

Case Report

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Bilateral gynecomastia: a report of one case

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Abstract

Background: Gynecomastia is defined as the presence of excessive breast tissue in males, which can appear unilateral or bilateral. Bilateral gynecomastia is frequently found in the neonatal period, early in puberty, and with increasing age. Prepubertal bilateral gynecomastia in the absence of endocrine abnormalities is extremely rare, with only a few cases in literature.

Methods: We report the case of a healthy boy of 17 years old with bilateral breast masses. No abnormalities were found on ultrasonography and all endocrine parameters were within normal limits. Treatment consisted of peripheral liposuction followed by subcutaneous partial resection of the gland, conducted through an infra-areolar incision and a left side drainage in aspiration for a week for firm tissue removal intraoperative skin laceration.

Results: Microscopy of the subcutaneous mastectomy specimen revealed gynecomastia without signs of malignancy. Postoperative course of patient was uncomplicated, with no signs of recurrence of breast tissue.

Conclusions: Atypical presentations of gynecomastia are often not recognized, with little attention to breast development in prepubertal non-obese children. Since prepubertal gynecomastia could be a sign of possible underlying diseases, a thorough examination and further research is recommended. If there is no causal treatment, surgical resection is the therapy of first choice. Peripheral liposuction and surgical resection of the gland tissue are the mainstay of treatment. In summary, we describe two cases of prepubertal unilateral gynecomastia with a normal endocrine workup. Further research is needed to establish the pathophysiologic mechanisms of prepubertal gynecomastia, since underlying etiology in most cases remains unclear.

Keywords: Bilateral gynecomastia, puberty, Etiology, Surgery

Introduction

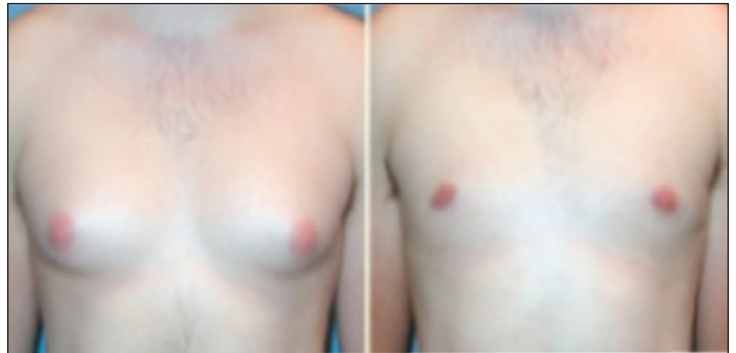
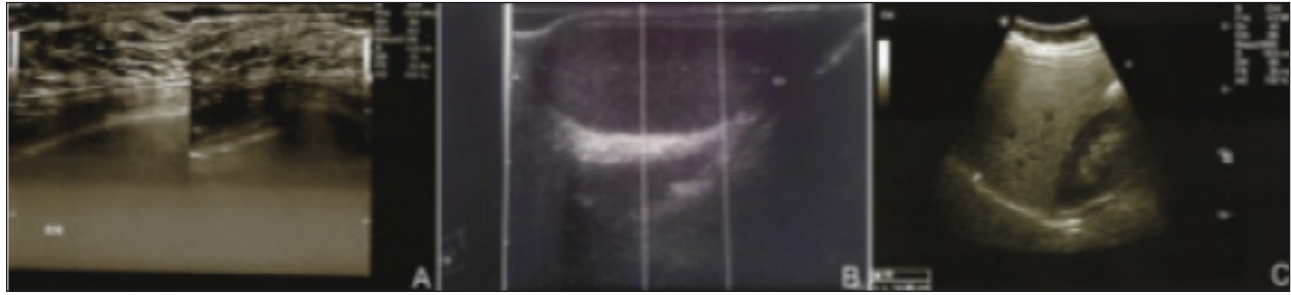
Gynecomastia is characterized by the presence of unilateral or bilateral breast tissue in males. This benign condition accounts for 60% of all disorders of the male breast [1,2]. While bilateral gynecomastia is common in the neonatal period, early in puberty, and with increasing age, prepubertal unilateral gynecomastia is a rare condition, with only a few cases in literature [3]. We report a unique clinical presentation of a boy with pubertal onset of bilateral gynecomastia and discuss the etiology, workup, and therapeutic intervention.

Case report

Patient

An 17-year-old boy presented with a 18-months history of bilateral breast enlargement. The patient also complained of tenderness of the breast mass and of great inconvenience in everyday life. In this case, there was no family history of breast malignancies or gynecomastia. No drugs or dermal applications were used. Physical examination showed a healthy-appearing lively boy. The patient was 178 cm tall without accelerated growth velocity, weighing 68 kg.

Palpation of the right breast revealed a firm mass measuring approximately 5 cm in diameter, with tenderness on deep palpation, Tanner stage V. There was a pronounced asymmetry of the breast, with abnormal-appearing right breast (Fig.) and minor in left breast 3 cm mass measuring. No history or sign of galactorrhea existed.



A summary of all endocrine parameters tested with corresponding results is given in Table 1. All parameters were found to be within normal limits. Ultrasound examination of the right breast showed retro-aureolar glandular tissue with normal aspects and swelling containing subcutaneous fat tissue. An abdominal computed tomography scan excluded any estrogen-producing tumor. Surgery (conducted by MB - AC) under general anesthesia consisted of peripheral liposuction (with the 2 mm PAL liposuction system) and subcutaneous partial resection of the gland, conducted through an infra-areolar incision. The specimen dissected measured $5.0 \times 3.0 \times 2.5$ cm. and $3.2 \times 2.1 \times 1.5$ cm on the left side where we needed a chest tube drainage for skin tissue removal intraoperative laceration [see pics 2]. Pathologic examination revealed normal glandular breast tissue, without evidence of malignancy. Microscopy of the subcutaneous mastectomy specimen showed mammary ducts with hypertrophic ductuli and mild lymphocytic infiltrates lining the ducts. The concluding diagnosis was normal gynecomastia tissue. Recovery was uneventful after a week. At a postoperative clinical evaluation 2 months later, there were no signs of breast development (Fig. 1b).

Discussion

Gynecomastia is a condition in which the glandular components of the male breast proliferate, resulting in an enlargement of one or both breasts. In the age distribution, three distinct peaks are identified. The first is found in the neonatal period, in which palpable breast tissue develops in 60% to 90% of all newborns

due to transfer of estrogens across the placenta. The second peak occurs during puberty, as a result of an imbalance between estrogens and androgens within the breast tissue. The last peak is found in the adult population, with the highest prevalence among 50- to 80-year-old males [1,3]. Common known causes for gynecomastia in adults include liver disease, as well as the use of drugs such as digitalis and tricyclic antidepressants [5]. In contrast to gynecomastia in adolescent boys and adult men, prepubertal gynecomastia is rare.

A specific cause is hardly ever identified, and in 90% of patients, prepubertal gynecomastia is classified as idiopathic [2,6]. Known causes of breast enlargement in children are diverse [3,6-8]. Therefore, further exploration of the etiology is advised, particularly to rule out any endocrine or malignant abnormalities. A variety of endocrinopathies, mostly as a result of an increased ratio of circulating estrogens to androgens, induce stimulation of breast tissue leading to gynecomastia. Calzada et al. showed that the presence of hormone receptors in gynecomastia may provide a setting favorable for mammary glands to develop gynecomastia [9]. The family history of our patient revealed gynecomastia on the father's side. Stratakis et al. described increased extraglandular aromatization of androgens to cause the unusual entity of familial gynecomastia, named aromatase excess syndrome [10].

This syndrome has been correlated to serum estradiol and estrone excess, which were within normal limits in both of our patients. Therefore, a mild aromatase excess syndrome could be excluded as a cause of the prepubertal gynecomastia and familial occurrence.

None of our patients were exposed to exogenous estrogens. However, our patient was using Melatonin, of which in literature only one case is reported. The case described a possible relationship of the usage of this drug and a painful bilateral gynecomastia in an adult male [11], making the relationship between this drug and gynecomastia in our first patient very unlikely. Furthermore, obesity is documented in 31% of the boys, with excessive fat tissue as a possible cause of prepubertal gynecomastia [3]. Increased adipose tissue causes an increased aromatization of mostly testosterone in adipose tissue, leading to an imbalance between estradiol and testosterone levels.

Our patient had no signs of being overweight; therefore, obesity was excluded as the etiology for the gynecomastia.

In summary, atypical presentations of gynecomastia are frequently underappreciated, with little attention to breast enlargement in the nonobese prepubertal and pubertal boy. Since prepubertal gynecomastia could be a sign of possible underlying diseases, a thorough examination and further research is recommended. If a reversible cause can be excluded, peripheral liposuction in combination with surgical resection of the gland tissue by an infra-areolar approach is the first therapy of choice. We report this case of pubertal bilateral gynecomastia, in which the cause of the gynecomastia remains unclear, while none of the above discussed causes could be identified.

Our patient had no signs of being overweight; therefore, obesity was excluded as the etiology for the gynecomastia.

References

1. Braunstein GD. (1993) Gynecomastia. *N Engl J Med.* 328(7):490–495. doi: 10.1056/NEJM199302183280708.

[PubMed] [Cross Ref]

2. Cho YR, Jones S, Gosain AK. (2008) Neurofibromatosis: a cause of prepubertal gynecomastia. *Plast Reconstr Surg.* 121(3):34e–40e. doi: 10.1097/01.prs.0000299299.46365.

7e. [PubMed] [Cross Ref]

3. Einav-Bachar R, Phillip M, Aurbach-Klipper Y, Lazar L. (2004) Prepubertal gynecomastia: aetiology, course and outcome. *Clin Endocrinol.* 61(1):55–60. doi: 10.1111/j.1365-2265.2004.02059.x. [PubMed] [Cross Ref]

4. Marshall WA, Tanner JM. (1970) Variations in the pattern of pubertal changes in boys. *Arch Dis Child.* 45(239):13–23. doi: 10.1136/adc.45.239.13. [PMC free article] [PubMed] [Cross Ref]

5. Harigopal M, Murray MP, Rosen PP, Shin SJ. (2005) Prepubertal gynecomastia with lobular differentiation. *Breast J.* 11(1):48–51. doi: 10.1111/j.1075-122X.2005.21442.x. [PubMed] [Cross Ref]

6. Henley DV, Lipson N, Korach KS, Bloch CA. (2007) Prepubertal gynecomastia linked to lavender and tea tree oils. *N Engl J Med.* 356(5):479–485. doi: 10.1056/NEJMoa064725. [PubMed] [Cross Ref]

7. Ersoy B, Yoleri L, Riza Kandiloğlu A. (2002) Unilateral galactoceles in a male infant. *Plast Reconstr Surg.* 109(1):401–402. doi: 10.1097/00006534-200201000-00076. [PubMed] [Cross Ref]

8. Kauf E. (1998) Gynecomastia in childhood: pathologic causes unusual but serious. *Fortschr Med.* 116(35–36):23–26. [PubMed]

9. Calzada L, Torres-Calleja J, Martinez JM, Pedrón N. (2001) Measurement of androgen and estrogen receptors in breast tissue from subjects with anabolic steroid-dependent gynecomastia. *Life Sci.* 69(13):1465–1469. doi: 10.1016/S0024-3205(01)01227-9. [PubMed] [Cross Ref]

10. Stratakis CA, Vottero A, Brodie A, et al. (1998) The aromatase excess syndrome is associated with feminization of both sexes and autosomal dominant transmission of aberrant P450 aromatase gene transcription. *J Clin Endocrinol Metab.* 83(4):1348–1357. doi: 10.1210/jc.83.4.1348. [PubMed] [Cross Ref]

11. Bleeker JL, Lamont BH, Verstraete AG, Schelfhout VJ. (1999) Melatonin and painful gynecomastia. *Neurology.* 53(2):435–436. [PubMed]