Proposal of the TBN Classification of Thoracic Anomalies and Treatment Algorithm for Poland Syndrome

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Background: Poland syndrome is a congenital deformity characterized by unilateral anomalies of pectoralis muscles, breast, nipple, axillary fold, subcutaneous tissue, ribs, and upper limb. The thoracic anomaly, which is the pathognomonic malformation of Poland syndrome, presents a wide phenotype variability and has been classified by different authors. However, these classifications do not include all the possible phenotypes of Poland syndrome. The aim of this study is to propose a simple classification of the whole spectrum of thoracic anomalies and a treatment algorithm that could have a practical value for determining the surgical approach.

Methods: Since 2008, 100 patients have been evaluated by the same plastic surgical team at San Martino Hospital-IST and Istituto Gaslini of Genoa, Italy, using the thorax, breast, nipple-areola complex (TBN) classification. Thoracic anomalies were classified as follows: thorax (T), from T1 (muscle defect only) to T4 (complex deformity with rib and sternal involvement); breast (B), in B1 (hypoplasia) or B2 (amastia); and nipple-areola complex (N), from N1 (dislocation <2 cm) to N3 (athelia).

Results: The most frequent thoracic anomalies were T1 (47 percent) and N2 (74 percent), whereas in female patients, B1 was more frequent than B2. The surgical approach to breast and pectoral reconstruction was based not only on the patient's age and sex, but also on the type of anomaly according to the TBN classification. In particular, a two-step approach with tissue expanders was required in N2 and N3 cases, whereas in N1 patients a single step was sufficient.

Conclusion: The TBN classification can be a useful tool for surgical decision-making according to each specific thoracic anomaly. (*Plast. Reconstr. Surg.* 138: 50, 2016.)

CLINICAL QUESTION/LEVEL OF EVIDENCE: Diagnostic, IV.

Poland syndrome, characterized by deficiency of the pectoralis major muscle,[1–4] presents a wide phenotype variability, including partial agenesis or deformity of the cartilage rib; hypoplasia or aplasia of the breast and nipple-areola complex, axillary fold, and subcutaneous tissue; sternal deformities; and anomalies of the ipsilateral arm.[5–7] The thoracic malformation is the pathognomonic feature of Poland syndrome and is the scope of our study. Although comprehensive classifications of all types of upper arm anomalies in Poland syndrome have been published and are recognized internationally as a reference tool for experts in this field,[8,9] thoracic abnormalities still lack a comprehensive classification. In the past, several classifications attempted to establish a degree of severity of the thoracic malformations in Poland syndrome (Table 1)[4,7,10–13]; however, (1) they do not include all the possible phenotypes of Poland syndrome; (2) although some prior classifications assumed a correlation between the malformation of the breast and the thoracic cage, this was not observed in our experience, as the severity of the thoracic anomaly is not necessarily correlated

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Table 1. Clinical Presentation and Classification of Thoracic Defect in Poland Syndrome: A Review of the Literature

Author	Cases	Degree	Pectoral Major Muscle Anomalies	Breast Anomalies	Rib/Sternal Involvement
Pegorier et al[10] 1994	8F 1M	Type A	Absence of sterno-costal head	Asymmetry Variable clinical presentation, NAC displacement	None
		Type B	Absence of sterno-costal head	Asymmetry Variable clinical presentation, NAC displacement	Rib anomalies
		Type C		Complete thoracic malformation	
Glicenstein[7] 2001	13F 7M		Absence of sterno costal head in all cases	Hypoplasia or aplasia, NAC and breast displacement, athelia	I Hypoplasia II: Sternal rotation or depression, pectus carinatum
			Alterations of other muscles	Contralateral breast: normal, hypoplasia, or hypertrophy	III: Rib aplasia
Foucras et al 2003[11] 2005[12]	19F 8M[11]	I	Hypoplasia of PMM	Moderate hypoplasia	Mild thorax asymmetry
		II	Aplasia of PMM	Major breast asymmetry	Possible moderate rib malformation Thorax asymmetry
	23F 14M[12]	III	Complete aplasia of PMM Other muscle aplasia associated	Aplasia of the breast	Major thorax asymmetry Rib aplasia, sternal deformation
Ribeiro et al[13] 2009	28 F	Mild	None, or partial absence of PMM	Amastia, hypomastia or areolar asymmetry	None
		Severe	Total absence of PMM; different alterations of the muscles	Hypomastia or amastia, areolar asymmetry	Different alterations of bones of ipsilateral chest
		Very severe	Different manifestations	Amastia; areolar asymmetry	Different manifestations
Seyfer et al[4] 2010	41F 22M	Simple form	Absence of the sternocostal head	Breast is smaller; NAC smaller and displaced toward axilla; color may be lighter	Hemithorax slightly smaller
		Complex form	Absence of sternocostal head	Breast and NAC are rudimentary or absent; if present, NAC is lighter in color and displaced toward the axilla	Hemithorax is smaller, upper anterior portions of rib absent or hypoplastic, and sternal aberrations
Baratte et al[14] 2011 (Foucras)	11F	Grade I	Hypoplasia	Moderate hypoplasia	None
		Grade II	Aplasia	Severe hypoplasia	Possible moderate rib malformation
		Grade III	Aplasia	Aplasia	Rib malformations
Zhu et al[15] 2012	24 F	Type I (Mild)	Hypoplasia of sternocostal head	Normal or hypoplastic	
		Type II (Moderate)	Moderately or severely hypoplastic	Moderately hypoplastic	
		Type III (Severe)	Severely hypoplastic or absent	Severely hypoplastic or absent	Usually severe rib and sternum deformities
Stylianos et al[16] 2012	7F 3M	1st	Hypoplasia		
		2nd	Absence of sternocostal head		
		3rd	Total absence of major or both pectoralis muscles		
		4th	Hypoplasia or absence of the pectoralis muscles		Skeletal anomalies of the thoracic bones (sternum or rib cage)

NAC, nipple-areola complex; PMM, pectoralis major muscle.
*Female patients.

with the severity of breast anomaly; (3) some of these classifications are based on female series only and therefore are not comprehensive of all Poland syndrome phenotypes; and (4) they are descriptive of Poland syndrome spectrum of anomalies, but their practical use is limited, as the surgical approach is not based on any of them. The aim of this study is to propose a new classification of Poland syndrome thoracic malformations and a treatment algorithm that, describing all the single aspects of the thoracic anomaly, could be helpful for surgeons to determine the type of surgical treatment.

PATIENTS AND METHODS

Since 2008, 100 patients were evaluated for Poland syndrome at San Martino Hospital-IST

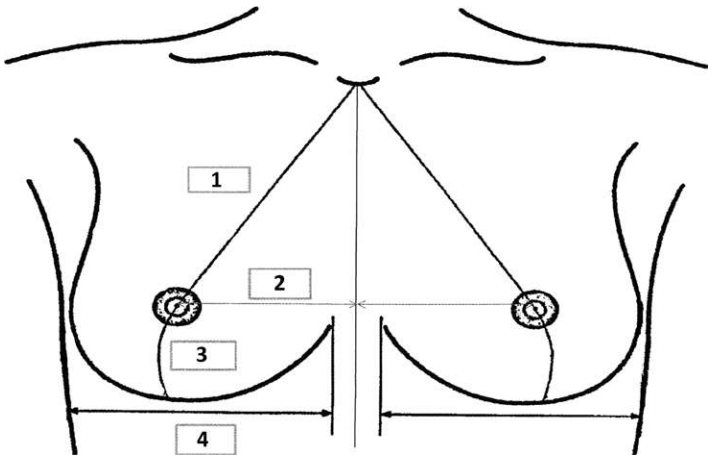

Fig. 1. Anthropometric measures.

and Istituto Gaslini of Genoa, Italy (national reference centers). The main criterion required for the diagnosis of Poland syndrome was pectoralis major muscle absence or hypoplasia, usually associated with breast, nipple, and soft-tissue ipsilateral defect. Rib agenesis, sternal deformities, and upper arm anomalies were also present in some cases. Clinical evaluation included measurement of the breast base; the nipple-areola complex diameter; and the distance between the sternal notch and the nipple-areola complex, the sternal midline and the nipple-areola complex, and the inframammary fold and the nipple-areola complex (Fig. 1). In all cases, photographic and echographic evaluations were available. In selected cases, computed tomography and/or magnetic resonance imaging of the chest were performed. Among the 100 evaluated patients, 70 underwent surgical treatment by the same plastic surgery team.

Table 2. TBN Classification of Thoracic Anomalies in Poland Syndrome

	Anomaly
T	Thoracic
T1	Hypoplasia or aplasia of pectoralis muscles and soft tissue
T2	T1 and sternal deformity, pectus excavatum and/or carinatum
T3	T1 and rib aplasia
T4	T1, T2, and T3 (muscle, sternum, and rib defect)
B	Breast
B1	Breast hypoplasia
B2	Breast aplasia
N	Nipple-areola complex
N1	NAC hypoplasia with dislocation of <2 cm
N2	NAC hypoplasia with dislocation of >2 cm
N3	Absent NAC

NAC, nipple-areola complex.

The first author (M.V.R.) created a classification of Poland syndrome thoracic abnormalities into the following features: thorax, breast, and nipple-areola complex (Table 2 and Figs. 2 through 4). This classification was called the thorax, breast, nipple-areola complex (TBN) classification.

The surgical approach (one or more steps, type of surgical treatment) was determined on the basis of age and sex of the patient, on the experience of the surgical team, and, particularly during the last few years, on TBN classification. A treatment algorithm is proposed (Fig. 5).

RESULTS

There were 56 male patients and 44 female patients; in 67 percent of them, the right side was affected, with a similar distribution in both sexes (66 percent in male patients and 68 percent in female patients). The distribution of each type of thoracic deformity in our series is shown in Tables 3 through 5.

The most frequent anomalies in male patients were T1N2 and T2N2, respectively (23 percent of all male patients); whereas in female patients, the most frequent anomaly was T1B1N2 (37 percent of all female cases). There was a similar distribution of T anomaly in both sexes with a high prevalence of T1, observed in one-half of patients. T2 affected one-third of patients (most frequent in male patients), T4 was present in 16 percent, and T3 was observed in 7 percent of patients of both sexes. Notably, cases of pectus excavatum and/or carinatum associated with Poland syndrome (T2 and T4) were very frequent, representing 46 percent in our series.

B1 was more frequent than B2 (75 percent versus 25 percent), with a high prevalence of N2 in both B1 and B2 groups. Regarding N, the most

Fig. 2. Thorax defect: T1 to T4. (*Above*, *left*) T1: hypoplasia or aplasia of pectoralis major muscle. (*Above*, *right*) T2: T1 and pectus excavatum and/or carinatum. (*Below*, *left*) T3: T1 and rib agenesis. (*Below*, *right*) T4: T1, T2, and T3.

frequent anomaly was N2 in both sexes; 86 percent in female patients and 64 percent in male patients. In male patients, N1 was presented in 27 percent of cases and N3 was uncommon (9 percent), whereas in female patients N1 and N3 were observed in the same number of cases (7 percent).

Concerning surgical treatment (Table 6), in T1 patients, characterized by agenesis of the pectoralis major muscle and soft tissue only, without sternum and rib malformation, thoracoplasty was never required. Conversely, patients with T4 presented a complex malformation and required thoracoplasty in all cases. In this group, thoracoplasty was always performed as the first step. In T2 and T3 patients, the indication for thoracoplasty was evaluated on a case-by-case basis in the specific anomaly and in our algorithm. In our series, there were seven T3 patients, but only two underwent surgery, and none of them required thoracoplasty, as rib agenesis was limited to one rib. Patients with T2 required thoracoplasty in 23 percent of cases; in 18 percent of cases costal deformity was performed in a single surgical step correction associated with positioning of a tissue expander.

On the basis of anthropometric parameters we have decided whether to perform reconstruction in one (N1) or two steps (N2 and N3). In female patients, all those cases with an important displacement of the nipple-areola complex (N2 or N3, representing 93 percent of all cases) needed the positioning of a tissue expander to mobilize the nipple and improve symmetry.

In male patients, a tissue expander was positioned in 77 percent of the patients with important

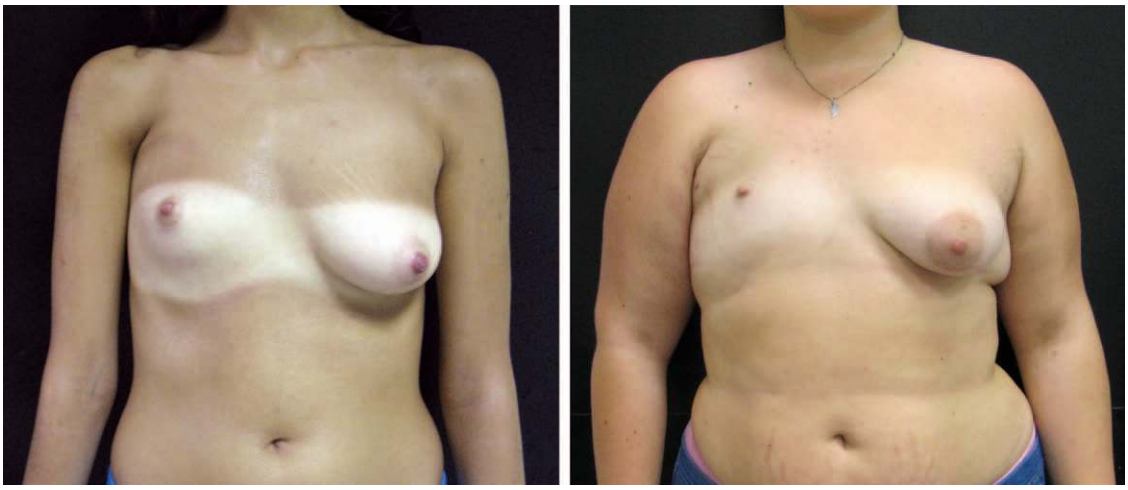

Fig. 3. Breast anomaly classification. (*Left*) B1: breast hypoplasia. (*Right*) B2: breast agenesis.

Fig. 4. Nipple anomaly classification. (*Left*) N1: nipple-areola complex dislocation less than 2 cm. (*Center*) N2: nipple-areola complex dislocation greater than 2 cm. (*Right*) N3: B2 (amastia) and atelia.

displacement of nipple-areola complex (N2 and N3, representing 73 percent of cases). All female patients underwent augmentation mastoplasty. In most cases, this step was preceded by fat grafting and/or positioning of a tissue expander, depending on N classification.

DISCUSSION

Considering the wide phenotype variability of thoracic abnormalities in Poland syndrome, a comprehensive classification was needed. Our aim was to propose a simple classification of the whole spectrum of Poland syndrome thoracic anomalies, which can be very complex. In our opinion, previous classifications did not describe precisely the thoracic malformation, as they did not include the whole phenotype range. Some classifications included only female patients.[13–15] In others, Poland syndrome was scored according to the severity of all clinical variables (e.g., muscle hypoplasia or aplasia, rib and/or sternal involvement, breast and nipple-areola complex hypoplasia or aplasia) considered together[16] (Table 1). To our knowledge, our series is the largest series of Poland syndrome cases reported in the literature. The availability of a wider phenotype range of Poland syndrome thoracic anomalies allowed us to propose a new, more comprehensive classification. In our experience, the single aspects of the thoracic malformation in Poland syndrome patients were variably associated. For example, we observed female patients with a severe rib anomaly (T4) who had only breast hypoplasia (B1).

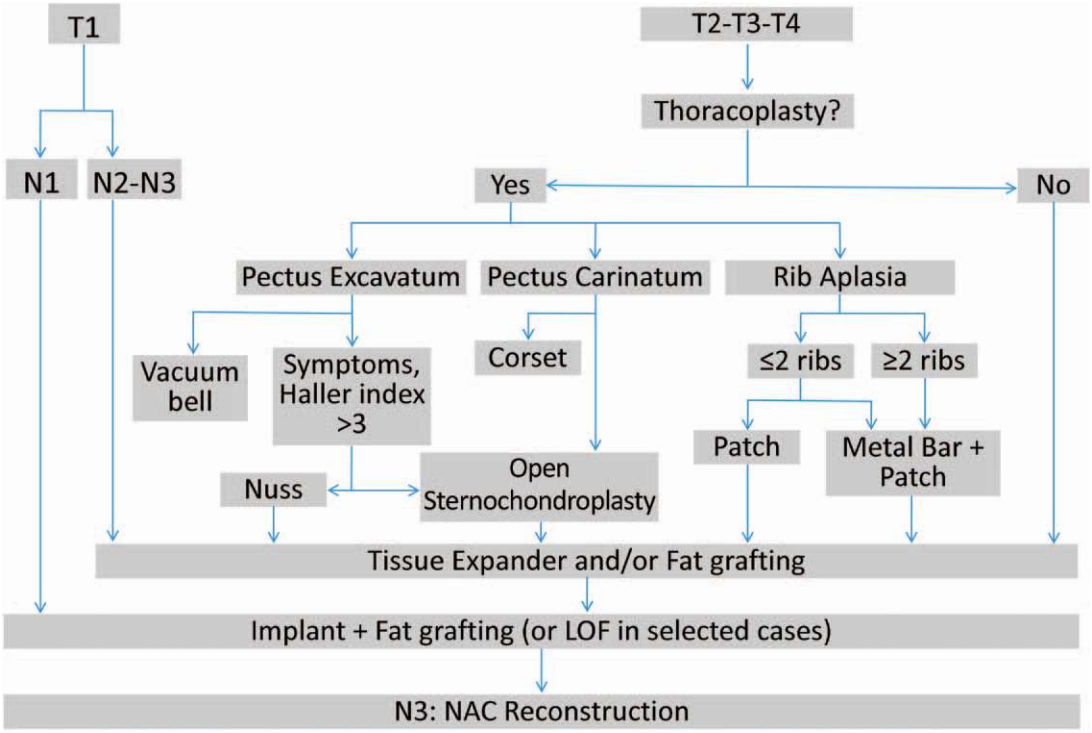

Fig. 5. Algorithm for treatment of Poland syndrome. Laparoscopic omental flap[18] (LOF) was indicated when fat grating was not possible. *NAC*, nipple-areola complex.

Table 3. Patients Classified According to Type of Chest Deformity.

Sex	No.	T1 (%)	T2 (%)	T3 (%)	T4 (%)
Male	56	23 (41)	20 (35)	4 (7)	9 (16)
Female	44	24 (54)	10 (23)	3 (7)	7 (16)
Total	100	47 (47)	30 (30)	7 (7)	16 (16)

Table 4. Male Patients Classified According to T and N Anomalies

	N1	N2	N3	Total
Total	15 (27%)	36 (64%)	5 (9%)	56
T1	10	13	0	23
T2	5	13	2	20
T3	0	3	1	4
T4	0	7	2	9

Table 5. Female Patients (*n* = 44) Classified According to T, B, and N Anomalies

	N1	N2	N3	Total
B1*	3 (7%)	38 (86%)	3 (7%)	
T1	3	17	0	20
T2	0	9	0	9
T3	0	1	0	1
T4	0	3	0	3
B2†				
T1	0	4	0	4
T2	0	1	0	1
T3	0	0	2	2
T4	0	3	1	4

*33 patients (75%).
†11 patients (25%).

Among male patients, there were cases with less severe thoracic abnormality (T2) who had absent nipple-areola complex (N3), whereas others with T4 had a nipple-areola complex, though it was abnormal (N2). All of these features are different manifestations of Poland syndrome with practical implications for surgical decision-making.

Several surgical approaches and techniques were adopted in our cases but are not the topic of this study, which is focused on the classification of Poland syndrome thoracic anomalies. The results of surgical treatment of Poland syndrome in our series require a separate study and will be submitted for publication in the future. However, we present here the algorithm of the surgical treatment adopted to show that the TBN classification can have practical implications for decision-making.

In all Poland syndrome patients, we indicate fat grafting[17] to improve volume, quality of skin, and elasticity, and it is especially useful in these types of patients with hypoplastic skin. A laparoscopic omental flap[18] was indicated when fat grating was not possible.

In our series, the measurement of sternal notch–to–nipple-areola complex and sternum-to–nipple-areola complex distance, and the

Table 6. Patients Distributed According to the TBN Classification and Surgical Treatment

Classification	Operated On* (%)	Requiring Thoracoplasty (%)
T	70	
T1	47 (67)	0
T2	13 (18.5)	23
T3	2 (2.9)	0
T4	8 (11.4)	100
N	70	
N1	22 (31.4)	14†
N2	44 (62.8)	90†
N3	4 (5.7)	100†
B (female patients only)	37	
B1 (33)	30 (81)	70†
B2 (11)	7 (19)	100†

*Of a total of 70.
†% requiring two-step breast reconstruction with tissue expander.

Fig. 6. Postoperative result of the T1N1 patient in Figure 2, *above*, *left* after fat grafting and pectoral implant.

Fig. 7. Postoperative result in the T3B1N2 patient in Figure 2c after fat grafting, reconstruction in two steps, and contralateral mastopexy.

Fig. 8. Postoperative result in the T3B3N3 patient in Figure 4, *right* after fat grafting, reconstruction in two steps, and nipple reconstruction.

determination of N were useful to determine the positioning of a tissue expander and to obtain nipple-areola complex symmetry improvement. Although N1 patients could be treated in a single step (Fig. 6), N2 or N3 patients required tissue expander positioning as a first step in the vast majority of cases (100 percent of female patients and 77 percent of male patients) (Figs. 7 and 8). The cutoff of 2 cm of dislocation of nipple-areola complex (see algorithm) is based on a lesson learned: the patients with a dislocation greater than 2 cm require a different surgical treatment (tissue expander) to obtain a satisfactory correction, whereas in the others cases, a good result could be achieved also without a tissue expander.

This difference between female patients and male patients was attributable to some male patients who preferred a single step. In our opinion, however, the best result in terms of symmetry is achievable with tissue expander positioning in all cases with dislocation more than 2 cm. Thoracoplasty was required in all T4 patients because of the severity of the malformation and in none of the T1 patients, whereas in T2 and T3 patients the decision to perform a thoracoplasty was made on a case-by-case basis.

Although every single patient with Poland syndrome is unique and the thoracic anomaly can present with different aspects even in the same group (not all T2 or T3 patients are affected by the same anomaly), we based our surgical indication

on the following criteria: T2 patients with two or more ribs affected were offered thoracoplasty [with Gore-Tex (W. L. Gore and Associates, Flagstaff, Ariz.) and/or metal bar]. T2 patients with only one rib affected were usually treated as T1 patients.

In T3 patients, the surgical indications for pectus carinatum and excavatum surgical treatment were the same as for the other pectus excavatum or carinatum patients without Poland syndrome.[19] In particular, the severity of the anomaly (in case of pectus excavatum evaluated with the Haller index and pectus correction index),[20,21] the presence of symptoms, and psychological discomfort were the indications for surgery. Younger patients, presenting without stiff thorax, were offered in the last 1 or 2 years a conservative treatment, based on a vacuum bell for pectus excavatum[22] and A dynamic compression system for pectus carinatum.[23] We have summarized the innovations and lessons learned in these years of experience.

Innovations and Lessons Learned

1. TBN classification.
2. Timing of surgery: we start in adolescence, performing different steps along with the physical development of the patient.
4. Preoperative and postoperative objective assessment: introduction of parameters and anthropometric measurements and photographic evaluation.
5. Algorithm of surgical treatment: identification of the surgical approach based on TBN classification.
6. Tissue expanders: indicated in N2 and N3 patients.
7. Lipofilling: always necessary to correct hypoplasia of subcutaneous tissue (always present in Poland syndrome).
8. Omental flap: indicated as an alternative to lipofilling in those patients in which the latter is not feasible.
9. Multidisciplinary team and collaboration with the thoracic surgeon: interventions with a team composed of both thoracic and plastic surgeons, to treat simultaneously the deformity of the rib cage and the pectoral/breast anomaly.
10. Considering not only the operation but the holistic care of the patient, including his or her psychological well-being.

The collaboration with the thoracic surgeon allowed us to treat these patients in a single session in most cases: during the same procedure, after the thoracoplasty performed by the thoracic surgeon, the plastic surgeon could insert the tissue expander. Unfortunately, a scientific evaluation of the surgical results of the treatment based on the proposed algorithm and analysis of the aesthetic results in the different groups of the TBN classification were not possible: this study is retrospective and the algorithm has been adopted only recently as the result of many lessons learned, but surgical treatment and indications have changed over the years, making this analysis very difficult.

CONCLUSIONS

In conclusion, Poland syndrome presents with a large variability of thoracic anomalies, and different surgical options have been reported in the literature. In our opinion, previous classifications did not include the whole spectrum of thoracic abnormalities, and therefore we propose a more comprehensive new classification that allows us to better define the thoracic anomaly in Poland syndrome and has proven to be helpful for determining the surgical approach.

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PATIENT CONSENT

The patients provided written consent for the use of their images.

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