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## CASE REPORT

## Massive bilateral breast reduction in an 11-year-old girl: 24% ablation of body weight

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### KEYWORDS

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Hypertrophic recurrence;  
Free nipple grafting

**Summary** An 11-year-old girl with massive virginal breast hypertrophy is presented. The breasts had begun to grow rapidly at puberty and had reached an enormous size within a year, to the point of causing physical impairment and respiratory compromise. Routine blood chemistry and endocrine investigation was normal, as was an MRI scan of the pituitary fossa. A bilateral reduction mammoplasty with free nipple grafts was performed, removing 12.5 kg of tissue in all (24% of the total body weight).

There was no recurrence at a 2 year follow up, and no requirement for additional surgery. A review of the literature reveals that breast regrowth is less frequent when free nipple grafting is used, and this technique is recommended for these extraordinary cases.

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Virginal or juvenile hypertrophy comprises rapid and excessive breast development out of proportion to the growth of the child.<sup>1–3</sup>

The most remarkable example of breast enlargement, described by Durston in 1669, was that of a 23-year-old woman whose breasts enlarged 'overnight' to a combined weight of 104 pounds (47.2 kg), and who died immediately after attempted mastectomy.<sup>4,5</sup>

No abnormalities of oestrogen levels have been demonstrated in these patients, and breast enlargement is attributed to unusual end organ sensitivity to normal hormone levels.<sup>6–8</sup>

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**Figures 1, 2** Preoperative views of the massive 'virginal hypertrophy' of the breasts in an 11-year-old girl.

Histological section reveals stromal hyperplasia, cystic degeneration of the mammary ducts and interstitial oedema.<sup>6</sup> There is an abundance of connective tissue surrounding and separating the ducts. The mammary ducts

manifest minimal branching, so that lobule formation is, at best, abortive.

### Case report

A girl with a 6-month history of massive breast hypertrophy was initially seen 2 weeks after her 11th birthday.

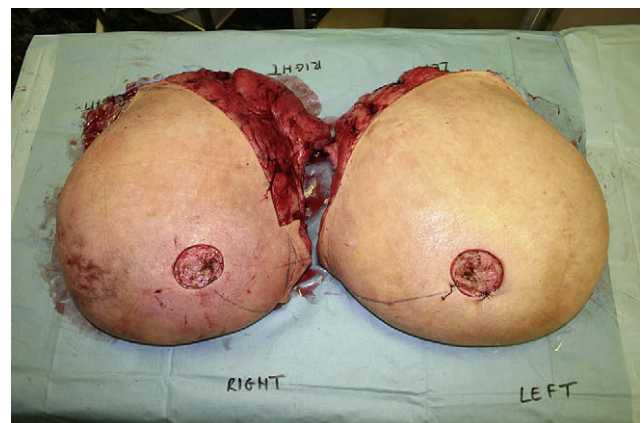
At a second consultation, 4 months later, her body weight had increased from 42.4 kg to 52 kg, and this was mostly breast tissue (Figures 1, 2). She was 145.8 cm tall and the rest of her body remained slim. The rate of increase in breast size was dramatic, and resulted in both physiological and psychological problems.

An ultrasound scan of both breasts showed oedema of the soft tissues. An MRI scan of the brain, including the pituitary gland, did not reveal an abnormality. The endocrine profile was normal.

A bilateral breast reduction was carried out using a free nipple graft technique. Six kilograms were removed from the right breast, and 6.5 kg from the left (Figure 3). There were periods of profuse bleeding and the measured total blood loss was 2900 ml, which was 80% of the patient's estimated total blood volume of 3640 ml. Four units of blood were transfused.

Following the surgery the anaesthetist noticed a significant improvement in the compliance of the patient's lungs – respiratory compliance was noted to be 22.5 ml/cmH<sub>2</sub>O soon after induction of anaesthesia, and 36.4 ml/cmH<sub>2</sub>O after completion of the resection, an improvement of 62%. This could be directly attributed to the removal of a restrictive effect on the chest wall imposed by the weight of the patient's breasts. Her body weight, recorded as 52 kg just prior to the surgery, had decreased to 39.5 kg, a reduction of 24%.

Histological examination showed minimal adipose tissue and a massive increase of stroma, containing cellular areas of fibroblast-like cells with anastomosing bands of hyalinised collagen. Some areas resembled pseudoangiomatous stromal hyperplasia (PASH).<sup>9</sup> Elsewhere there was paucicellular stroma with pseudocystic change. Throughout the stroma ducts were found, but not lobules.<sup>10</sup>



**Figure 3** Breast tissue removed (combined tissue weight: 12.5 kg).

**Table 1** Breast reduction: pedicled techniques<sup>1,11,13,15,16,21,26,28</sup>

Author	Year	Patient age	Total tissue weight (g)	Hormonal therapy	Recurrence	Follow up
Ship	1971	22	1358	Progesterone	Yes	3 months
Sperling	1973	11	3294	No	Yes	< 1 year
Mayl	1974	11	?	No	Yes	Immediate - weeks
Mayl	1974	20	3800	No	Yes	Immediate
Mayl	1974	21	3200	No	Yes	3 months
Oberman	1979	17	?	No	Yes	7 months
Ryan	1985	22	2650	Dydrogesterone	Yes	2 months
Ryan	1985	22	1320	Tamoxifen	Yes	3 months
Urribe Barreto	1991	16	?	No	Yes*	6 months
Kupfer	1992	12	5948	No	Yes	5 months
Kucukaydin	1994	13	?	No	Yes	4 months
Baker	2001	14	?	No	Yes*	10 years

\* During pregnancy.

Overall the histopathologic appearances suggested an abnormal response to hormonal stimulation and were in keeping with the clinical diagnosis of virginal hypertrophy.

## Discussion

Although 350 years have passed since Will Durston's vivid description, the treatment of macromastia in the actively enlarging breast remains difficult and controversial.<sup>5,11,12</sup>

Any surgical treatment shy of total ablation of all breast tissue may result in recurrent hypertrophy during the period of end-organ hypersensitivity.<sup>13</sup>

A few authors have suggested the use of endocrine therapy to control the growth of the breasts following reduction mammoplasty<sup>14–17</sup> and a variety of drugs have been used alone or in combination with surgical treatment to prevent recurrence.<sup>18–21</sup> Medroxyprogesterone acetate (Provera), dydrogesterone (Gynorest) and tamoxifen citrate (Nolvadex) have anti-oestrogenic properties and have proved successful in some cases of virginal hypertrophy,<sup>11,14,15</sup> but were completely ineffective in others.<sup>16</sup>

No short-term complications have been reported in the few patients treated with progesterones or tamoxifen, but

potential side effects on subsequent peri-pubertal development may outweigh the purported advantage of using these hormones.<sup>13</sup>

The main operative alternatives are total subcutaneous mastectomy or reduction mammoplasty. The more radical option, subcutaneous mastectomy, is not completely without risk of recurrence, as reported by Cardoso de Castro.<sup>22</sup> Hypertrophy of incompletely resected breast tissue will often enlarge to the point that it requires further surgical removal.<sup>23–25</sup>

Although mastectomy with implant reconstruction would seem to offer definitive treatment, there is a lifelong risk of the complications of implants in addition to the extreme nature of the surgery.

Reduction mammoplasty is the preferred first line treatment, and indeed it is the most common surgical therapy used.<sup>18</sup> However, recurrence is frequent<sup>11,16</sup> and secondary procedures may be required.<sup>22,26–29</sup>

A review of the literature, comparing pedicled versus free nipple graft techniques, reveals a noticeable difference in the rate of recurrence. Of a total of 12 cases of pedicled reductions, all recurred (Table 1), in contrast to only five of a total of 12 cases using a free nipple graft

**Table 2** Breast reduction: free nipple graft technique<sup>6,22,26,29–35</sup>

Author	Year	Patient age	Total tissue weight (g)	Hormonal therapy	Recurrence	Follow up
Fisher	1971	10	4000	No	Yes	1 month
Fisher	1971	18	?	No	Yes	8 months
Cardoso de Castro	1977	12	8000	No	Yes	6 months
Samuelov	1988	12	8200	No	Yes	8 months
Arcscott	2001	12	9500	Bromocriptine	Yes	3 months
Gaines	1936	14	4990	No	No	?
Erich	1960	13	?	No	No	?
Fisher	1971	22	?	No	No	5 months
Sagot	1990	11 1/2	1820	Dydrogesterone	No	18 months
Khan	2000	n/a	1316	No	No	1 year
O'Hare	2000	14	1305	No	No	2 years
Baker	2001	17	?	No	No	20 years



**Figure 4** Postoperative result at a 2-year follow up. There is no evidence of recurrent hypertrophy.

technique (Table 2). This difference is statistically significant ( $P = 0.005$ , Fisher's exact test).

In conclusion, an 11-year-old girl with massive virginal hypertrophy of the breasts was treated with a reduction mammoplasty using a free nipple graft technique, removing 12.5 kg of tissue in all (24% of body weight).

A 2 year follow up revealed she had no breast regrowth and did not require additional surgery (Figure 4).

A review of the literature seems to suggest that hypertrophic recurrence is less frequent when the free nipple graft technique is used.

It is suggested that more research should be carried out in order to find possible further evidence that this technique is actually associated with a lower risk of recurrence.

## References

- Oberman HA. Breast lesions in the adolescent female. *Pathol Annu* 1979;14:175.
- Daniel WA, Mathews MD. Tumors of the breast in adolescent females. *Pediatrics* 1968;41:743.
- Bower R, Bell MJ, Ternberg JL. Management of breast lesions in children and adolescents. *J Pediatr Surg* 1976;11:337.
- Deaver JB, McFarland J. *The breast: its anomalies, its diseases and their treatment*. Philadelphia: Blakiston; 1917.
- Durston W. *Concerning a very sudden and excessive swelling of a women's breasts*. Phil Trans, vol. IV, for Anno 1669, pp 1047–1049. London: Royal Society; 1670.
- Fisher W, Smith JW. Macromastia during puberty. *Plast Reconstr Surg* 1971;47:445.
- Gillispie JB, Hurter AJ. Virginal hypertrophy of the breast. *J Pediatr* 1949;35:240.
- Wulsin JH. Large breast tumors in adolescent females. *Ann Surg* Jul 1960;152:151–9.
- Gow KW, Mayfield JK, Lloyd D, et al. Pseudoangiomatous stromal hyperplasia of the breast in adolescent females. *Am Surg* Jul 2004;70:605–8.
- Eliassen CA, Cranor ML, Rosen PP. Atypical duct hyperplasia of the breast in young females. *Am J Surg Pathol* 1992;16:246–51.
- Mayl N, Vasconez LO, Jurkiewicz MJ. Treatment of macromastia in the actively enlarging breast. *Plast Reconstr Surg* 1974;54:6–12.
- Lettreman G, Schurter M. Will Durston's "mammoplasty". *Plast Reconstr Surg* 1974;53:48.
- Kupfer D, Dingman D, Broadbent R. Juvenile breast hypertrophy: report of a familial pattern and review of the literature. *Plast Reconstr Surg* Aug 1992;9:303–9.
- Gliosci A, Presutti F. Virginal gigantomastia: validity of combined surgical and hormonal treatments. *Aesthetic Plast Surg* 1993;17:61–5.
- Sperling RL, Gold JJ. Use of anti-estrogen after reduction mammoplasty to prevent recurrence of virginal hypertrophy of breasts. *Plast Reconstr Surg* 1973;52:439.
- Ryan RF, Pernoll ML. Virginal hypertrophy. *Plast Reconstr Surg* 1985;75:737–42.
- Taylor PJ, Cumming DC, Corenblum B. Successful treatment of D-penicillamine-induced breast gigantism with danazol. *Br Med J* 1981;282:362.
- Bauer BS, Jones KM, Talbot CW. Mammary masses in the adolescent female. *Surg Gynecol Obstet* 1987;165:63–5.
- Gargan TJ, Goldwyn RM. Gigantomastia complicating pregnancy. *Plast Reconstr Surg* 1987;80:121–4.
- Kullander S. Effect of 2 br-alpha- ergocryptin (CB 154) on serum prolactin and the clinical picture in a case of progressive gigantomastia in pregnancy. *Ann Chir gynaecol* 1976;65:227–33.
- Kucukaydin M, Kurtoglu S, Okur H, et al. Virginal hypertrophy. Case report. *Turk J Pediatr* 1994;36:243–8.
- De Castro CC. Subcutaneous mastectomy for gigantomastia in adolescent girl. Recurrence after reduction mammoplasty. *Plast Reconstr Surg* 1977;59:575–8.
- Urribe Barreto A. Juvenile mammary hypertrophy. *Plast Reconstr Surg* Mar 1991;87:583–4 [correspondence].
- Hollingsworth D, Archer R. Massive virginal breast hypertrophy at puberty. *Am J Dis Child* Feb 1973;125:293–5.
- Fournas DW. Subcutaneous mastectomy for juvenile hypertrophy of the breast: report of a case. *Br J Plast Surg* 1982;35:367–70.
- Baker SB, Burkey BA, Thornton P, et al. Juvenile gigantomastia: presentation of four cases and review of the literature. *Ann Plast Surg* May 2001;46:517–25.
- Netscher DT, Mosharafa AM, Laucirica R. Massive asymmetric virginal breast hypertrophy. *South Med J* 1996;89:434–7.
- Ship AG, Shulman J. Virginal and gravid mammary gigantism: recurrence after reduction mammoplasty. *Br J Plast Surg* 1971;24:396–401.
- Samuelov R, Siplovich L. Juvenile gigantomastia. *J Pediatr Surg* 1988;23:1014.
- Arcscott GD, Craig HR, Gabay L. Failure of bromocriptine therapy to control juvenile mammary hypertrophy. *Br J Plast Surg* Dec 2001;54:720–3.
- Gaines JA. Massive puberty hypertrophy of the breast. *Am J Obstet Gynecol* 1937;34:130.
- Erich JB. Plastic correction of excessive hypertrophy of the breasts at puberty: report of a case. *Proc Staff Meet Mayo Clin* 1960;35:192.
- Sagot P, Mainguene C, Barriere P, et al. Virginal breast hypertrophy at puberty; a case report. *Eur J Obstet Gynecol Reprod Biol* Mar 1990;34:289–92.
- Khan A, Mohammed-Emamdee R, Lalla R, et al. Massive virginal breast hypertrophy. *West Indian Med J* Jun 2000;49:181–2.
- O'Hare PM, Frieden IJ. Virginal breast hypertrophy. *Pediatr Dermatol* 2000;17:277–81.