

CASE REPORT

Familial congenital symmastia: Ultrastructurally abnormal breast tissue

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Abstract

Reports about congenital symmastia and its surgical treatment are few. We report two patients – a mother and daughter – with congenital symmastia in whom breast and fatty tissue was found to be mobile adhering poorly to the chest wall. Although histological examination showed no abnormality of the tissue bridge between the breasts, ultrastructural investigation of breast tissue (including Cooper's ligaments) showed an abnormal arrangement of collagen fibres. Satisfying aesthetic results were achieved by resection of excess soft tissue in the cleavage area through a submammary incision and fixation of the skin with subcutaneous interrupted sutures to the sternal periosteum.

Key Words: *Symmastia, congenital, familial, surgical correction, submammary approach, Cooper's ligaments*

Introduction

Spence et al. [1] in 1984 described a rare form of breast malformation characterised by the presence of a tissue bridge between the two breasts, with a resulting loss or reduction of cleavage between the breasts. As a consequence, the breasts seemed to blend with each other into one structural unit ("bread-loafing"). The authors called this condition symmastia. Factors leading to congenital symmastia are not clear. Spence et al. [1] described two patients, one of which was corrected by raising a flap and an inverted V to Y plasty, and the other by a modified reduction mammoplasty. Salgado and Mardini [2] in 2004 described liposuction through the periareolar approach for its treatment.

We describe here our approach to two even more unusual cases of symmastia, as the two patients were mother and daughter. We present also the results of electron microscopic investigation of the resected

tissue which showed abnormal arrangement of collagen fibres, although histological examination showed no abnormality. This suggests that the condition may have a familial component.

Case reports

Case 1

A 17-year-old girl with symmastia underwent a corrective operation in 1978 (Figure 1a). A submammary approach was used to remove fatty tissue from the soft-tissue bridge. A year later, because of her dissatisfaction with the postoperative aesthetic results, a reduction mammoplasty according to Biesenberger's technique [3] was performed and the remaining glandular breast tissue in the cleavage was resected. At follow-up 26 years later, the patient said she was pleased with the results achieved after the second intervention (Figure 1b, c).

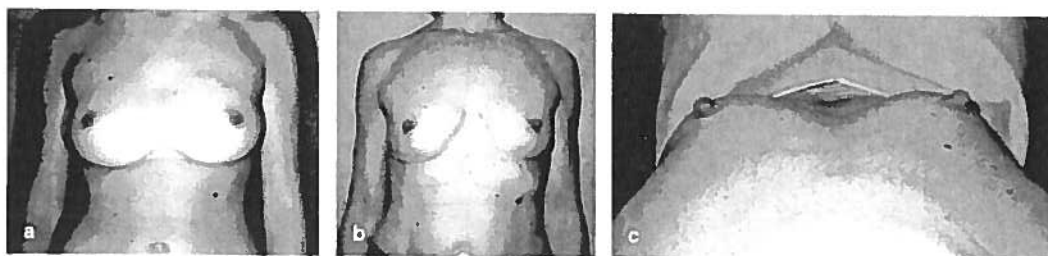


Figure 1. (a) Breast confluence before operation. (b) Postoperative result. Frontal view. (c) View from above after second surgical intervention 26 years later.

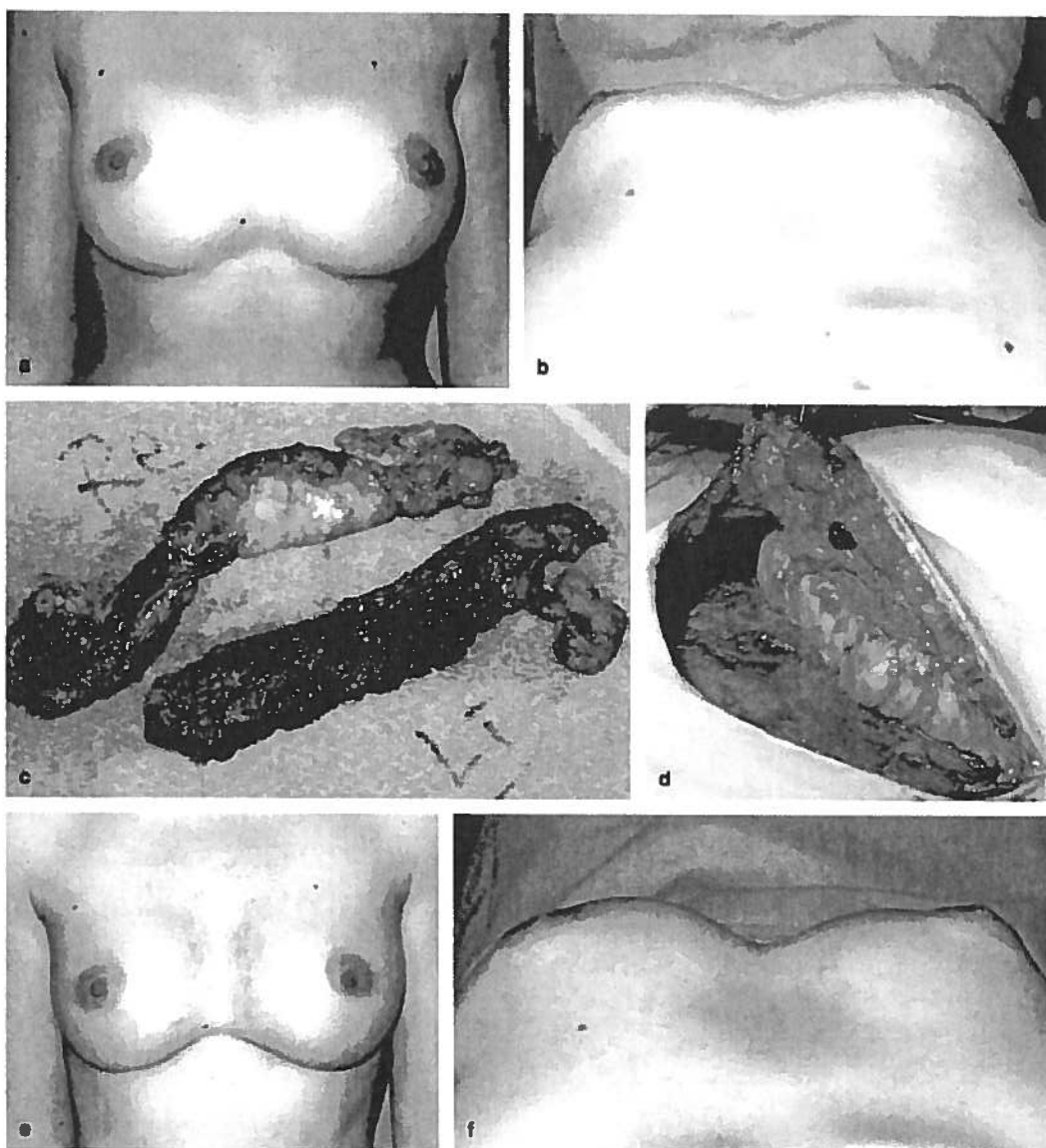


Figure 2. (a) Congenital familial symmastia in a 16-year-old girl. (b) View of the breasts from above. (c) Specimens of resected breast tissue from the cleavage area. (d) Fixation of the medial part of the left breast on to the pectoral muscle. (e) Frontal view 24 months postoperatively. (f) View from above.

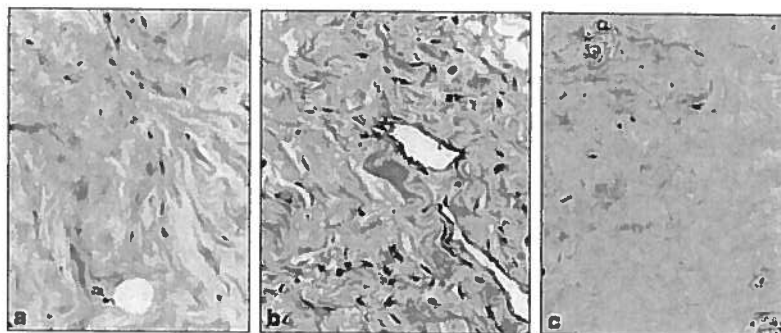


Figure 3. Photomicrographs showing no obvious difference between (a) normal breast tissue and (b, c) that from patients with symmastia (haematoxylin and eosin, original magnification $\times 400$).

Case 2

Her daughter presented to our clinic in 2002 for the first time at the age of 14 with congenital symmastia. We decided to adopt a wait and see policy because of her youth. The tissue bridge in the cleavage increased considerably during the following two years. Premenstrual breast pain, pain in the cleavage area, and severe psychological distress caused by shapeless breasts, were indications for operation (Figure 2a, b).

A submammary approach was used on both sides. We undermined a part of the lower medial breast quadrant prepectorally. Breast and fatty tissue were a mobile mass adhering poorly to the chest wall. A part of the tissue bridge measuring 11 cm \times 8 cm was removed subcutaneously (Figure 2c). The

skin was fixed to the sternal periosteum with subcutaneous interrupted sutures to help adherence of the skin and tissue to the sternum. At the medial border, breast tissue was fixed with sutures to the pectoral fascia (Figure 2d-f).

Histological examination of the resected tissue from the bridge as well as haematoxylin and eosin stains showed normal breast tissue (Figure 3a) in both patients with inconspicuous ducts, glandular structures, bland-looking loose stroma in the lobules, and collagen-rich stroma in between the lobules (Figure 3b, c). Histochemical stains for collagen bundles such as trichrome and Gordon and Sweet stain also showed no difference between breast tissue from patients with symmastia and those taken from women during reduction mam-moplasty. However, at the ultrastructural level,

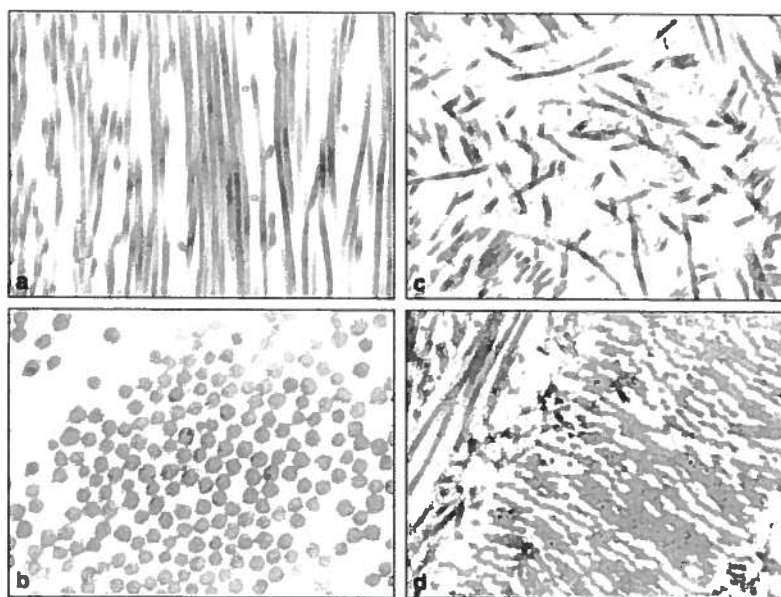


Figure 4. At ultrastructural level normal breast tissue shows a well-structured arrangement of collagen fibres (a) in longitudinal section (original magnification $\times 20,000$) and (b) in cross section (original magnification $\times 50,000$). Patients with symmastia show (c) criss-cross arrangement in longitudinal sections (original magnification $\times 20,000$) and (d) clotted fibres in cross sections (original magnification $\times 20,000$).

normal breast tissue showed a well-structured arrangement of collagen fibres in longitudinal section (Figure 4a) and in cross-section (Figure 4b), while breast tissue from the patients with symmastia showed a criss-cross arrangement in longitudinal sections (Figure 4c) and clotted fibres in cross-sections (Figure 4d).

Discussion

Although there are a few reports of congenital symmastia, we are not aware of any case report about what may be termed familial symmastia. We reported two patients, a mother and her daughter, who were both operated on for the same condition 26 years apart. Reported cases of symmastia are isolated and rare, so it is difficult to judge whether the patients presented here can be explained as similarity in a developmental defect in two related people or attributed to some underlying genetic component. It might hint perhaps at a familial disposition. Available reports contain no information or histological evaluation of resected web tissue, and there are no discussions on the possible causes of familial congenital symmastia. In our patients, the resected tissue from the bridge was evaluated histologically, but showed no abnormality. However, ultrastructural investigation showed abnormal arrangement of collagen fibres of breast tissue including Cooper's ligaments. This might be why the breast tissue crossed the midline and led to symmastia.

Congenital symmastia can be corrected by resection of the subcutaneous fatty and glandular breast tissue in the area where the cleavage should normally be. There are, however, variations of the proposed surgical techniques. Medial confluence of breasts can be corrected by reduction mammoplasty with a transverse, upside-down V to Y flap operation, lateral mobilisation of excess soft tissue to form contours, and fixing the lifted presternal skin to the sternal periosteum with interrupted subcutaneous sutures [1]. Where necessary, horizontal incision and excess skin can be resected through a horizontal incision, as described by McKissock [4]. Schönegg et al. [5] did liposuction for the excess presternal breast parenchymal tissue using McKissock's technique [6] after reduction mammoplasty by the technique of Keppke and Matta [7]. To avoid visible scars, Salgado and Mardini preferred a minimally

invasive periareolar approach [2], and subcutaneous non-absorbable interrupted sutures to attach the skin to the sternal periosteum. Visible puckering of the skin is said to disappear with decreasing tension of the suture materials over time.

We suggest resection of excess soft tissue in the cleavage area through a submammary incision, as it offers a better overview of the preparation and the resected soft tissue and helps to avoid visible scars. The suggested periareolar approach and liposuction [2] seem to be promising, and require further observation. Transdermal interrupted sutures to support adherence of skin to the sternum and the pectoral fascia are perhaps not to be recommended, because hypertrophic scars can develop from radical pull-out sutures that go deep down through several tissue layers and cause dimpling [2,8]. Instead, we recommend fixing the skin to the sternal periosteum with subcutaneous interrupted sutures. It must be emphasised that the results depend on the systematic use of postoperative compression on the skin, with individually designed and fitted bandages.

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