



Case Report

Coexistence of Gynecomastia and Poland Syndrome: Case Report

Erkan Yüce,¹ Selami Serhat Şirvan,² Işıl Akgün Demir,² Hikmet Ihsan Eren,² Semra Karşıdağ²

¹Department of Plastic Surgery, Kirikkale State Hospital, Kirikkale, Turkey

²Department of Plastic, Reconstructive, and Aesthetic Surgery, University of Health Sciences, Sisli Hamidiye Etfal Training and Research Hospital, Istanbul, Turkey

Abstract

Coexistence of the Poland syndrome and gynecomastia is a rare condition. Poland syndrome requires soft tissue augmentation of the affected side, whereas gynecomastia necessitates reduction of the breast tissue. To provide symmetry, breast reduction and fat grafting techniques should be combined.

We report a 29-year-old male patient with left gynecomastia and right sided Poland syndrome. In order to correct his asymmetry on the anterior chest wall, left breast tissue resection and fat grafting to the right breast were performed.

Having these two opposite conditions at the same time and on the same patient makes the deformities look more dramatic than they are separately. Accurate planning and selection of proper techniques enable to provide symmetry in such cases.

Keywords: Breast reduction; gynecomastia; fat graft; poland syndrome.

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Coexistence of gynecomastia and Poland Syndrome in a patient is a rare condition. In the literature, three cases have been reported in different publications with this condition,^[1–3] which remained under-researched. The two chest wall deformities, namely gynecomastia and Poland Syndrome, arise from different etiologic factors.

Poland syndrome was defined by Sir Alfred Poland about 170 years ago.^[4] The incidence of Poland syndrome has been reported as 1/30000.^[5] This syndrome usually occurs sporadically, but a familial inheritance is observed in below 1%.^[3] It is usually unilateral, affecting the right side and males more. Hypoplasia or aplasia of the pectoralis major and minor muscles, breast, nipple-areola complex, subcu-

taneous tissue and axillary hair are seen on the affected side. Abnormalities of the chest wall and upper extremities are also observed.^[4–6] To be diagnosed with Poland syndrome, the patient must have one or more other characteristics, except pectoral muscle involvement. Concomitant anomalies are often seen in other parts of the body. Poland syndrome is classified into three groups as mild, severe and very severe in Ribeiro et al.'s study (Table 1).^[7]

Gynecomastia is the most common breast disease in males and arises from the benign increase of stromal and ductal elements in the breast and incidence varies 32-36% and is reported to be 40% in autopsy series.^[8] In differential diagnosis, pseudogynecomastia, breast cancer, hypogonadism,

Address for correspondence: Işıl Akgün Demir, MD. Sağlık Bilimleri Üniversitesi Sisli Hamidiye Etfal Eğitim ve Araştırma Hastanesi, İstanbul, Turkey

Phone: +90 535 399 87 36 **E-mail:** isilakgundemir@gmail.com

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Table 1. Clinical and radiological classification of the Poland syndrome

Degree of presentation	Mammary alterations	Musculoskeletal chest alterations	Ipsilateral superior limb alterations	Other congenital alterations
First (mild)	Amastia; hypomastia or areolar asymmetry	None, or partial absence of pectoralis major muscle	None	May be present
Second (severe)	Hypomastia or amastia; areolar asymmetry	Total absence of pectoralis major muscle; different alterations of the muscles and/or bones of the ipsilateral chest	No or small alterations	May be present
Third (very severe)	Amastia; areolar asymmetry	Different manifestations	Present	May be present

hyperprolactinoma and lymphangiomas should be considered. Idiopathic gynecomastia is thought to develop as a decrease in circulating androgen, an increase in estrogen, or a physiological response to the defect in androgen receptors. Rohrich et al.^[8] classified gynecomastia in four stages in their study (Table 2).

Coexistence of these two conditions described above makes both deformities appear more dramatic and requires a complex surgical approach in the reconstruction. In this case, we report a simultaneous approach to two deformities in a patient with Poland syndrome and gynecomastia to contribute to the relevant literature.

Case Report

A 29-year-old male patient with a normal body mass index admitted to our outpatient clinic due to an asymmetric appearance in his chest. The physical examination of the patient revealed that asymmetry was because of hypertrophic breast tissue on the left side and undeveloped breast tissue on the right side (Fig. 1a, b). While congenital growth failure was seen in the right breast, it was learned that there was an overgrowth of the left breast after puberty. Significant asymmetry was observed on the vertical axis between the nipple levels. Rohrich et al.'s study showed that left-sided breast hypertrophy was evaluated as 3rd-

grade gynecomastia.^[8] Thoracic magnetic resonance imaging (MRI) was carried out with the indication of right-sided breast agenesis, suggesting Poland syndrome. The findings showed that the right pectoral muscle did not have a sternal head. No additional upper extremity and anterior chest wall anomalies were observed, and it was evaluated as mild Poland syndrome in Riberio et al.'s^[7] study. In terms of the etiology of gynecomastia, the patient was screened according to endocrinology consultation, and no hormonal disorder was detected. No mass was detected on the breast ultrasound. We learned that the patient had not had routine drug or marijuana use, and he had alcohol consumption at the level of social drinking. The gynecomastia was determined to be idiopathic.

The operation was recommended to correct the asymmetry. After obtaining the consent of the patient, left mastectomy and fat injection for the right breast were planned to be performed. With a left inferior areolar intervention, through semilunar incision line, resection of the fat and glandular tissues of the breast was performed to provide a proper contour. Tumescence solution infiltration, prepared with 1000 ml isotonic, 100 ml 1% lidocaine and 1 ml 1:1000 adrenaline, was applied to the abdominal area. Liposuction was performed with standard Coleman technique. The fat grafts were taken with an oil aspiration cannula with the diameter of 2 mm and 10 cc Luer Lock syringe. The material was separated by gravity, and a 75 cc oil graft was obtained. Fat grafts were infiltrated into subcutaneous tissue and subpectoral fascial planes to form a contour which is proper for the right pectoral area, and the operation was terminated. Because of the partial resorption of the fat grafts six months after the operation, the operation was planned again. A 125 cc fat graft obtained from the abdomen with the same technique as the previous operation was injected to ensure proper contour and symmetry (Fig. 2a, b). There were no complications after the operations, and the patient was discharged the next day. During the follow-up, an operation was recommended to correct the nipple-areola complex asymmetry; however, the patient voluntarily refused further intervention.

Table 2. Gynecomastia classification by Rohrich et al.

Grade I	Minimal hypertrophy (<250 g of breast tissue) without ptosis I A. Primarily glandular I B. Primarily fibrous
Grade II	Moderate hypertrophy (250-500 g of breast tissue) without ptosis II A. Primarily glandular II B. Primarily fibrous
Grade III	Severe hypertrophy (>500 g of breast tissue) with grade I ptosis
Grade IV	Severe hypertrophy with grade II or III ptosis

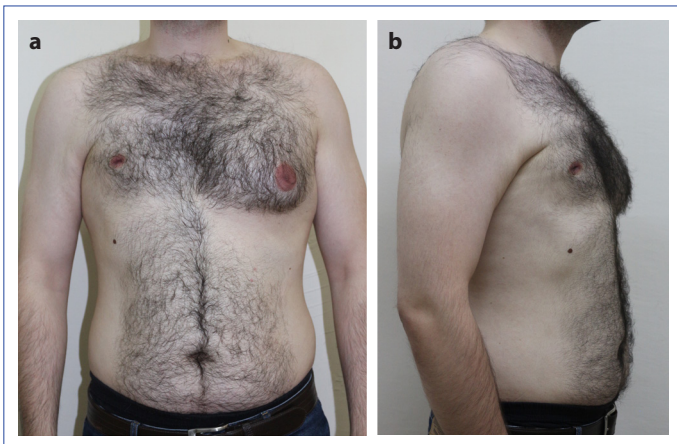


Figure 1. (a) Preoperative anterior appearance. (b) Preoperative oblique appearance.

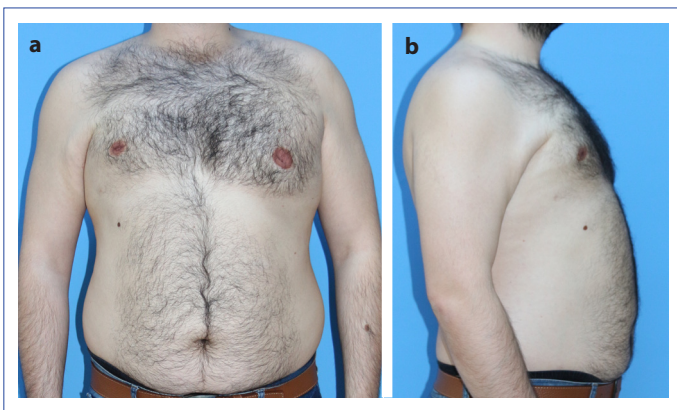


Figure 2. (a) Postoperative anterior appearance. (b) Postoperative oblique appearance.

Discussion

Gynecomastia treatment should be planned on a patient basis without following a standard protocol. The presence of excess skin, as well as the density of glandular or adipose tissue, is an important factor affecting the treatment approach. As a result of the evaluations, excision or liposuction of the breast tissue, or a combination of two methods may be preferred.^[9] While liposuction alone may be sufficient in gynecomastia with high adipose tissue, excision provides better results in cases where glandular tissue is more intense.^[8] In severe gynecomastia, there are cases where reduction is provided by methods, such as Lejour mastopexy.^[8, 9] Additionally, in patients undergoing liposuction, pull through method for residual breast tissue can be used.^[10] In our patient, mastectomy through the areolar incision was preferred because of more intense glandular tissue.

The main purpose of the reconstruction of Poland syndrome is to provide contralateral chest wall and breast contour. Contour symmetry, as well as nipple symmetry, should be considered and costal reconstruction according

to the degree of the syndrome should be included in the planning. The treatment options in male patients include implant or autologous tissue reconstruction.^[6-11] Implants can be custom designed or ready to use rectangular forms in various sizes can be used.^[12, 13] However, as in all implanted cases, the complication rate is higher than the case in reconstruction with autologous tissue. In reconstruction with autologous tissues, fat grafts, pedicled flaps or free flap options are available. Although the pedicled latissimus dorsi flap is preferred, transverse musculocutaneous gracilis flap^[14] and anterolateral thigh flap^[15] can be evaluated among other options according to the surgeon's request.

The first fat graft application was reported by Czerny in 1895. Lipomatous tissue taken from the back was used in breast augmentation.^[16] Coleman has popularized fat graft applications.^[17, 18] Fat grafting is widely used as a soft tissue augmentation method in the face, breast, gluteal region, hands and under the scars.^[17] Fat graft application is a quite ideal method regarding the possibility of complications, duration of application and ease of operation. Breast fat graft applications are increasing over time. Contrary to the discussions, breast fat graft application is reported being a safe and effective method.^[19] Complications that may occur after fat injections to the breast include palpable induration and nodule formation, deep and superficial infections, loss of sensation, pain, hematoma, sepsis, abnormal secretion from the breast, pneumothorax and fat necrosis.^[20, 21] The incidence of radiological changes after fat injection into the breast was reported to be 13% after one year, and there is no significant difference when compared to other surgical procedures of the breast.^[20] Fat injection does not cause difficulty in distinguishing a suspicious radiological change (Veber et al.).^[22] Fat injections are also frequently used in the treatment of Poland syndrome.^[6, 11, 21] As in our case, infiltration of the grafts which are obtained after separation of the fat tissue by various methods, by the liposuction of the abdominal area to achieve the contralateral chest wall and breast symmetry, provides satisfactory results. The amount of infiltration required to capture symmetry varies between 25-200 cc (Pinsolle et al.).^[21] In general, the desired symmetry can be achieved in more than one session. In our case with unilateral 3rd-grade gynecomastia, the desired contour after breast tissue resection was achieved by applying a total of 200 cc fat graft. In cases without a history of gynecomastia, the contralateral pectoral region can be directly used as a base; however, in cases similar to our case, the amount of fat injection should be determined after achieving the desired reduction in the breast with gynecomastia. While fat grafting alone can be sufficient, it can be used to support reconstruction with implant or flap and to remove minor residual contour deformities.

Disclosures

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

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Conflict of Interest: None declared.

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