CASE REPORT

Multimodality approach in the diagnosis and management of bilateral giant juvenile breast fibroadenoma

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SUMMARY

Juvenile giant fibroadenoma is a very rare breast disease affecting young girls of premenarche and adolescent ages. It is a benign fibroepithelial tumour characterised by stromal and epithelial proliferation that causes rapidly growing breast mass. Bilateral symmetrical involvement is extremely rare. In this article, we describe this entity in a girl aged 13 years who presented with bilateral gigantically enlarged breasts. Ultrasonography and MRI showed large, multilobulated masses involving both breasts entirely. Endovascular embolisation of bilateral internal mammary arteries and lateral thoracic arteries supplying the masses was performed prior to surgery to reduce their vascularity. The patient subsequently underwent excision of bilateral breast masses and reduction mammoplasty. Histopathologically, bilateral breast masses were confirmed to be juvenile fibroadenomas.

BACKGROUND

Girls of prepubertal or adolescent age group presenting with a palpable mass in the breast is quite rare.¹ Even rarer is massive or gigantic rapid enlargement

of unilateral or bilateral breasts. Juvenile giant fibroadenoma causes such a clinical presentation. Ultrasonography and MRI help to characterise these breast masses better. Imaging guidance is also useful for an accurate fine needle aspiration or core needle biopsy to differentiate this mass as benign or malignant. This facilitates better surgical management. We could not find the MRI and digital subtraction angiographic (DSA) features of this entity in the literature until now. Endovascular embolisation of these huge masses is useful preoperatively to reduce the tumour vascularity which aids in easier surgical excision.

CASE PRESENTATION

A girl aged 13 years presented with massively enlarged bilateral breasts with severe discomfort for 8 months. Both breasts grew rapidly to the present size starting soon after menarche. She had no history of trauma, fever, discharge, loss of weight or loss of appetite and no family history of breast malignancy. On clinical examination, gigantic enlargement of bilateral breasts was seen extending downward until bilateral iliac regions with multiple dilated superficial veins (figure 1A). Skin over both

Figure 1 (A) Clinical photograph showing gigantic enlargement of bilateral breasts reaching up to bilateral iliac regions, (B) sonography with high-frequency transducer showing circumscribed hypoechoic mass component (arrow), (C) colour flow imaging showing few dilated vessels (arrowhead) within the mass and (D) pulse Doppler showing low resistance waveform within the arteries.

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breasts showed areas of redness and hyperpigmentation. Bilateral nipple–areola complexes were stretched out by the masses. There was no regional lymphadenopathy.

INVESTIGATIONS

Sonography findings

Well-circumscribed, multilobulated masses, with predominantly hypoechoic components were seen occupying both breasts (figure 1B). Few irregular cystic areas were also seen in between the hypoechoic components. There were no calcifications. Few dilated intervening vessels were seen in the mass that showed low resistance waveform on Doppler (figure 1C, D).

MRI findings

The enlarged breasts were too large for the breast coil, so a routine magnetic resonance (MR) mammography could not be performed. A torso coil was used to cover the entire breasts spanning the chest and abdomen. Multilobulated heterogeneous masses were seen involving the entire breasts bilaterally; right side measuring 39×22×17 cm (craniocaudal×transverse×anteroposterior) and left side measuring 40×24×18 cm. They appeared predominantly hypointense to isointense when compared with muscle on T1-weighted images (figure 2A); mildly hyperintense to muscle on T2-weighted images (figure 2B). Postcontrast fat-suppressed T1-weighted images showed relatively homogeneously enhancing mass components, separated by intervening non-enhancing septae (figure 2C, D). The masses were abutting the pectoral muscles bilaterally; however, no infiltration was seen. Time-resolved MR angiogram showed multiple arterial feeders from the branches of the subclavian artery—mainly the lateral-thoracic artery and internal mammary artery and their branches

(figure 2E). The lesions were moderately vascular, enhancing in the early delayed phase. Dilated venous channels were seen within the lesion, draining into the subclavian and jugular veins. Few dilated veins were seen in the suprasternal notch also. A radiological diagnosis of bilateral juvenile giant fibroadenoma was made.

Fine needle aspiration cytology

Smear showed diffusely cellular stroma, sheets of epithelial cells and multiple bare nuclei with no atypia. These features were representative of benign fibroepithelial tumour.

DIFFERENTIAL DIAGNOSIS

The clinical differential diagnoses clinically are juvenile (virginal) breast hypertrophy and phyllodes tumour. Juvenile hypertrophy causes massive diffuse enlargement of both breasts with no distinct mass within. This was ruled out by sonography and MRI. Phyllodes tumour usually affects unilateral breast and is much more common in adults (quite unusual in prepubertal age or adolescence).² However, excision biopsy was performed to rule out malignant phyllodes tumour.

Rapidly growing breast mass could rarely be metastasis from rhabdomyosarcoma, leukaemia, lymphoma, primitive neuroectodermal tumours, Ewing sarcoma, malignant melanoma and renal cell carcinoma.³ However, the size of such masses is not usually as gigantic as in our patient.

TREATMENT

The patient underwent endovascular embolisation for bilateral breast masses, prior to surgery, to reduce the vascularity of the large-sized masses. DSA of bilateral subclavian arteries showed the internal mammary arteries with their branches and the

Figure 2 (A) Axial T1 and (B) axial T2-weighted MR images showing multilobulated mass (white arrows) appearing isointense to muscle on T1 and mildly hyperintense (white arrows) on T2-weighted images; (C) axial and (D) coronal contrast-enhanced fat-suppressed T1 images showing enhancement within the mass (white arrow) sparing the septae (dotted white arrow). (E) MR angiography showing bilateral lateral thoracic arteries (white arrowheads) and dilated bilateral internal mammary arteries (black arrowheads) and their branches.

Figure 3 Digital subtraction angiography of the left side. (A) Dilated left lateral thoracic artery and its branches (*white arrowhead*), (B) dilated branch of the left internal mammary artery (*black arrowhead*), (C) postembolisation of the left lateral thoracic artery and its branches (*white arrowhead*)—no flow within them and (D) postembolisation of the left internal mammary arterial branches (*black arrowhead*)—no flow within the branches.

lateral thoracic arteries supplying bilateral breast masses ([figure 3A, B](#) of the left breast; [figure 4A, B](#) of the right breast). The internal mammary arterial branches and lateral thoracic arterial branches of each side were superselectively catheterised and embolised using graded gel foam particles. Postembolisation angiogram showed no flow in these arteries ([figure 3C, D](#) of the left breast; [figure 4C, D](#) of the right breast).

Subsequently, the patient underwent surgery. The masses in bilateral breasts were totally excised. The mass from the right breast weighed 10 kg and from the left breast was 11 kg ([figure 5A](#)). Reduction mammoplasty was performed using free nipple grafting technique. Bilateral nipple–areolar grafts were harvested and symmetrically repositioned ([figure 5B](#)). Owing to the massive size of bilateral breast masses occupying the entire breasts, pedicle technique could not be performed.

Histopathology of bilateral breast masses showed uniform stromal hypercellularity with predominant pericanalicular pattern; tubular or oval ducts lined by epithelial and myoepithelial cells; and multiple bare, bland nuclei in the background. No atypia was seen. These features confirmed the diagnosis as fibroadenoma ([figure 5C, D](#)).

OUTCOME AND FOLLOW-UP

Postoperatively, the patient made an uneventful recovery. She was satisfied with the surgical outcome. On follow-up after 3 months, the wound site had healed well and there was no recurrence. The patient was advised to follow-up after 6 months or in case she noticed any recurrent mass.

DISCUSSION

Breast masses in children and adolescents are mostly benign in nature.⁴ Juvenile fibroadenoma is the most common among them, comprising nearly 75% of breast masses in puberty.⁵ Giant juvenile fibroadenoma, a variant of fibroadenoma that is defined as larger than 5 cm or >500 g, is very rare.⁶ This entity is reported mostly in females of East Asian, African or Caribbean origin.⁷ Bilateral symmetrical giant juvenile fibroadenomas are even rarer, with only about 16 cases until.^{8–11} This benign fibroepithelial tumour consists of stromal (connective tissue) and epithelial (ductal) proliferation.

Ultrasonography is the preferred imaging modality for initially evaluating a breast mass in a child or adolescent¹² as it is

Figure 4 Digital subtraction angiography of the right side. (A) Right lateral thoracic artery and its branches (*white arrowhead*), (B) right internal mammary artery and its branches (*black arrowhead*), (C) postembolisation of the right lateral thoracic artery and its branches (*white arrowhead*)—no flow within them and (D) postembolisation of the right internal mammary arterial branches (*black arrowhead*)—no flow within the branches supplying the mass.

Figure 5 (A) Gross resected breast mass, (B) postsurgical clinical photograph, (C) low power histological photomicrograph and (D) higher power image showing uniform stromal hypercellularity (*star*)—with predominant pericanalicular pattern, surrounding the ducts (*black arrow*), myoepithelial and epithelial cells (*black arrowhead*) without atypical features.

non-invasive and has no hazards of radiation. It helps to assess whether the lesion is solid or cystic, and to characterise most of the lesions as benign or malignant. Mammography is not useful in young patients because of the increased density of fibroglandular breast parenchyma in this age. Typical adult fibroadenomas are well circumscribed, hypoechoic, oval or round, macrolobulated (not more than three lobulations) masses with transverse dimension larger than anteroposterior.¹³ Juvenile giant fibroadenomas may not conform to most of these features. Rapidly enlarging breast mass is a suspicious feature and requires histopathology for confirmation. Fine needle aspiration cytology and core needle biopsy may help to broadly differentiate between benign and malignant aetiology of the mass, prior to surgery. However, these cannot often accurately differentiate between juvenile fibroadenoma and phyllodes tumour; and this warrants excision biopsy.¹⁴

When the mass appears suspicious for malignancy on clinical examination or on ultrasonography, MR mammography is a very useful imaging modality for further evaluation. Juvenile fibroadenoma mostly has smooth, well-defined margins. Juvenile giant fibroadenoma in bilateral breasts may appear as symmetrically large masses with heterogeneous signal intensities. On T1-weighted imaging, the mass typically appears mildly hypointense relative to fibroglandular parenchyma and hypointense to isointense when compared with muscle. On T2-weighted images, it appears hyperintense to muscle. Multilobulated masses can be seen in each breast with intervening T1 and T2 hypointense, non-enhancing septae. Postcontrast administration T1-weighted images show diffuse relatively homogeneous contrast enhancement with persistent enhancement (type 1) which is typically noted in benign masses.¹⁵ It may be due to the underlying pectoral muscles; however, it does not cause infiltration.

In extremely huge masses like in our case, endovascular tumour embolisation using DSA is beneficial to reduce the vascularity before the surgery. Angioembolisation minimises the intraoperative bleeding, improves the visualisation for resecting the tumour effectively with less chances of recurrence; and reduces the duration of the surgery.¹⁶

Giant juvenile fibroadenoma often being a very large mass at clinical presentation requires surgery as treatment unlike smaller fibroadenoma. Preserving the normal breast parenchyma and nipple–areola complex is recommended as the compressed/displaced parenchyma can fill in after the excision of the tumour and obviate the need for reconstruction.^{7 17} Although it is benign in nature, recurrences have occurred after complete excision. Reduction mammoplasty is widely used for removing large

breast masses and has less postoperative complications.¹⁸ For difficult cases of gigantic masses, mastectomy could be performed with free nipple–areolar complex grafting.^{8 19} Breast reconstruction can be performed subsequently, especially for unilateral giant fibroadenoma cases.

Learning points

- ▶ When a young girl presents with a rapidly developed, large-sized breast mass, giant juvenile fibroadenoma is to be considered as a differential.
- ▶ On ultrasonography, it can appear similar to adult fibroadenoma in some cases with well-circumscribed, oval, solid, macrolobulated, homogeneously hypoechoic appearance. It also can be heterogeneous and multilobulated, with predominantly solid component and few cystic areas within the mass.
- ▶ Ultrasound-guided fine needle aspiration cytology or core needle biopsy may provide further clue to characterise the mass as benign or malignant; however, excision biopsy is proven more accurate due to the large size of these masses.
- ▶ MRI appearance is more often homogeneously T1 isointense to muscle, T2 homogeneous or heterogeneous with predominant T2 hyperintense signals. Postcontrast homogeneous or heterogeneous enhancement may be seen. T1 and T2 hypointense non-enhancing septations may be seen within. No infiltration of the underlying muscles is seen in spite of the large size and rapid growth.
- ▶ In extremely huge masses like in our case, endovascular tumour embolisation using digital subtraction angiographic is beneficial to reduce the vascularity before the surgery as it minimises the intraoperative bleeding, improves the visualisation for resecting the tumour effectively, in less time, with less chances of recurrence.

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Contributors NR and RR made the radiological diagnosis. SJ performed the endovascular embolisation of the masses. KD performed the surgery and involved in patient follow-up. NR acquired the data and wrote the manuscript, RR edited the manuscript; KD and SJ acquired the data, reviewed and approved of the manuscript.

Competing interests None declared.

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Patient's perspective

Soon after menarche, both my breasts grew into an extremely large size within 8 months. They were so large and heavy that I was very uncomfortable to go to school or involve in any social activities; and for past few months, I was unable to do so. I also suffered from back pain on standing and walking, which reduced my daily activities even further. This made me feel sad and anxious. Nobody knew the treatment for this in my native place, and I was referred here for evaluation and treatment. After surgical removal of the masses from both breasts weighing over half my body weight, I am very relieved with the result. The biopsy also turned out to be benign. I am happy that I can resume going to school and do my routine activities.

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