Technical Refinements Utilizing the Latissimus Dorsi Muscle Flap in Male Poland Syndrome

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ABSTRACT

Poland's syndrome is a rare congenital anomaly. The incidence has been estimated at 1:16.500 live births, the ratio of men to women is 3:1 and 75% of patients are affected on the right side. It mostly, if not always, unilateral, although a single bilateral case has been reported.

It is a non genetic, congenital disorder with low (less than 1%) risk of reoccurrence in the same family. It is mostly related to impairment of the blood supply of the upper limb bud adjacent to the chest wall associated with hypoplasia of the ipsilateral subclavian artery or one of its branches.

Poland syndrome is characterized by hypoplasia of the breast, nipple and subcutaneous tissue, absence of the costosternal portion of the pectoralis major muscle, aplasia of the pectoralis minor muscle, the costal cartilages or ribs, loss of hair of the axillary and mammary region, and unilateral brachysyndactyly. Other muscles like latissimus dorsi, external oblique, and serratus anterior muscles might be affected as well. Involvements of these components are variable and it is rare for all features to be present in the same individual. In association with Poland syndrome, dextrocardia, Möbius syndrome, leukemia, non-Hodgkin's lymphoma, cervical cancer, leiosarcoma might be presented.

Management of Poland's syndrome should be guided by the degree of the deformity, the age, and the sex of the patient. Reconstruction could be achieved using autologous tissue like bone grafts and muscle flaps as latismuss dorsi to correct the muscle deficiency which has a drawback of donor site morbidities. On the other hand synthetic materials like prosthetic mesh patch, breast implants and custom made implants for male chest wall reconstruction which is expensive, need special facilities, and carries the risk of infection and extrusion could be used as well. Endoscopic assisted reconstructions for the male and female breast were used utilizing tissue expanders and custom made implants in a way trying to minimize scaring in the anterior chest wall.

INTRODUCTION

Alfred Poland, a student demonstrator in anatomy, described his case in 1841; however, there were earlier reports of this anomaly: In 1826 (Lallemand) and in 1839 (Froriep) [1-3]. The full anatomical spectrum of the disease was first sum-

marized by Thompson in 1895. In 1900, Furst suggested a common cause. In 1962, Clarkson coined the name Poland's syndactyly after more than 300 patients had been reported [4,5-7].

Surgical intervention may be indicated for the following reasons: 1- Unilateral depression of the chest wall and the possibility of its progression. 2- Lack of adequate protection of the heart and lung. 3- Paradoxical movement of the chest wall. 4- Hypoplasia or aplasia of the female breast and 5- Cosmetic defect due to lack of the pectoralis major muscle and axillary fold in male patients. In children with mild forms of Poland's syndrome, which is limited to absence of the pectoralis muscles andbreast hypoplasia, the operation should be postponed until after puberty [8]. The first report on the surgical correction of Poland's syndrome was published in 1952 by Ravitch and Handelsman [9].

Fouras et al., [10] proposed in 2003 a classification in three grades of the forms affecting the chest wall and breast. Using this clinical classification, appropriate treatment can be proposed according to the degree of involvement. The main goal is to restore muscular contour, subclavicular hollowing and absence of the anterior axillary fold, and also chest wall aplasia in severe forms.

Reconstruction thus involves several tissues requiring a variety of surgical correction techniques. These include insertion of a breast implant or customised chest wall implant [11-16], use of a pedicled latissimus dorsi flap [17-19], microsurgical transfer of a deep inferior epigastric (DIEP) flap [20], or even osteoplastic surgery of the rib cage [8,21]. Among the new surgical techniques which may be proposed in Poland's syndrome, lipostructure by Coleman's technique [22,23].

Borschel et al., [24] describe a two stage minimally invasive technique for the reconstruction of female Poland syndrome that requires a single 6-cm scar in the axilla. The first operation uses an endoscopically assisted approach for placement of a tissue expander to increase the size of the skin envelope. The second operation is then performed endoscopically to remove the tissue expander, place a permanent implant, or transfer a latissimus dorsi muscle.

Aim of our study is to describe a technique which avoids the scarring over the back and anterior chest wall, via single incision along the mid axillary line to harvest and inset latismus dorsi flap for reconstruction of male patients with Poland syndrome.

PATIENTS AND METHODS

We adopted our technique over the past five years (September 2008 till November 2013), The study was conducted in the Plastic Surgery Department, Countess of Chester Hospital, UK. and Ain Shams University Hospital, Cairo, Egypt. The study included twelve male patients with Poland syndrome, eight right side and four left side reconstructions were done. The age of patients ranged from 20 to 39 years old with an average age ranged of 29.5 years.

Preoperatively history taking, examination (the back for the latismus dorsi muscle while the patient pressing on his iliac crest) and routine investigations were carried out to all patients. The surgical procedure and the risk of complications were explained to all patients. Nine patients were concerned and asking for minimal scaring, breast symmetry and correction of the infraclavicular hollowness, while the other three patients in addition to the previous concerns were looking for reconstruction of the anterior axillary line reconstruction as well. Consent forms were signed for the surgical procedure as well as the medical photography. Photos were taken to all patients before and after the surgical reconstruction. All patients were followed-up at three, six and nine months.

Operative procedure:

Marking are made with the patient standing up, sitting down and in the supine position. It is carried immediately before surgery, marking of the healthy side starting with the midline of anterior chest wall, medial and lateral boundaries of the breast, infraclavicular hollowness, infra mammary fold and the anterior axillary fold. Mirror image marking is carried on the diseased side and the incision site

(six to eight centimeter in length) over the mid axillary line is carried out as well, see Fig. (1A,B).

The operation was performed under general anathesia. All patients were operated in supine position. Single dose of intravenous antibiotics were administered to all patients immediately after endotracheal intubation. The surgical procedure consisted of three steps: Dissection of the recipient pectoral region, harvesting of the latismuss dorsi muscle flap, insetting of the flap in the proper position.

Dissection of the recipient pectoral region:

Injection of 1:200000 adrenaline and 10ml lignocaine 1% in the site of incision and the pectoral region is carried to facilitate the dissection and minimize the blood loss. Sharp dissection was done just superficial to the pectoral fascia, using the electro-cautery and the lighted retractor in combination with blunt finger dissection as well, starting from lateral to medial and then from above downward. The boundaries of the recipient pocket were planned according to the preoperative marking.

Harvesting of the latissimus dorsi muscle flap:

Dissection is carried first superficial to the muscle from lateral to medial 1cm from the mid line of the back and from the upper border of the muscle, determined by palpating the angle of the scapula, till the lower border of the muscle just above the iliac crest. Deep to the muscle dissection is carried out between the latismuss dorsi muscle, serratous muscle medially, trapezious at the upper border and intercostals muscles at its lower portion. Cutting the muscle from its origin starts from the lower portion at the iliac crest, the medially at the mid line and lastly its upper border just inferior to the angle of the scapula. Reinserting its tendon anterior to the biseptal groove is carried in three patients asked for reconstruction of the anterior axillary line.

Insetting of the flap in the proper position:

The latissimus dorsi muscle flap is anchored to the anterior chest wall Flap with 2-0 prolene sutures (10-14 days), anchored to periosteum and secured with multiple bolster sutures at the upper, medial and lower borders of the muscle. In five patients we folded the muscle over itself to increase its volume to match the healthy side, see Table (1). Careful homeostasis, application of two suction drains one at the back and the other underneath the muscle in the pectoral region. Closure in layers starting with the deep subcutaneous tissue using 2/0 vicryle, followed by intradermal closure using the 3/0 proline suture. Light dressing applied to

the back and adhesive transparent dressing is applied to the mid axillary incission and pectoral bolster stitches as well.

Postoperative care and follow-up:

Patients stay in the hospital for two to three days or till they will be able to stay at home safely. Postoperative analgesics and antibiotics were given. Instructions were given to the patients to avoid muscular effort or exercise over the first six weeks, take a shower after five days. Follow-up in the outpatient clinic is carried out at one week, three weeks, three, six and nine months.

RESULTS

Over five years, twelve male patients with Poland syndrome have undergone reconstruction using the latismus dorsi muscle flap with some technical refinements. After an average follow-up period of nine months, no major general or local complications were reported. In terms of subjective

evaluations all of the patients were satisfied by the aesthetic outcome a detailed questionnaire were filled by all patients and scoring was done for each of the following items: Minimal hidden scar, breast volume and symmetry, infraclavicular fullness and anterior axillary fold reconstruction. Patients scored their satisfaction from 1 till 10, poor satisfaction will take a score (1,2,3), moderately satisfied will take a score (4,5,6,7) and very satisfied will score (8,9,10). See Table (2) and Figs. (2A,B and 3A,B, C,D).

Suction drains on the back were removed within an average Seven days while the pectoral one within an average two days. In four patients post operative pain was maximum at the site of the bolster stitches (33.3%). Seroma developed in one patient (8.3%). It responded to repeated aspiration and use of a compression garment and resolved in 14 days. One patient unfortunately fell from a height, lead to disruption of the muscle flap from its bed, which required secondary revision.

Table (1): Technical refinements utilizing latismus dorsi flap, complications and its management.

Technical Refinements of the latismus dorsi muscle flap	Complications	Management	
Reconstruction with the local muscle flap and attachment of the flap via bolsters: (12 patients)	Pain at the bolster sites in four patients	Pain Killers administered and patients' reassurance that it is self limited	
	Seroma in one patient	Repeated tapping and wearing a compression garment	
Latismus dorsi flap folding: (5 patients)	None	None	
Latissmus dorsi tendon reinsertion: (3 patients)	Accidental disruption	Secondary reinsertion	

All flaps were anchored to the anterior chest wall using bolster stitches, four patients suffered post operative pain, which improved with pain killers and patient reassurance, one patient developed seroma, which resolved after repeated tapping and wearing a compression garment. There is no complication in the five patients with folded muscle flap. Finally three patients were asking for anterior axillary fold reconstruction via latissmus dorsi tendon reinsertion, unfortunately one patient fell from a height and required secondary reinsertion.

Table (2): Patient satisfaction score as regard: Post operative pain, Scar in the mid axillary region, Breast Symetry, infarclavisular fullness and Anterior axillary fold reconstruction.

Satisfaction Score	Not satisfied	Moderately satisfied	Very satisfied
Post operative bolster stitches pain	1 patient	3 patients	8 patients
Scaring	None	1 patient	11 patients
Breast symmetry	None	2 patients	10 patients
Infraclavicular fullness	None	None	12 patients
Anterior axillary fold reconstruction	None	1 patient	2 patients

Post operative bolster stitch pain was not satisfactory in one patient and three were moderately satisfied and eight were very satisfied. All patients were moderately (1) to very satisfied (11) patients. All Patients were moderately to very satisfied with the breast symmetry. All were very satisfied with the infraclavicular fullness and anterior axillary fold reconstruction, except one patient who accidentally fell from a height.

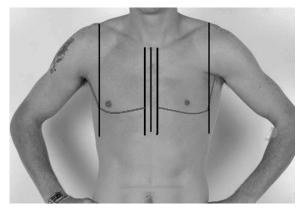


Fig. (1A): Marking the patient showing the midline of anterior chest wall, medial and lateral boundaries of the breast, infra mammary fold and the anterior axillary fold

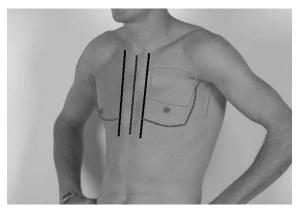


Fig. (1B): Photo for the same patient in oblique position showing the marked mid axillary incision and the infraclavicular hollowness.





Fig. (2A): Preoperative photos of male patient 26 years old, having left side moderate degree of male Poland syndrome deformity of the anterior chest wall.

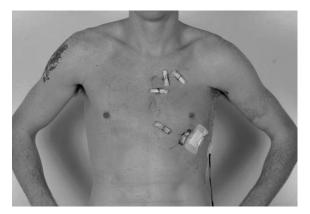


Fig. (2B): Early (five days) Postoperative photo of the same patient in Fig. (2A), showing much improvement of the infraclavicular hollowness and anterior chest wall symmetry.

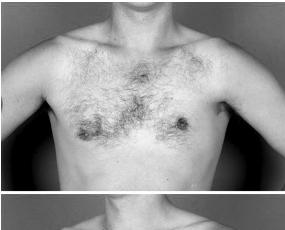




Fig. (3A): Preoperative photos of male patient 32 years old, having left side mild to moderate degree of male Poland syndrome deformity of the anterior chest





Fig. (3B): Early (seven days) Postoperative photo of the same patient in Fig. (3A).



Fig. (3C): Late postoperative photo (nine month) for the same patient in Fig. (3A).



Fig. (3D): Close up view showing minimal scarring over the hidden mid axillary region.

DISCUSSION

Several techniques have been proposed to achieve satisfactory results in patients with Poland syndromes starting with rib osteotomies [25], rib grafts [26] and microsurgical tissue transfer [27]. However, these operations are technically challenging, invasive, and result in significant donor site morbidity. The latissimus dorsi muscle flap provides

infraclavicular fullness, anterior chest wall and anterior axillary fold reconstruction, but despite these aesthetically pleasing results that can be achieved with this technique, substantial donor site morbidity exists because of the muscle harvest [28].

Endoscopic surgery offers the opportunity to minimize the aesthetic deformity associated with large incisions. Visualization is facilitated with the use of the endoscope, thus potentially allowing better control of perforating vessels, easier and more precise insetting of the flap, and reducing trauma to the flap during the raising and insetting portions of the procedure. However Potential drawbacks of endoscopic latissimus dorsi muscle harvest include increased operating time, the need for special equipment, and familiarity with endoscopic techniques [29].

Zhang et al., reported the use of customised textured silicone implant in seven male patients with a lateral thoracic incision [30]. The risks of implant related complications are infection, haematoma and seroma formation, implant extrusion, and capsule formation [31]. The customized pectoral silicone implant cannot correct the lost lateral sweep of the pectoral major muscle and it can only reconstruct the anterior chest wall skeletal and muscular deformity [32].

In male Poland's syndrome of the chest wall and breast, autologous fat injection seems to be a particularly useful technique in patients with mild deformity to fill the subclavicular and anterior axillary fold defects. Complication like fat necrosis in the injection area was recorded, probably due to crossing of the injection tunnels. In order to reduce this risk, a strict injection technique should be adhered to and injection of too much fat should be avoided. It is of course impossible to define the ideal volume because this depends on several factors: The patient's morphology, the quality of the teguments, the volumes treated. Autologous fat injection is a technique with negligible residual scarring, however it could not be used alone for treatment of all grades of male Poland's syndrome and several sessions at intervals of 4 to 6 might be carried [33].

By adopting our technique and its technical refinements, the results showed that it provides minimal scarring, decreasing the incidence of keloid or hypertrophic scaring in absence of endoscopic facilities, avoids the need for prosthetics with its drawbacks and we could reach a very good aesthetic outcome by the natural and almost symmetrical contouring of the anterior chest wall, contouring the infraclavicular hollowness and the anterior axillary fold. The latissimus dorsi muscle flap is very versatile as we could reconstruct mild, moderate and even sever grades of male Poland syndrome according to Foucras et al., 2004 by folding the flap over itself adding bulk of tissue to compensate for the sever anterior chest wall deficiency.

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