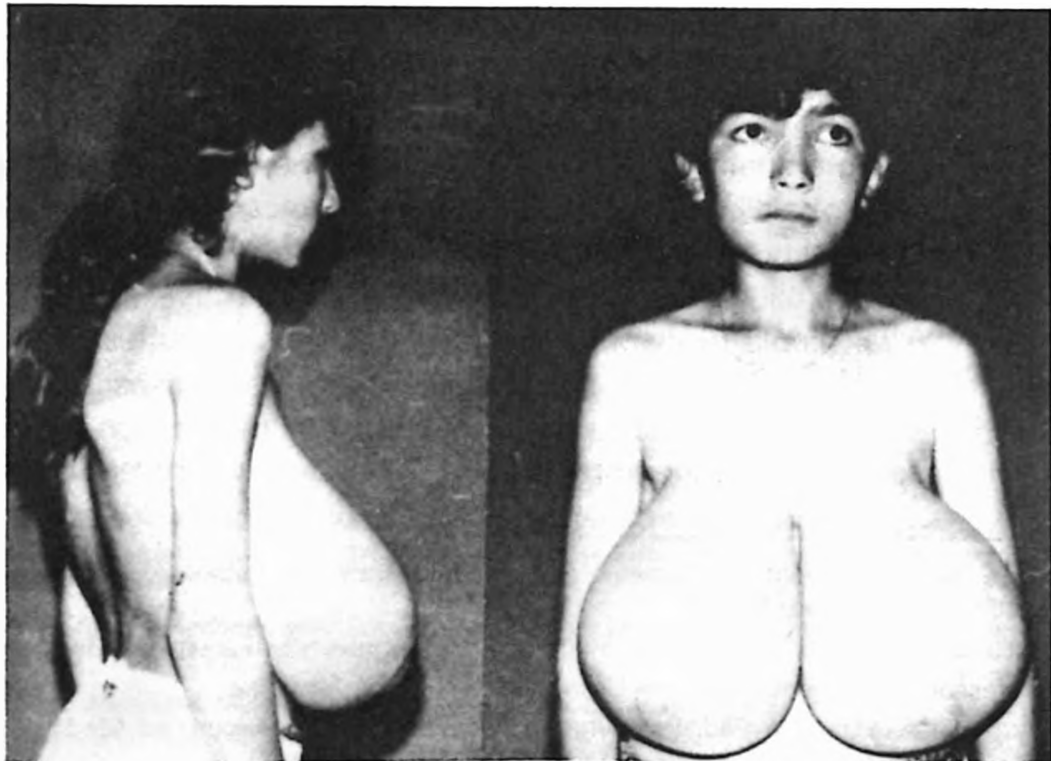


Fig. 1: 13-year-old girl. Both breasts were greatly enlarged.

Routine blood chemistry, complete blood count and urinalysis were normal. The endocrinological investigations, including baseline and thyrotropin releasing hormone-stimulated prolactin levels, baseline luteinizing hormone, follicle stimulating hormone and luteinizing hormone releasing hormone stimulated levels, were normal. Estrogen and progesterone levels were also normal. The computerized tomography scan of the sella was unremarkable. Lateral chest x-ray showed considerable kyphosis of the vertebral column which could have been caused by the weight of the breasts.

A bilateral reduction mammoplasty was performed as described by Blomquist⁵, resulting in the removal of 2400 g tissue from the left breast and 2600 g from the right. Macroscopically the tissue consisted of mainly lobular parenchyma with little fat.

Histological analysis revealed a breast parenchyma with little fat infiltration. Embedded were numerous lactiferous ductules with verruciform proliferating epithelia without atypia. In a few places, the periductal stroma was myxoid (Fig. 2). These features confirmed the diagnosis of virginal hypertrophy. The patient had an uneventful recovery and was discharged from the hospital on the 24th day.

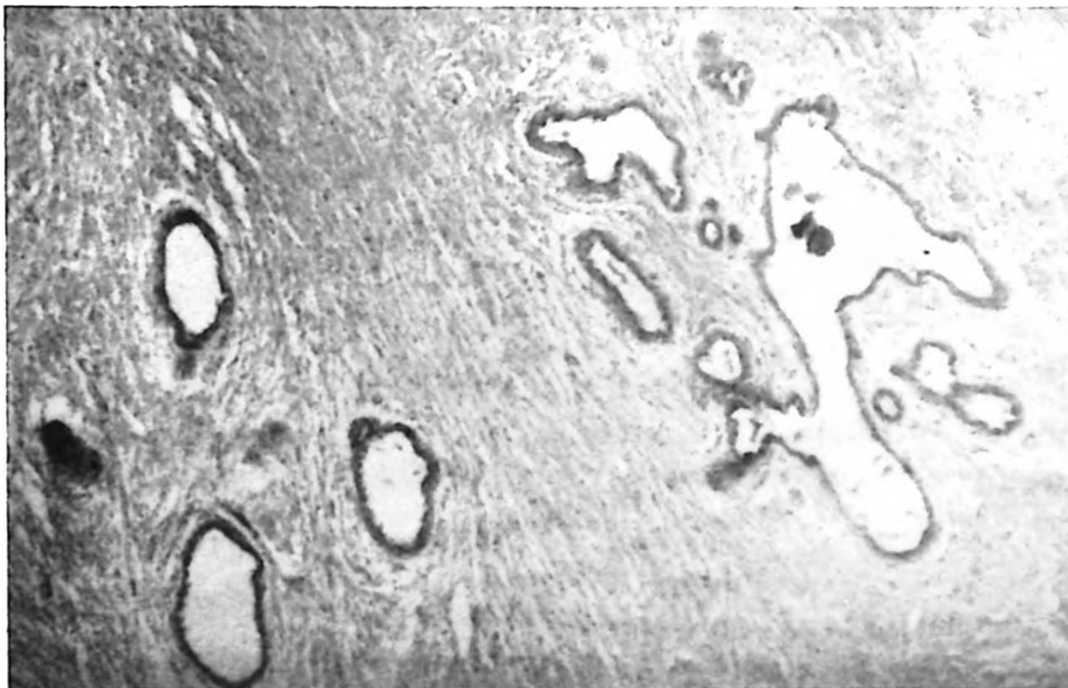


Fig. 2: Histology of excised breast tissue showing a parenchyma with numerous lactiferous ductules without atypia (H and E x 63).

Four months after the operation, her breasts started to grow again. After seven months they had enlarged significantly, and a second reduction mammoplasty was done. A total of 950 g tissue from the right and 850 g tissue from the left was resected. At that time, tamoxifen citrate 20 mg/day was given for four months.

Twenty months after the second operation, there had been no further breast growth (Fig. 3).

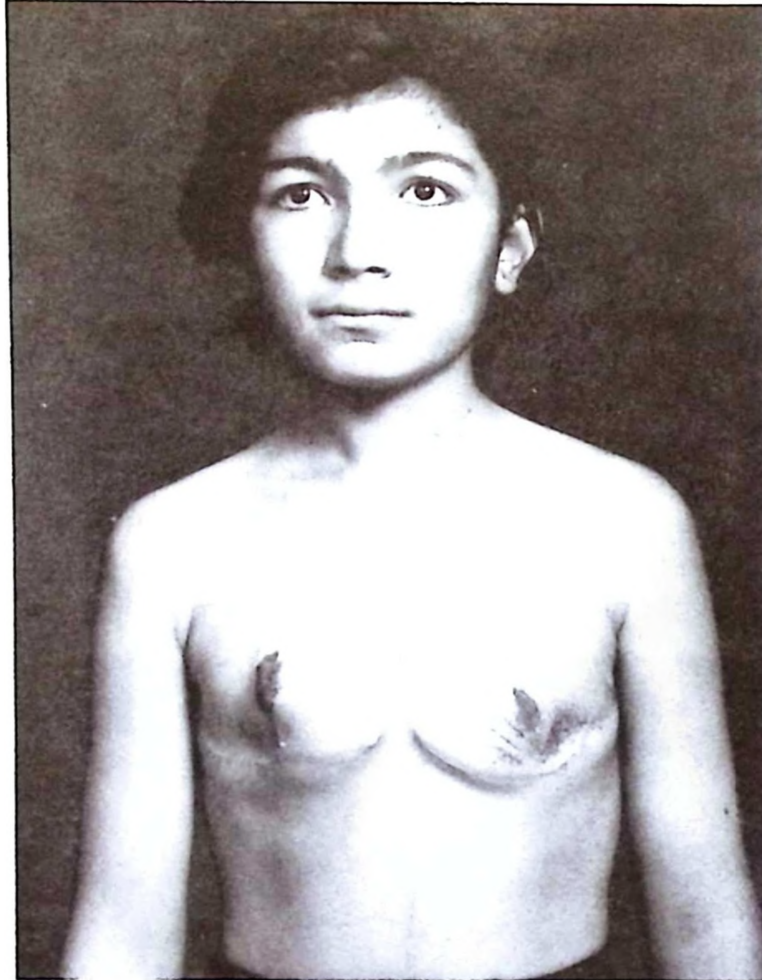


Fig. 3: Result 20 months after surgery.

Discussion

Virginal hypertrophy of the breast is a disease of unknown etiology. It is characterized by massive growth of both breasts, usually at the time of puberty. The breasts may become very pendulous and heavy. Even moderate hypertrophy can cause emotional symptoms similar to those in patients with hypoplasia. The breasts occasionally become gigantic, either at puberty or, more often, in later life in association with pregnancy. A plastic reduction in breast size is indicated whenever the breasts are large enough to cause real distress to the patient⁴.

In 1974 Mayl⁶ described an 11-year-old girl with virginal hypertrophy who initially underwent a reduction mammoplasty but had to undergo a complete mastectomy because of recurrence at twelve years of age. Fisher and Smith⁷ reported a ten-year-old girl who needed surgery for virginal hypertrophy, and in this case, a

postoperative recurrence occurred as well. In 1977, Gardoso de Castro⁸ published a case of a 12-year-old girl who underwent a breast reduction. In this case, repeated recurrence occurred and subsequently subdermal mastectomy was unavoidable. In 1991, Piza-Katzer and Umbricht-Sprüngli⁹ presented an 11-year-old girl who required surgery for infantile breast hyperplasia. Postoperative recurrence did not occur.

The decision of surgery ultimately depends on the degree of suffering and the individual symptoms of the patient. Frequent complaints are: heaviness of breasts, grooves and scars on the shoulders from the pressure of bra straps, shoulder pain, back pain and deformities which may even cause kyphosis, breast tension, orthopnea, restriction of mobility, dermatoses, and headaches which often are augmented premenstrually¹⁰. Nearly all of the complaints mentioned above, especially kyphoses and shoulder pain, were noted in our case. In addition to somatic complaints, psychosocial symptoms were present, including poor social relations with friends, nervousness, and feelings of inferiority and insecurity. When the symptoms are so advanced, surgery is usually unavoidable in spite of the risk of recurrence.

To prevent recurrence, medications are often advised. The administration of dihydrogesterone or tamoxifen citrate has been used with some success¹¹. Bilateral recurrence occurred in our patient, and after the second mammoplasty tamoxifen citrate was administered to prevent recurrence.

In less acute cases, one should not be operated on until the breast is completely developed. The initial examination should involve mammagraphy. Carcinomas are rarely found in this age-group. Cystosarcoma phylloides is found more often, and can only be verified histologically¹².

It is currently being discussed whether hormonal imbalances as well as hypersensitivity of estrogen receptors to estrogen may be the cause of infantile hypertrophy of the breasts. A frequent argument against extensive reduction plasty is that such a procedure would inhibit proper functioning of the breasts during lactation. It must be mentioned that hypertrophic breasts rarely function during lactation^{3,11,13}.

This report shows the importance of the use of tamoxifen citrate to prevent recurrence after surgical reduction of the breasts in virginal hypertrophy cases.

Acknowledgment

I am grateful to Mr. D.M. Burge, Consultant and Pediatric and Neonatal Surgeon, Wessex Regional Centre for Paediatric Surgery, U.K, for helping me in the detailed revision of the paper.

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