

Massive Assymetrical Virginal Breast Hypertrophy: A Case Report

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Virginal breast hypertrophy (VHB) is a rare , distinct disorder of unknown etiology with the rapid onset of macromastia at the onset of puberty. We reported a 12 year old, peripubertal girl presented to us with abnormal assymetrical growth of her breasts in 10 months. Due to the enormous breast volume, which caused her physical and psychological problems, she curtailed her social life. On examination, left breast was enlarged more in comparison to right ,with associated skin changes. Endocrinological investigations were normal. A bilateral reduction mammoplasty with free nipple graft was performed. Histological analysis of the breast tissue revealed the diagnosis of virginal hypertrophy. During the follow-up period of 13 months, no recurrence was noted and patient is physically and psychologically satisfied.

Introduction

Virginal hypertrophy of the breast (VHB) is an uncommon, benign disorder and typically occurs in peri-pubertal females¹⁻⁵. This entity was first described by Durston in 1669. The etiology of VHB is uncertain¹⁻⁵. VHB usually develops sporadically, but familial cases have also been reported^{1,6}. It occurs more commonly in girls between 8 and 16 years of age, and is clinically characterized by rapid enlargement of the breast¹⁻³. The overgrowth of the breasts is usually bilateral, although unilateral VHB has been described^{4,5}. It can cause several clinical problems such as breast pain, back and neck pain, dilatation of superficial veins, and skin ulcerations. It may also cause some serious psychological and cosmetic disturbances. We present a case of 12 year old female having assymetrical bilateral VHB with left breast larger than right and reaching upto left groin crease. Associated psychological and social morbidity is also discussed in brief.

Case Present



A healthy, postmenarchal 12-year-old girl was seen in our Plastic surgery OPD with chief complaint of enlargement of both the breasts since last ten months. This massive breast enlargement created multiple socio-cultural problems for her along with the medical complains. Constant nagging by peers and response of general public to her appearance made her home bound. The patient dropped out of school and curtailed her social appearance to bare minimum. She also had difficulty fitting clothes of any size and at presentation to the OPD said that she even contemplated suicide but could not muster enough courage. In addition to the breast enlargement, she had bilateral mastalgia and pain in shoulders.

The growth was more enormous and rapid in left breast. Her past medical history was unremarkable and she was not on any medications or oral contraceptive pills. General survey revealed, a girl with thin built and average state of nutrition having slumped shoulders and sagging posture. On local examination, the left breast was markedly enlarged, reaching upto left groin crease. Nipples and areola were flattened and stretched. She developed a pressure ulcer of size 4 X 3 cm over inframammary crease on left side. The right breast was

moderately enlarged (Fig. 1 and Fig 2). The skin over the left breast was hyperemic, tender and warm. Palpation of the left breast revealed firm, poorly defined masses (4-7cm in diameter), whereas right breast had a uniformly firm texture without any discrete mass. No axillary nodes were noted. Other secondary sexual characters were normal. FNAC from both the breasts was suggestive of virginal hypertrophy. Hormonal studies revealed no elevated estrogens or hypothyroidism. Preoperative ultrasonography suggested multiple hyperechoic mass lesions on right side and no parenchymal abnormality on left side.

Surgical reduction with free-nipple graft was performed. The weight of the removed mammary tissue was 2510gms and 1010 gms, respectively, for the left and the right breast. The breast was reduced by combining technique of superior pedicle reduction with partial breast amputation. Nipple –areola complex was applied as free graft. The patient had an uneventful recovery and was discharged on 10th post operative day. Pathological findings were characterized by hypertrophy of cellular connective tissue and ductal epithelium, with absence of circumscribing capsule. The ducts were distorted, swollen and lined with hyperplastic epithelium. At six months after surgery, the patient was satisfied psychologically and physically with good cosmetic appearance of breast. Clinical and ultrasonographic examinations have not indicated any persistent tumors.

The patient rejoined her school education after three months of surgery. She leads a normal social life now and intermingles enthusiastically with her peers. Postoperative psychiatric evaluation at 6 months, have found no signs of depressive behavior or suicidal tendency in her.

Discussion

In most girls, thelarche is usually the first sign of puberty and the usual breast development occurs during the period of 3-5 years of onset of puberty. Complex hormonal influences affect breast development. Ductal and lobular-alveolar development is mainly influenced by estrogen and progesterone respectively³⁻⁵. Juvenile hypertrophy of the breast is an uncommon disorder that occurs near the time of menarche and results in pathologic overgrowth of the breasts¹. In VHB, the overgrowth of the breasts is usually bilateral. Initially, rapid enlargement of the breast occurs for about three to six months followed by continuous but slow growth of the breast^{1,3}. In our case there was rapid growth for initial 6 months followed by slow growth in next 2 months. The breast can grow to weight as much as 13.5 kg to 22.5 kg^{4,12}. In VHB, the breasts are usually pendulous and diffusely firm, with or without any discrete mass lesions. It can cause breast pain, and back and neck pain. Dilatation of superficial veins or skin ulcerations may be present. Physical and psychological problems may develop.

The etiology of VHB is uncertain. In these individuals hormonal level and number of estrogen receptors are normal^{1,5}. End organ hypersensitivity is assumed to be reason for massive enlargement of breasts^{1,3,5,7}. The differential diagnosis of abnormal breast enlargement during childhood includes pseudo-gigantomastia associated with obesity, juvenile phyllodes tumor, fibroadenomas, lymphomas⁸ and sarcomas. Ultrasonographic (US) examination of the breasts is rarely useful for differential diagnosis. USG breast examination of our patient showed irregular, hyperechoic, mass lesions in the right breast, which suggested multiple giant fibroadenomas.

In our patient, the final diagnosis of VHB was made by histopathological examination. Histologically, this condition is an exaggeration of the normally developing breast. Characteristically the breast tissue shows varying degrees of stromal and ductal hyperplasia, often with dilatation and cystic degeneration of the ducts with interstitial and periductal edema. Many modalities of treatment have been recommended in VHB, including reduction mammoplasty, mastectomy with implantation of prosthesis, hormonal manipulation, and combination of surgery and medications^{1-5,9}.

Appropriate surgical intervention should be performed in late adolescence or early adulthood when breast growth rate stabilizes and ideally when no change in size is detected over the last 12 months¹⁻⁵. As in our patient, early surgical interventions are required in cases with severe mastalgia, shoulder

and back pain, pressure ulceration, and social and psychological stress associated with gigantomastia. Breast reduction surgery is usually the treatment choice. The most commonly applied procedure is reduction mammoplasty with free grafting of nipple areola complex^{3,10}. Hormonal manipulation remains controversial because of unknown long-term effects. Antiestrogen drugs such as medroxyprogesterone, dydrogesterone, and tamoxifen citrate have been shown to be useful. Several cases with VHB who were treated successfully with tamoxifen citrate have been reported in the literature and it was found to be the most effective agent for preventing recurrence^{3,10,11}. However, potential side effects of tamoxifen citrate limit its use in children.

There is huge amount of psychological distress associated with massive breast enlargement. There is not only limitation of physical activities but also embarrassment due to easily observable huge breast, which ultimately leading to social isolation, loss of love life and low self confidence. As in our case, due to social problems, girl refused to go to school and was unable to mingle with her friends. Postoperatively patient was not only physically satisfied, but there was drastic improvement in social behavior and personal attitude. We believe that VHB has profound psychological impact on the patient. Our interaction with the patient leads us to believe that psychological factors should be considered importantly in deciding the time of surgery.

Conclusion

VHB is a rare benign disorder that should be kept in mind during the differential diagnosis of abnormal breast enlargement in pubertal girls. Fibroadenomas should be considered in the differential diagnosis of VHB. Especially juvenile and giant forms of fibroadenomas are more likely to mimic VHB^{2,3}. Definitive diagnosis can be made by histopathologic examination. Surgery is only treatment for severe symptomatic cases. Role of hormonal therapy in young girl is controversial. Problem of associated psychological stress should not be underestimated. Optimal treatment strategy should be based on the patient's clinical and psychological status.

References

1. Kupfer D, Dingman D, Broadbent R. Juvenile breast hypertrophy: report of a familial pattern and review of the literature. *Plast Reconstr Surg* 1992; 90: 303-9.
2. Bauer BS, Jones KM, Talbot CW. Mammary masses in the adolescent female. *Surg Gynecol Obstet* 1987; 165: 63-5.
3. Baker SB, Burke BA, Thornton P, et al. Juvenile gigantomastia: presentation of four cases and review of the literature. *Ann Plast Surg* 2001; 5: 517-26.
4. Neinstein LS. Breast disease in adolescents and young women. *Pediatr Clin North Am* 1999; 46: 607-29.
5. Templeman C, Hertweck SP. Breast disorders in the pediatric and adolescent patient. *Obstet Gynecol Clin North Am* 2000; 27: 19-34.
6. Govrin-Yehudain J, Kogan L, Cohen HI, et al. Familial juvenile hypertrophy of the breast. *J Adolesc Health* 2004; 35: 151-5.
7. Jabs AD, Frantz AG, Smith-Vaniz A, et al. Mammary hypertrophy is not associated with increased estrogen receptors. *Plast Reconstr Surg* 1990; 86: 64-66.
8. Di Noto A, Pacheco BP, Vicala RE. Two cases of breast lymphoma mimicking juvenile hypertrophy. *J Pediatr Adolesc Gynecol* 1999; 12: 33-35.
9. Cardoso de Castro C, Aboudib JH, Salema R, Valladares B. Massive breast hypertrophy in a young girl. *Ann Plast Surg*. 1990 Dec;25(6):497-501.
10. Govrin-Yehudain J, Kogan L, Cohen HI, et al. Familial juvenile hypertrophy of the breast. *J Adolesc Health* 2004; 35: 151-155.
11. Kucukaydin M, Kurtoglu S, Okur H, et al. Virginal hypertrophy. Case report. *Turk J Pediatr* 1994; 36: 243-248

Tongue Entrapment in an Aluminium Milk Can: An Unusual Cause of Tongue Injury.

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Peri-oral injuries are common findings in paediatric patients; however, tongue injury following entrapment in bottles and cans is rare and has not been reported in our locality. A case of a 9-year old previously healthy female child who got her tongue tightly entrapped in an half opened aluminium milk can while in school is hereby presented. This case highlights the result of careless and often dangerous play and misadventures of children and the challenge of management. It calls for vigilance and close supervision of children by caregivers at home and at school. Early presentation, immediate intervention and treatment can prevent grave consequences.

Introduction

Peri oral injuries are common findings in paediatric patients, and they may have significant medical, dental and psychological consequences in the affected children¹. In the USA, the tongue is the second most common site of oral mucosal lesion in children and youths after the lip and this is commonly due to bites, followed by stomatitis, herpes labialis and geographic tongue². In Nigeria however, the tongue is the most common reported site of oral mucosal lesion and this is most commonly due to fall followed by road traffic accident³.

Tongue injury may also result during convulsions in children with epilepsy, intense oro - facial spasms in Leigh diseases⁴, forceful insertion of spoon into the mouth of children with febrile convulsion⁵ and from baby walker injury⁶. A complete tongue amputation during a fight had been reported⁷. It is seen more commonly in boys than girls^{2,3} within the age range of 3 months to 17years with the highest occurrence in the 0-5 years old^{2,3}. These may be due to the fact that children are more restless, exploitative and adventurous⁸, and are constantly exploring⁹. Most tongue injuries are minor injuries that can be managed conservatively³. There are few report of tongue injury from tongue entrapment in bottles^{10,11} and can¹² from the western world but we are not aware of any such report from the tropics.

We are reporting the case of a 9-year old Nigerian girl who had tongue entrapment in an old half opened condensed milk can while trying to lick 'garri' and sugar contained in the can.

Case Report:

O. J., a 9-year old Nigerian girl presented to the paediatric emergency unit of our hospital with history of tongue entrapment in an old aluminium milk can 20 minutes before presentation while licking garri (A staple Nigerian food) and sugar contained in it while at school. There was associated pain, drooling of saliva and minimal blood loss. There was no difficulty in breathing. There was neither a previous history of similar incidence nor history suggestive of mental derangements. She had no history of previous hospital admission, surgery or blood transfusion. She was the only child of deceased parents, and lives with her maternal grandmother. Review of systems revealed no abnormality.

Examination showed a healthy school girl in obvious painful distress, sweating profusely and drooling saliva. Vital signs were within normal limits. The anterior 1/3 of the tongue was trapped in a half opened milk can (Figure 1). It was massively edematous and cyanotic. There was minimal bleeding. The ear, nose, oro-pharynx and other systems were essentially normal.

The tongue release was done under sedation with intravenous diazepam in the emergency paediatric department, using a strong Mayo's scissors to cut open the can. Fragments of the can were removed and the oral cavity copiously irrigated with normal saline (Figure 2).

Post procedure findings were minimal abrasions on the dorsum tongue at the junction of the anterior and middle one-third. The anterior one-third of the tongue was massively edematous. This however returned to normal size within one and a half hour (Figure 3) which began to resolve quickly. She was given 750 i.u of anti-tetanus serum after a test dose, 0.5ml of tetanus toxoid, amoxicillin- clavulanic acid antibiotics, analgesic and regular warm salt water gargle.

She was subsequently discharged the same day to the ear, nose and throat clinic for follow up in the outpatient clinic after several hours of observation but was lost to follow up.



Figure 1. Tongue was trapped in a half opened milk can



Figure 2. Post procedure findings showing minimal abrasions on the dorsum tongue at the junction of the anterior and middle one-third. The anterior one-third of the tongue was massively edematous.



Figure 3. Normal sized Tongue within one and a half hours after release.

Discussion

Injury commonly occur in children due to the fact that they are more restless, exploitative and adventurous and are constantly exploring^{8,9}. These injuries commonly follows falls from heights, febrile convulsions, burns and electrical injuries, drowning and near drowning and usually affect major body parts^{3,5,13} and occasionally the tongue. Tongue injury due to entrapment had been reported as isolated cases mainly in the United State of America^{10,11,12}. This 9 year old girl presented with tongue entrapment in an-old half opened milk can while trying to lick garri (a powdered Nigerian staple cassava food) contained in it while at school.

In injury to the tongue, the most common location is the dorsum of the anterior 1/3^{2,14}. This is also the site in this index case. The age of our patient is also within the commonly affected age group involved in oro- facial tissue injury^{2,3}. The tongue on presentation was massively edematous because of impaired venous return due to constriction by the edge of the half opened can. After removal of the can and copious irrigation, the edema resolved within one and a half hour (fig. 3). The challenge in the management of this case is that metal cutting facilities are not part of the regular surgical armamentarium and repeated attempts were made with different instruments to cut open the can. This type of problem is well illustrated in a tongue entrapped in a bottle in which a professional glazier was involved in the management¹¹. This may lead to delay in the intervention unless an alternative is readily available. Early presentation and immediate intervention are important. This was the case of this patient who presented within 30 minutes of the incidence and this prevented the complications that could follow prolonged entrapment which include ecchymosis, lingual edema, ischaemia, gangrene and auto amputation^{7,10,15}.

Conclusion

Tongue entrapment though rare, is a cause of peri-oral injury which is preventable¹ but not without grave consequences. However, early presentation, immediate intervention and treatment can prevent these. This case highlights the result of careless and often dangerous play and misadventures of children and the challenge of management. It calls for vigilance and close supervision of children by caregivers at home and at school.

References

1. Rothman DL. Pediatric orofacial injuries. J Calif Dent Assoc. 1996; 24(3): 37-42.
2. Shulman JD. Prevalence of oral mucosal lesions in children and youths in the USA. Int J Paediatr Dent. 2005; 15(2): 89-97.
3. Bankole OO, Fasola AO, Denloye OO. Oro-facial soft tissue injuries in Nigerian children: a five-year review. Afr J Med Med Sci. 2004; 33(2): 93-7.
4. Diab M. Self-inflicted orodental injury in a child with Leigh disease. Int J Paediatr Dent. 2004; 14(1): 73-7.
5. Ndukwe KC, Folayan MO, Ugboko VI, Elusiyan JB, Laja OO Orofacial injuries associated with prehospital management of febrile convulsion in Nigerian children. Dent Traumatol. 2007; 23(2): 72-5.
6. Al-Nouri L, Al-Isami S. Baby walker injuries. Ann Trop Paediatr. 2006; 26(1): 67-71.
7. Toure S, Fall I, Diallo BK, Diouf R, Sane JC, Diouf M, Neissem B, Diop R, Diop EH [Emergency reimplantation of the tongue after complete traumatic amputation]. Rev Stomatol Chir Maxillofac. 2003; 104(1): 52-4.
8. Lather M, Borchard S, St-vil D, et al. Falls from heights among children: A retrospective review. J. Paediatric Surg 1999; 34: 1060-3.
9. Broz L, Kripner J, Brucek S. Emergency care of severe burn children (an experience of Pragua burn center). Acta Chir Plas 1995; 37: 89-93.
10. Green DC. Bottleneck entrapment of the tongue. Otolaryngol Head Neck Surg. 1995; 113(4): 508-9.
11. Guha SJ, Catz ND Lingual ischemia following tongue entrapment in a glass bottle. J Emerg Med. 1997; 15(5): 637-8.

12. Bank DE, Diaz L, Behrman DA, Delaney J, Bizzocco S. Tongue entrapment in an aluminum juice can. *Pediatr Emerg Care.* 2004; 20(4): 242-3.
13. Shimoyama T, Kaneko T, Nasu D, Suzuki T, Horie N. A case of an electrical burn in the oral cavity of an adult. *J Oral Sci.* 1999; 41(3): 127-8.
14. Lamell CW, Fraone G, Casamassimo PS, Wilson S. Presenting characteristics and treatment outcomes for tongue lacerations in children. *Pediatr Dent.* 1999; 21(1): 34-8.
15. Singh K. Partial glossectomy for lingual edema following injury. *Indian Pediatr.* 2004; 41(5): 520.