Letter to the Editor

Unilateral juvenile (virginal) hypertrophy of the breast

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To the Editor,

We have read the case report entitled "Unilateral juvenile (virginal) hypertrophy of the breast" published in the Turkish Journal of Pediatrics (2008; 50: 278-281). To the best of our understanding, the patient is the first evaluated in the Department of Pediatric Hematology-Oncology, and the diagnosis was established with biopsy, which was performed following general surgery consultation. However, the medical history, age at onset of pathologically rapid breast enlargement as well as presence of erythema and tenderness are sufficient for a clinical diagnosis of "juvenile (virginal) hypertrophy of the breast". In addition, imaging studies are useful in further differential diagnosis of juvenile breast hypertrophy from benign or malignant tumors^{1,2}. We think that the patient underwent an unnecessary invasive procedure since this entity is well known by pediatric endocrinologists and should be diagnosed based on history and physical examination. Furthermore, following the diagnosis, the patient was left untreated (as shown in Figure 1 in the article) owing to the stable clinical picture. However, it would have been more appropriate if the case underwent a surgical intervention by the end of pubertal development.

In our practice, we observed favorable results with tamoxifen in a 12.5-year-old female patient, who presented during rapid breast enlargement. We prefer surgical correction in patients who present later in the course of the disease with the aim of improving mastalgia, back pain, limited motion, and cosmetic problems.

With this letter, we want to (i) emphasize that unnecessary interventions should be avoided and (ii) underline the necessity of a continuous multidisciplinary approach to such patients.

REFERENCES

- 1. Kuhl CK. MRI of breast tumors. Eur Radiol 2000; 10: 46-58.
- 2. Touraine P, Youssef N, Alyanakian A, et al. Breast inflammatory gigantomastia in a context of immune mediated diseases. J Clin Endocrinol Metabol 2005; 90: 5287-5294.

Reply,

We are grateful for the comments of Dr. Böber and Dr. Büyükgebiz about our article in the Turkish Journal of Pediatrics (2008; 50: 278-281). This case was firstly admitted to the Department of General Surgery of our center in August 2001, and Pediatric Oncology consultation was done after biopsy was performed. Ultrasonographic (US) examination is a useful method for evaluation of breast masses in children and adolescents, and usually further radiologic evaluation is not necessary¹. Nowadays magnetic resonance imaging (MRI) of the breast is also considered a useful method for detection of invasive cancers, preinvasive cancers, and premalignant lesions, and it has also been proposed as an additional screening method for young women at high risk of breast cancer²,³. The breast architecture and pathologic lesions may be defined by MRI; nevertheless, MRI is not able to distinguish between benign, inflammatory lesions and carcinoma⁴. Because of the rarity of breast tumors during childhood and adolescence, experience in radiologic evaluation of breast masses is also limited in children. In our case, US findings were considered as giant fibroadenoma or juvenile adenomatoid tissue. Although malignant breast tumors of childhood, including malignant carcinoma, malignant or borderline phylloides tumor, and breast lymphoma

are seen relatively rarely, they should be considered in the differential diagnosis⁵⁻⁷. Fine needle aspiration cytology (FNAC) of breast masses in children may be helpful in the differential diagnosis⁷. Because the clinical and radiologic findings were suspicious for malignant or borderline breast tumors, biopsy was considered for definitive diagnosis in this case. Diagnosis of juvenile breast hypertrophy was done by incisional biopsy without any sequelae. She was also consulted with our Pediatric Endocrinology Department, and hormonal treatment with antiestrogen drugs (tamoxifen) was not offered because of its unknown long-term effects in children at that time. Furthermore, both the effectiveness and ineffectiveness of tamoxifen has been published in the literature, and treatment experience with tamoxifen is still limited in childhood juvenile breast hypertrophy⁸⁻¹⁴. As we stated before, no medical or surgical treatment was performed in this case. She was followed up without any further enlargement of the right breast by the Pediatric Oncology and General Surgery Departments until 18 years of age, and was then followed up solely by our General Surgery Department. We agree that unnecessary surgical interventions should be avoided and that a multidisciplinary approach is necessary for such patients.

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REFERENCES

- 1. García CJ, Espinoza A, Dinamarca V, et al. Breast US in children and adolescents. Radiographics 2000; 20: 1605-1612.
- 2. Riedl CC, Ponhold L, Flöry D, et al. Magnetic resonance imaging of the breast improves detection of invasive cancer, preinvasive cancer, and premalignant lesions during surveillance of women at high risk for breast cancer. Clin Cancer Res 2007; 13: 6144-6152.
- 3. Lord SJ, Lei W, Craft P, et al. A systematic review of the effectiveness of magnetic resonance imaging (MRI) as an addition to mammography and ultrasound in screening young women at high risk of breast cancer. Eur J Cancer 2007; 43: 1905-1917. Epub 2007 Aug 2.
- 4. Touraine P, Youssef N, Alyanakian MA, et al. Breast inflammatory gigantomastia in a context of immune-mediated diseases. J Clin Endocrinol Metab 2005; 90: 5287-5294. Epub 2005 Jun 21.
- 5. Gutierrez JC, Housri N, Koniaris LG, Fischer AC, Sola JE. Malignant breast cancer in children: a review of 75 patients. J Surg Res 2008; 147: 182-188. Epub 2008 Apr 14.
- 6. Di Noto A, Pacheco BP, Vicala R, Itala J, Pellegrino J, Mendez Ribas J.Two cases of breast lymphoma mimicking juvenile hypertrophy. J Pediatr Adolesc Gynecol 1999; 12: 33-35.
- 7. Kapila K, Pathan SK, Al-Mosawy FA, George SS, Haji BE, Al-Ayadhy B. Fine needle aspiration cytology of breast masses in children and adolescents:experience with 1404 aspirates. Acta Cytol 2008; 52: 681-686.
- 8. Gerber B, Krause A, Lochmüller EM, Janni W, Friese K. Juvenile gigantomastia. Case report and literature survey. Zentralbl Gynakol 2004; 126: 167-169.
- 9. Baker SB, Burkey BA, Thornton P, LaRossa D. Juvenile gigantomastia: presentation of four cases and review of the literature. Ann Plast Surg 2001; 46: 517-525; discussion 525-526.
- 10. O'Hare PM, Frieden IJ. Virginal breast hypertrophy. Pediatr Dermatol 2000; 17: 277-281.
- 11. Küçükaydin M, Kurtoğlu S, Okur H, et al. Virginal hypertrophy. Case report. Turk J Pediatr 1994; 36: 243-248.
- 12. Morimoto T, Komaki K, Mori T, et al. Juvenile gigantomastia: report of a case. Surg Today 1993; 23: 260-264.
- 13. Ryan RF, Pernoll ML. Virginal hypertrophy. Plast Reconstr Surg 1985; 75: 737-742.
- 14. Gerber B, Krause A, Lochmüller EM, Janni W, Friese K. Juvenile gigantomastia. Case report and literature survey. Zentralbl Gynakol 2004; 126: 167-169.