Bilateral Gigantomastia with Congenital Gum Hypertrophy in a 12 year-old - Girl

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Abstract

Title: Bilateral Gigantomastia with Congenital Gum Hypertrophy in a 12 year-old - girl

Report of the case: In this case report we present a unique finding in a 12-year-old-girl with juvenile gigantomastia of the bilateral breast (rapid and disproportionate increase in the size of the breast during puberty in 1 year, with secondary skin ulcerations) and congenital hypertrophy of gum. Her BMI was 20.6 kg/m². There was no auxiliary lymphadenopathy. Hormonal levels of Prolactin, follicle-stimulating hormone, and serum estradiol were normal. The medical genetic analysis report did not associate with any syndrome. Radiological imaging of the breast did not report anything suggestive of Neoplastic. Reduction mammoplasty was done and debulking of gums would be planned. After 4 months of follow-up, no recurrence is reported. After an extensive literature search, we could not find any similar case reported.

Discussion: Gigantomasatia is a rare case where continued breast growth occurs during puberty. The exact underlying aetiology for Gigantomastia has not been fully elucidated, but several theories have been proposed. The treatment modalities in Gigastomastia involved the following strategies: (1) surgical management, (2) medical therapy administered either preoperatively (3) postoperatively and (4) medical therapy alone.

Conclusion: Reduction mammoplasty is well accepted by adolescents for Gigantomastia which can improve physical and psychological outcomes. Gigantomastia with congenital gum hypertrophy has only been described sporadically in the literature and this case will further contribute to the knowledge and future studies in establishing the optimal treatment modality for this debilitating condition.

Categories: Plastic Surgery, Psychology, Quality Improvement

Keywords: psycho-social, reduction mammaplasty, breast cancer biology, common puberty, gigantomastia

Introduction

Introduction

Juvenile gigantomastia is a benign condition manifesting as an atypical, rapid, and continued increase in either unilateral or bilateral breast size, disproportionate to other body parts during puberty [1]. There are a myriad of terms describing this entity in the literature including virginal hypertrophy, juvenile gigantomastia, and juvenile macromastia [2].

It is a relatively rare condition. Neinstein reviewed 15 publications regarding breast lesions in adolescents spanning a period of nearly 40 years and reported that gigantomastia accounts for only 2% of all breast lesions in this group of patients [3]. In 2011 Hoppe et al. in their meta-analysis of case reports identified 65 reported cases between 1910 and 2009 [4]. Hisham et al. The study yielded additional nine cases from 2010-2017 [5]. Our own literature search to the best of our knowledge yielded additional 10 cases from 2017 till date, but in our study, this is the first case where bilateral gianatomastia with congenital hypertrophy of gum was reported.

The etiology of gigantomastia has not been fully explained, but several theories have been proposed. The popular theory includes the imbalance of endogenous hormone production [6]. Some suggest that it may be a consequence of an increasingly unhealthy lifestyle, a hormone-laden diet and obesity [7].
The development of gigantomastia 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. The management of this disease includes hormonal interventions, surgery, and a combination to prevent recurrence [8].

Here we reported a case of a 12-year-old girl with bilateral Gigantomastia with Congenital Gum hypertrophy underwent bilateral breast reduction mammoplasty.

Case Presentation

Case Report

A 12-year-old girl was referred to plastic surgery because of progressive, massive, bilateral breast enlargement for a period of 1 year and enlargement of gums, along with ulcers in the lower quadrant of both breasts near inframammary folds with severe back and neck discomfort, incapacitating her from school and social activities, and causing her social embarrassment. She did not attain menarche till now. Her past medical and family history was unremarkable, and she was not receiving any medication. Preoperative pictures are in Figure 1.

FIGURE 1: Bilateral enlargement of breast and Congenital Gum Hypertrophy

Right side - Bilateral Enlargement of breast

Left Side - Congenital Gum Hypertrophy

On examination, she was a slim girl with a normal BMI of 20.6 kg/m2 with body weight 40 kg. The breast was asymmetrical, pendulous and disproportionately enlarged with inverted areolas. The right breast lump measure 24cm x 20cm and left breast lump measured 19cm x 15cm. Both the lumps were solitary, well circumscribed, and not fixed to the deeper structures. The right 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 investigation within normal limit. Hormonal levels of Prolactin, follicle-stimulating hormone and serum oestradiol are within normal limit. Medical genetic analysis report suggested no syndromic association. Ultrasound of the breast showed interstitial oedema. Magnetic resonance imaging of the breast revealed bilateral gigantomastia - Ulceration on right breast skin surface with right intramammary lymphadenopathy with Multiple diffuse cystic channels are seen involving all quadrants of breast with proliferation of fibro glandular tissue in both breasts.

A standard Wise-pattern skin resection was designed and bilateral breast reduction was performed with supromedial pedicle flap. A total of 4900 grams of tissue had been resected, accounting for 12.25% of patient
total body weight (Figure 2). The resultant defect was closed in an inverted-T scar fashioned closure. No intraoperative blood transfusion was needed.

**FIGURE 2: Gross Specimen and Histopathology**

Right side - Gross Specimen

Left side - histopathology

HPE report after operation was bilateral Juvenile hypertrophy of bilateral breast. Postoperative period was uneventful, and the patient was discharged on day 10 after the operation. Figure 3

**FIGURE 3: Post Operative Images**

Right side - Post-operative Day -10

Left Side - follow up after 4 months

**Discussion**

Discussion

Gigantomastia is a rare case where continued breast growth occurs during puberty. The differential diagnosis includes giant fibroadenomas, phylloides tumor, and malignant tumor such as lymphoma and sarcomas. Although malignant tumour 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 [8]. 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.

The exact underlying aetiology for Gigantomastia has not been fully elucidated, but several theories have been proposed. The popular theories include end-organ hypersensitivity to normal levels of circulating oestrogen increased oestrogen or progesterone receptor expression, imbalance of endogenous hormone production, and excessive local oestrogen production [9].

Hereditary and autoimmune causes have also been described but in most cases the condition is sporadic. More recently, genetic basis for this disease has also been postulated involving the PTEN (phosphatase and tensin homologue) tumour-suppressing gene. In 2002, Li et al. on a murine model discovered that mutation
and deletion of the PTEN gene has been linked to precocious lobuloalveolar development, excessive ductal branching, delayed involution, reduced apoptosis and mammary epithelial hyper proliferation [10]. However clinical correlation is still unclear. Two case reports that performed the PTEN gene mutation analysis on their pathologic samples were found to be negative [11]. Our patient had neither the family history nor association with any autoimmune diseases.

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. In this case the breast tissue grows rapidly and massively for over 1 year. 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 [12]. 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 Gignastomastia [13].

The treatment modalities in Gignastomastia involved the following four strategies: (1) surgical management, (2) medical therapy administered either preoperatively (3) postoperatively and (4) medical therapy alone [6]. In this case reduction mammoplasty over of both breast 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].

It successfully alleviates discomfort and helps eliminate skin break down and infection commonly associated with breast enlargemen which positively influence physical and psychological outcomes. Studies have demonstrated that reduction mammoplasty markedly enhance self-perceived body image [15]. Both surgical and medical treatments have been attempted to manage Gignastomastia . 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.

Medical therapy such as hormone modulators has been attempted in the treatment of this disease, such as tamoxifen, danazol, or bromocriptine [16]. However, the evidence of the efficacy and safety in long-term use of pharmacotherapy is currently unknown.

Conclusions

Juvenile Gigantomastia is a rare and benign condition where breast tissue grows rapidly and massively in peripuberty. Reduction mammoplasty is well accepted by adolescents for Gigantomastia which can improve physical and psychological outcomes. Gigantomastia with congenital gum hypertrophy has only been described sporadically in the literature and this case will further contribute to the knowledge and future studies in establishing the optimal treatment modality for this debilitating condition.

Additional Information

Disclosures

Human subjects: Consent was obtained or waived by all participants in this study. Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work. Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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