

# Virginal breast hypertrophy: A case report

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## Abstract

A 14-year-old girl presented with the rapid growth of bilateral breasts within a year, causing her chest discomfort, and pressure sores in the inframammary fold. Bilateral reduction mammoplasty with free nipple-areola graft was performed. Nine kilograms of breast tissue, accounting for 15% of her total body weight, were excised.

## KEYWORDS

Adolescent, gigantomastia, mammoplasty, macromastia, puberty

## INTRODUCTION

Virginal breast hypertrophy (VBH) is a benign condition manifesting as atypical, rapid, and continued increase in either unilateral or bilateral breast size, disproportionate to other body parts during puberty.<sup>1,2</sup> Patients experience an initial period of quickened growth for three to six months. If left untreated, a longer period of slower growth may last for childbearing years.<sup>3</sup> Terms like juvenile hypertrophy, gigantomastia, and virginal or juvenile macromastia have been used interchangeably to define VBH in the literature.<sup>4</sup> This is an extremely uncommon condition arising sporadically, though a familial case has been reported.<sup>5</sup> Hoppe et al<sup>4</sup> mentioned sixty five cases reported in the literature till 2010 A.D. Later on, Hisham et al<sup>6</sup> acknowledged nine more cases in addition to Hoppe et al from 2010 to 2016 A.D. Reduction mammoplasty with its modifications, is generally advisable to decrease the mass effect of the breast causing neck and back pain and to prevent hemorrhage and ulceration in the skin. However, there are chances of recurrences.<sup>7</sup>

Virginal breast hypertrophy has not been reported from Nepal to the best of our knowledge. Herein, we describe the case of a 14-year-old girl, who was diagnosed with this rare entity and corrected surgically.

## CASE PRESENTATION

A 14-year-old female student from eastern Nepal visited our center with massive and gradually progressive enlargement of bilateral breasts for a year, along with chest discomfort, and pressure sores in the inframammary folds and shoulders due to bra-straps. Her large breast size had incapacitated her from attending school and social activities. Her menstrual cycle was regular over the past year. She was not under any medications. Past medical and family history was not significant.

On examination, her bilateral breasts were pendulous and enlarged disproportionately to other body parts, with widened areola, and multiple areas of pressure necrosis over the skin (Figure 1). Breasts were non-tender and firm on palpation, without any discrete masses. Axillary lymph nodes were not enlarged. She had normal body mass index (BMI) of 23.15 kg/m<sup>2</sup> (weight=60 kg and height=161 cm). Complete blood counts, C-reactive protein, thyroid function test, follicle-stimulating hormone, luteinizing hormone, estradiol, progesterone, and prolactin were within normal limits.

Bilateral reduction mammoplasty with free nipple-areola graft was performed. Markings for neo-nipple areolar complex (neo-NAC) on both breasts were done at 23 cm from the suprasternal notch on breast meridian, with neo-NAC to inframammary fold (IMF) length of 8.5 cm (Figure 2). Full-thickness NAC was excised bilaterally. Skin and parenchymal resection of medial and lateral wedges of tissues and whole breast tissue inferior to the neo-NAC were excised till the fascia, which revealed encapsulated solid breast tissue. Specimens consisting of skin and breast tissue weighing five kilograms (kgs) and four kgs were respectively excised from the right and left breasts (Figure 3). Medial and lateral flaps were brought together and hitched at IMF. The skin over the intended neo-NAC was de-epithelized. Then, the full-thickness NAC was grafted over it and was secured with the tie over the bolster dressing. Neo-IMF and vertical limb of inverted T were sutured and the excised tissue was sent for histopathology. Histopathology revealed extensive fibrosis with collagen deposition in the mammary ducts lined by inner ductal epithelial cells and outer myoepithelial cells (Figure 4). There was focal cellular stroma without atypia. This was suggestive of pubertal macromastia without evidence of malignancy.

By the seventh day (Figure 5), her chest discomfort, neck, and back pain had subsided. During five months of follow up, she was doing fine and was satisfied with the outcome of the surgery. She improved her social interactions as well.

## DISCUSSION

Normally, breast development begins with the formation of the mammary ridge from the ectoderm on the ventral surface of the body during the sixth week of gestation.<sup>8</sup> Then, subsequent development of primary mammary buds, secondary mammary buds, and lactiferous systems occur. In the late fetal period, nipple and areola form from the mammary pits.<sup>8,9</sup>

Breast remains largely unchanged from birth to puberty though the lobular alveolar and the ductal systems grow under the strong influence of estrogen.<sup>9</sup> From puberty onwards, normal breast development occurs over two to five years involving all tissues of the breast due to the result of the anterior pituitary hormone through follicle-stimulating hormone, luteinizing hormone, growth hormone, and adrenocorticotrophic hormone along with the estrogen. Estrogen has the strongest effect on ductal growth.<sup>8,9</sup> In addition to these, progesterone and prolactin have a role in lobular-alveolar development.<sup>10</sup> Similarly, fibrous stroma and fatty tissues also increase in the puberty. However, sometimes massive or excessive growth of the unilateral or bilateral breasts can occur despite the normal level of gonadal hormones. Though the exact cause remains unknown, it is believed to be due to idiopathic end-organ hypersensitivity to the normal level of gonadal hormones.<sup>11</sup> Molecular studies regarding the VBH show that *PTEN* gene which encodes for tumor-suppressing phosphatase protein, which when mutated in the mice showed excessive ductal branching, precocious development, and severely reduced apoptosis in the mammary gland.<sup>12</sup> However, such testing is rarely done in the suspected case of VBH. Koves et al<sup>13</sup> reported the case which turned out negative for *PTEN* mutation.

With the increasing size of the breast in the initial period, the breast becomes unevenly large and pendulous with prominent superficial veins. Nipple and areola are typically stretched and may be difficult to distinguish.<sup>8</sup> The increasing size and weight of the breast may cause breast tenderness, difficulty in supporting the weight of the breast, postural problems, respiratory problems, neck pain, back pain, and sometimes decreased sensation of ulnar nerve distribution.<sup>5,8,14</sup> Due to chronic irritation of the bra straps, shoulders can be grooved and scarred.<sup>5,8</sup> Similarly, intertrigo and stretch marks can also be observed.<sup>14</sup> Our patient had chest discomfort and pressure sore in the inframammary folds and had grooved shoulders due to bra-straps. Diffuse firmness, or one or more rubbery masses on palpation may be present, and axillary lymph nodes are also not enlarged.<sup>8</sup> Such masses were not present in our case. And axillary lymph nodes were not enlarged.

As exemplified in our case, a sudden change of the body shape in comparison to peers, and unfitting clothes may lead to social embarrassment and even depression, which is a major risk that requires psycho-social care to the patient.<sup>3,5</sup> In our case, the patient had limited her social activities and stopped going to school due to the embarrassing size of her breasts.

Though cases of VBH have been mentioned in the literature, we could not find the true incidence of VBH and

it remains unknown. Being rare, the initial presentation of this entity can easily divert the surgeon to the other pathologies like fibroepithelial tumors (fibroadenoma and phyllode tumor), adolescent macromastia and fibrocystic changes which are more common than VBH in this age group.<sup>1,15</sup> Other conditions, like trauma, dermal infiltrative processes such as lymphoma and leukemia cutis, pancolitis, infection, and breast malignancies though rare, should be kept in mind before reaching the final diagnosis.<sup>2</sup> Two rare events about the primary and secondary lymphoma mimicking juvenile breast hypertrophies have been reported.<sup>16</sup> So, it is necessary to keep the differentials wide.

There are no specific treatment strategies. The common approaches used are either surgical, medical or both.<sup>4</sup> Medical management with hormone modulators like tamoxifen citrate, dydrogesterone and, medroxyprogesterone, either alone or post-operative can be used. But their results are variable and side effects cannot be overlooked.<sup>3,17</sup>

Surgery is the best treatment modality. Though the timing of surgery is debatable, consideration of the surgery should be made to eliminate the physical symptoms, and increase the post-operative potential. Mastectomy, usually subcutaneous with breast reconstruction, or breast reduction surgery with its modifications, free nipple graft, or pedicle-based technique, is generally preferred.<sup>4</sup> There is decreased chance of recurrence in mastectomy than in reduction mammoplasty, indicating complete removal of breast tissue.<sup>4</sup> However, in adolescents, psychological consequences after the procedure cannot be overlooked.

Free nipple graft reduction mammoplasty can be done in selective patients. Patients who need large volume reduction (resected tissue greater than two kgs on each side), as in our case, or sternal notch to nipple distance greater than 40 cm, is generally advocated for breast reduction surgery with free nipple graft technique.<sup>18</sup> Additionally, elderly patients, obese patients with gigantomastia, smokers, patients with peripheral vascular disease, or any systemic disease with impaired healing are also advised for reduction mammoplasty with free nipple graft.<sup>18</sup> Pedicle-based technique utilizes long fold of pedicle and can compromise the blood supply during large volume resection and lead to increased risk of nipple necrosis again favoring free nipple graft technique.<sup>14,18,19</sup> Similarly, studies have shown pedicle based technique has increased complications than free nipple graft.<sup>19</sup> Also, breast reduction surgery with free nipple graft technique has decreased the chance of recurrence.<sup>6,20</sup> And can be done rapidly with minimum blood loss and skin undermining.

Though free nipple graft reduction mammoplasty is preferred in massive gigantomastia (mass resection greater than two Kgs per side), it has the disadvantage of resulting in the loss of lactation, variable return of sensation and contractility of the nipple, graft failure, and at times pigmentary changes of the nipple-areola complex.<sup>18,21,22</sup> With careful deliberation in the department, proper counseling of the patient and her family members and adhering to the limited evidence; bilateral breast reduction surgery with free nipple-areola graft was performed in our case.

## CONCLUSION

VBH is a rare presentation in pubertal period requiring the consideration after ruling out wide differentials. Its management is often individualized by considering various factors such as massive volume reduction. Reduction mammoplasty with the free nipple graft is the better option for those requiring massive volume reduction and it would be helpful to counsel the patient and his/her family members about the limitations of this procedure.

## List of Abbreviations

BMI: Body mass index

IMF: Inframammary fold

Kgs: Kilograms

NAC: Nipple areola complex

VBH: Virginal beast hypertrophy

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## Declaration

### Ethics approval and consent to participate

Need for ethical approval waived. Consent from the patient and her father deemed to be enough.

### Consent for publication

Written informed consent was obtained from the patient's father for publication of this case report and any accompanying images. A copy of the written consent is available for review by the editor-in-chief of this journal.

### Availability of data and material

Not applicable

### Conflict of interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

### Author contributions

BP, SG, NBP, and NB: wrote the initial draft of the manuscript. JS, SR, and SS reviewed the manuscript. BP, SG, NBP, and NB: edited the draft and reshaped it into this manuscript. All authors approved the final version of the manuscript and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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## Captions of figures

Figure 1: The anterior (A) and lateral (B) views of the body showing breast hypertrophy just before surgery.

Figure 2: Markings of neo-NAC on breast meridian, 23 cm from the suprasternal notch, and neo-NAC to inframammary fold length of 8.5 cm.

Figure 3: Removed breast tissues.

Figure 4: Panel A (hematoxylin and eosin, x40), left breast specimen showing a section of the mammary duct with extensive collagen deposited areas; Panel B (hematoxylin and eosin, x100), magnified view of mammary duct showing normal appearing inner ductal and outer myoepithelial cells without any atypia, periductal stromal fibrosis.

Figure 5: Condition of breasts during the seventh postoperative day.









