Case Report

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Extreme Idiopathic gigantomastia

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ABSTRACT

Gigantomastia is a rare mastopathy of unknown cause. Due to mechanical and psychological complications related to excessive breast weights and volume, effective surgical treatment is required. Most cases of gigantomastia in the literature are associated with pregnancy or puberty and very rare cases of spontaneous gigantomastia have been reported We report a 38 years old woman with an idiopathic gigantomastia treated successfully with Thorek technique.

Keywords: Gigantomastia, Mammoplasty, Surgery, Treatment

INTRODUCTION

Gigantomastia is defined by the benign bilateral enlargement of the breast to a degree that requires a breast surgery reduction removing more than 1500 mg per breast. Gigantomastia's aetiologies were subdivided in four categories: Idiopathic, pregnancy associated, juvenile and drug induced.

Whereas, previous reports showed that pregnancy induced, and juvenile forms are the more common.² To our knowledge, idiopathic gigantomastia have been reported in only 11 cases.^{3,4} Authors report a rare case of idiopathic gigantomastia successfully treated with breast surgery reduction.

CASE REPORT

A thirty-eight years old Congolese woman attended our Institute for progressive enlargement of the breasts evolving since 7 years. She had restriction of her movement due to a severe backache. She needed help mobilising and bathing.

She was pre-menopausal and had four children. Her youngest child was 9 year-old. She had breast feed all of her children about one year. Her menarche was 14 years and had regular menstruations since then. She was not taking any medication including contraceptive pills.

Her body mass index was 29,25Kg/m². On physical examination, she had enormously hypertrophic breasts reaching below the waistline (Figure 1 and Figure 2). The right breast was obviously larger than the right breast. She had multiples scars and bleeding ulcerations located in the lower quadrants due to the chronic frictions with her bra. Palpations didn't find any mass.

Breast sonography finds galactophoric dilatation with several simples cysts. We were unable to perform mammography or magnetic resonance imagery due to the large size and density of the breasts. Laboratory essays for hormone levels including prolactin, progesterone, oestrogen, β -human chronic gonadotrophin, thyroid stimulating hormone and growth hormone were normal. On blood sampling, her hemoglobin level was 8,2 g/dL probably explained by the ulceration bleedings in the breasts.



Figure 1: Gigantomastia in a 38 years old woman before mammoplasty- frontal view.



Figure 1: Gigantomastia in a 38 years old woman before mammoplasty- lateral view.

She had blood transfusion and then was operated under general anaesthesia. She had a bilateral reduction mammaplasty with free nipple grafts (Figure 3). The specimen weighted 21,7Kg in total. Post operative course

was eventful. The patient was discharged at the third day.



Figure 3: One month post operative results.

The histological examination showed nodular proliferation of glandular breast tissue with oedematous fibrous stroma and focal lactational change.

After 6 months of the surgery there was no relapse in the hypertrophy of the breasts with a satisfactory esthetic result.

DISCUSSION

Idiopathic gigantomastia is extremely rare condition. Although the actual pathophysiology is not known, there are several theories that could explain the abnormal enlargement of the breast. Since most of the cases of gigantomastia are diagnosed within pregnancy or puberty, excessive release of endocrinology hormones or increased hormonal sensivity are suspected to be the cause.²

Juvenile gigantomastia can occur during per pubertal period and sometimes occurring with the onset of the larch. Levels of estrogen and progesterone are usually normal in this setting.⁵ Similar to fibroadenomas, this growth of normal breast tissue is explained by excess sensitivity to normal levels of hormones. Noczynska et al, report the cases of two 14- and 15-year-old girls with high levels of estrogen receptors in the mammary tissue. Immunohistochemical examination revealed hormones.6 hypersensitivity to these Juvenile gigantomastia can also occur in the context of Cowden syndrome.⁷ Gestational gigantomastia complicate 1 to 118000 deliveries.8 It starts usually in the first or second trimester of gestation.9 Two mechanisms are suspected to be the cause of gestational gigantomastia. An elevated prolactine level has been reported by Lafreniere et al, in an 18 years old patient. The administration of Bromocriptine stopped the enlargement of the breasts confirming the hormonal cause of gigantomastia.¹⁰

Another mechanism was hypothetitised by Gargan et al, consists in the creation of an antibody similar with Grave disease, which may interfere with the hormone receptor complex. 11 Several reports also described drug-induced gigantomastia such as D-penicillamine, neothetazone, cyclosporin and bucillamine. 12,13 Idiopathic gigantomastia is even more exceptional, reaching the adult woman over 20 years of age outside pregnancy. Histologically, there is hypertrophy of the connective tissue rather than hyperplasia of the glandular tissue. 14 Hedberg et al, reported an intracellular accumulation of a substance rather than a proliferation of connective tissue. 15

Due to the rarity of this condition, treatment is not well codified in gigantomastia. Hormonal treatment has been used to control the breast growth by many authors. Tamoxifen had been widely used instead of surgery in the juvenile forms or post-operatively to prevent relapses. ^{5,16} Bromocriptine was able to decrease the size of the breasts in a 26 years-old woman who had a pregnancy associated form without jeopardizing the infant to born. ¹⁷ Corticoids and diuretics may be used also post-operatively. ⁵

Surgical treatment has been considered after failure of medical treatment or upfront especially in the idiopathic and drug induced forms.² The standard technique is the described by Thorek in 1922 consisting in a nipple free reduction mammoplasty. The main inconvenient with this technique is a non-aesthetic breast and nipple with poor projection. It have been replaced with dermoglandular pedicle techniques.¹⁸ The main risk after mammoplasty is frequent recurrent hypertrophy of the remaining breast tissue.⁴ Pregnancy could be an aggravating factor due to the relative elevation of oestrogen.¹⁹

Some authors recommend a delay of pregnancy of two years after surgery. Which Knowing this risk of relapse, some authors prefer mastectomy with reconstruction employing tissue expanders or autologus breast reconstruction procedures mainly in the juvenile or pregnancy associated gigantomastia. The normal breast tissue remaining after mammoplasty after re-growth in an aggressive way. Nevertheless, axillary recurrence after mastectomy have been described by several reports. 1.5,20

CONCLUSION

Idiopathic gigantomastia is a rare entity, of indeterminate etiology. Diagnosis requires a careful elimination of other, more frequent causes, of gigantomatia such as pregnancy, juvenile or drug induced. It raises the problem of surgical treatment that can very rarely be radical. Long-term follow-up is necessary because of the possibility even after a radical surgery.

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REFERENCES

- 1. Kulkarni D, Beechey-Newman N, Hamed H, Fentiman IS. Gigantomastia: A problem of local recurrence. Breast. 2006;15(1):100-2.
- Dancey A, Khan M, Dawson J, Peart F. Gigantomastia--a classification and review of the literature. J Plast Reconstr Aesthet Surg. 2008;61(5):493-502.
- 3. Cho MJ, Yang JH, Choi HG, Kim WS, Yu YB, Park KS. An idiopathic gigantomastia. Ann Surg Treat Res. 2015;88(3):166-9.
- Chargui R, Houimli S, Damak T, Khomsi F, Ben Hasouna J, Gamoudi A, et al. [Relapse of gigantomastia after mammoplasty. Report of a case and literature review]. Ann Chir. 2005;130(3):181-5.
- 5. Baker SB, Burkey BA, Thornton P, Larossa D. Juvenile gigantomastia: presentation of four cases and review of the literature. Annals of Plastic Surgery. 2001;46(5):517-26.
- Noczyńska A, Wasikowa R, Myczkowski T. Hypersensitivity of estrogen receptors as a cause of gigantomasty in two girls. Polski merkuriusz lekarski: organ Polskiego Towarzystwa Lekarskiego. 2001;11(66):507.
- 7. Sood A, Garg R, Saily R, Dash R. A patient with congenital hypertrichosis, gum hyperplasia and macromastia. Journal of Pediatric Endocrinology and Metabolism. 2000;13(5):561-4.
- 8. Beischer NA, Hueston JH, Pepperell RJ. Massive Hypertrophy of the Breasts in Pregnancy: Report of 3 Cases and Review of the Literature, never think you have seen everything. Obstetrical and Gynecological Survey. 1989;44(4):234-43.
- 9. Swelstad MR, Swelstad BB, Rao VK, Gutowski KA. Management of gestational gigantomastia. Plast Reconstr Surg. 2006;118(4):840-8.
- 10. Lafreniere R, Temple W, Ketcham A. Gestational macromastia. The American Journal of Surgery. 1984;148(3):413-8.
- 11. Gargan TJ, Goldwyn RM. Gigantomastia Complicating Pregnancy. Plastic and Reconstructive Surgery. 1987;80(1):121-4.
- 12. Taylor P, Cumming D, Corenblum B. Successful treatment of D-penicillamine-induced breast gigantism with danazol. British Medical Journal (Clinical research ed). 1981;282(6261):362.
- 13. Sakai Y, Wakamatsu S, Ono K, Kumagai N. Gigantomastia induced by bucillamine. Annals of Plastic Surgery. 2002;49(2):193-5.
- Mamouni N, Erraghay S, Oufkir A, Saadi H, Bouchikhi C, Banani A. [Gigantomastia: report of a case and review of the literature]. Pan Afr Med J. 2014;18:154.

- 15. Albert H. Diffuse idiopathic hypertrophy of the mammary glands of the female: a report of a new case and a consideration of the etiology and pathology based on data of recorded cases. Journal of the American Medical Association. 1910;55(16):1339-43.
- 16. Demir K, Unuvar T, Eren S, Abaci A, Bober E. Tamoxifen as first-line treatment in a premenarchal girl with juvenile breast hypertrophy. Journal of Pediatric and Adolescent Gynecology. 2010;23(5):e133-e6.
- 17. Hedberg K, Karlsson K, Lindstedt G. Gigantomastia during pregnancy: effect of a dopamine agonist. American Journal of Obstetrics and Gynecology. 1979;133(8):928-31.
- 18. Lacerna M, Spears J, Mitra A, Medina C, McCampbell E, Kiran R. Avoiding free nipple grafts during reduction mammaplasty in patients with gigantomastia. Annals of Plastic Surgery. 2005;55(1):21-4.
- 19. Ship AG, Shulman J. Virginal and gravid mammary gigantism- recurrence after reduction mammaplasty. British Journal of Plastic Surgery. 1971;24:396-401.
- 20. Boyce SW, Hoffman Jr PG, Mathes SJ. Recurrent macromastia after subcutaneous mastectomy. Annals of plastic surgery. 1984;13(6):511-8.

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