Giant Breast Hypertrophy in Juvenile: A Rare Case

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ABSTRACT
Introduction: Juvenile hypertrophy of the breast (JHB) is a benign condition that can lead to gigantomastia in adolescence. The development of breast enlargement in adolescence can cause both physical and psychosocial problems. Therefore, appropriate investigation and proper management at an early stage of the disease are very important. One of the treatment modalities is breast reduction surgery which ideal and offers an improvement in the quality of life.

Case Presentation: A 13-year-old female with massive bilateral breast enlargement for a period of 8 months. The patient also complained of severe back pain and neck discomfort since her breast progressively enlarged. Psychosocial problems were recorded. Right breast reduction was performed and closed with inverted-T mammoplasty. The post-operative period was uneventful, and the patient was discharged on day 5 after the operation.

Conclusions: Reduction mammoplasty is well accepted by adolescents with juvenile breast hypertrophy and can improve physical and psychological outcomes.

INTRODUCTION
Juvenile hypertrophy of the breast (JHB) is a benign condition that can lead to gigantomastia in the peripuberty of female adolescents. This kind of pathologic lesion rapidly and massively increase in size over a short period of time [1]. It often follows a 6-month period of extreme breast enlargement, superseded by a longer period of slower but sustained breast growth [2]. It is relatively a rare condition. In 2011, Hoppe et al in their meta-analysis of case reports identified 65 reported cases between 1910–2009 [3]. Hisham et al. [4] study yielded additional nine cases from 2010–2017.

The etiology of JHB has not been fully explained, but several theories have been proposed. The popular theory includes the imbalance of endogenous hormone production [5]. Some suggest that it may be a consequence of an increasingly unhealthy lifestyle, a hormone-laden diet and obesity [6].

The development of macromastia in adolescence leads to a distressing condition during the peripuberty period. It causes physical and psychosocial problems. Physical problems such as back pain and shoulder pain. Social issues arise secondary to poor fitting clothing, negative body image, and trouble in exercising. Therefore, appropriate investigation and proper management at the early stage of the disease are very important. Breast reduction surgery is ideal and offers improvement with those problems mentioned above. The management of this disease includes hormonal interventions, surgery, and a combination to prevent recurrence [7].

Here, we presented a case of a 13-year-old girl with giant juvenile breast hypertrophy who underwent right breast reduction mammoplasty.

CASE PRESENTATION
A 13-year-old female came with a chief complaint of massive bilateral breast enlargement for a period of 8 months (Figure 1). There were no sores, discharge, or itchy. The patient also complained of severe back pain and neck discomfort since her breast progressively enlarged. This causes difficulty in finding a fit clothing for her, social embarrassment that incapacitating her from school and social activities. There was no history of trauma, fever, anorexia, or weight loss. She attained thelarche at 11 years old and menarche at 12 years old. Her past medical dan family history was unremarkable.

Clinical examination revealed that the breasts were asymmetrical and disproportionately enlarged with the
widened areola. The right breast lump measure 22cm x 20cm x 18cm and left breast lump measured 22cm x 21cm x 30cm. Both the lumps were solitary, well-circumscribed, and not fixed to the deeper structures. The left breast demonstrated an oval-shaped with skin ulceration area overlying the lump. There was no blood or nipple discharge on both breasts. There was no axillary lymphadenopathy.

Routine hematological and biochemical examinations were within normal limits. The patient has undergone a left breast biopsy one month prior to surgery at the previous hospital. The histopathology result showed mammary dysplasia with increased cellularity of stromal (Figure 2). There was no evidence of malignancy.

Right breast reduction was performed with removal of the excess tissue and nipple (Figure 3.A). The wound was closed with an inverted-T mammoplasty (Figure 3.B). No intraoperative blood transfusion was needed. The post-operative period was uneventful, and the patient was discharged on day 5 after the operation. Post-operative scar day 14 was good without remarkable complications.

DISCUSSION

Juvenile hypertrophy of the breast (JHB) is a rare case where continued breast growth occurs during puberty. Neinstein reviewed 15 publications regarding breast lesions in adolescents spanning a period of nearly 40 years and reported that JHB accounts for only 2% of all breast lesions in this group of patients [8]. In most cases, the clinical diagnosis of JHB is so singular and striking with consideration of other breast pathologies [9]. The differential diagnosis of JHB includes giant fibroadenomas, phyllodes tumor, and malignant tumor such as lymphoma and sarcomas. According to Neinstein the prevalence of this condition in adolescents with breast lesions was 1%, 0,4%, and 0,9% respectively.8 Although malignant tumor of the breast are rare in this age group, 2% of all primary malignant breast lesions occur under the age of 25 years in female [10].

The term gigantomastia may be used to refer to cases of extreme breast enlargement. This enlargement may be unilateral or bilateral and can occur at any time during puberty, sometimes occurring with the onset of thelarche [2]. In this case, the patient has bilateral breast enlargement that occurred few months after of her menarche.

The underlying etiology of JHB has not been fully explained, but several theories have been proposed. The popular theory includes an imbalance of endogenous hormone production, end-organ hypersensitivity to normal levels of gonadal hormones [5]. Some experts estimated that the pathophysiology of JHB is due to an overreaction of breast tissue to the estrogen hormone [11–13]. However, previous studies have found normal levels of estrogen, progesterone, gonadotropins, and growth hormone during the phase or rapid growth [2]. Even though hormonal levels weren’t determined in this case, but it may be necessary to examine the levels of estrogen and progesterone receptors from samples of breast tissue.

Ultrasound examination of the breast is rarely useful to rule out differential diagnoses. MRI can be more useful for determining breast architecture and
pathological lesions. Breast ultrasound will provide a picture of regular, hyperechoic mass that is like the picture of giant fibroadenoma ultrasound [14,15].

In this case, the breast tissue grows rapidly and massively for over 8 months. Due to a giant size of breast enlargement, the patient has difficulty finding clothes that fit in and social embarrassment that incapacitating her from school and social activities. As reported in adults with macromastia, the adolescent with breast hypertrophy suffer from significant emotional distress [16]. This occurs with significant social pressure to fit in. Embarrassment, dissatisfaction with body image, poor self-esteem, and disorder eating habit may be observed in JHB [16,17]. Unfortunately, adolescents often internalize these stressors and reluctant to seek appropriate help. Therefore, appropriate investigation and proper management at an early stage of the disease are very important to improve the quality of life.

The treatment modalities in JHB involve the following four strategies: (1) surgical management, (2) medical therapy administered either preoperatively or (3) postoperatively, and (4) medical therapy alone [5,7]. In this case, mammoplasty reduction of right breast reduction was successfully performed. The design was made prior to the operation, with the choice of the T-inverted design due to extensive mass resection [14]. Nipple areola complex sparing was not possible to be done because of the unclearly defined border of nipple-areola complex from surrounding tissue and due to reduction of the breast parenchyma. Delayed right nipple reconstruction was planned after the size of the breast was stabilized and no recurrence occurred.

During the surgery, parenchymal tissue was found bleed easily, then tourniquet binding was performed using intestinal clamps to reduce bleeding. The result of the operation was round shape right breast with 15 cm in diameter and inverted T-shaped surgical wound. The patient was discharged on postoperative day 5. Post-operative wound day 14 was good without remarkable complications.

Reduction mammoplasty is well accepted as a first-line treatment for symptomatic macromastia [2]. It successfully alleviates discomfort and helps eliminate skin break down and infection commonly associated with breast hypertrophy, which positively influence physical and psychological outcomes. Studies have demonstrated that reduction mammoplasty markedly enhance self-perceived body image [18,19].

Both surgical and medical treatments have been attempted to manage juvenile breast hypertrophy. Timely management of this condition is the main priority. This means a carefully planned surgery following a period of observation to confirm breast growth stabilization is recommended. The surgical option for this condition includes reduction mammoplasty with or without free nipple graft, or in extreme cases, subcutaneous mastectomy, and breast reconstruction in an immediate or delayed fashion. Untimely surgery in an active growth phase has been associated with a high recurrence of breast enlargement [2]. In giant juvenile fibroadenoma may recur after complete excision and the chance of recurrence becomes less after the third decade [20].

Medical therapy such as hormone modulators has been attempted in the treatment of this disease, such as tamoxifen, danazol, or bromocriptine [21]. However, the evidence of the efficacy and safety in long-term use of pharmacotherapy in juvenile breast hypertrophy is currently unknown. The well-known side effect of tamoxifen includes the risk of endometrial cancer, thromboembolism, host flashes, and decrease in bone density [2,5]. Our patient demonstrated a stable disease following the operation and was not commenced on any medical therapy.

CONCLUSIONS

Juvenile hypertrophy of the breast (JHB) is a rare and benign condition where breast tissue grows rapidly and massively that can lead to gigantomastia in peripuberty. Reduction mammoplasty is well accepted by adolescents with juvenile breast hypertrophy and can improve physical and psychological outcomes.

DECLARATIONS

Competing of Interest
The authors declare no competing interest in this study.

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