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Surgical Algorithm of Poland Syndrome Based on Thorax, Breast, and Nipple-areola Complex Classification

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Background: Poland syndrome (PS) is a rare congenital syndrome characterized by unilateral pectoralis major muscle defect. In 2016, we proposed the thorax, breast, and nipple-areola complex (TBN) classification and a subsequent treatment algorithm, which included conservative and surgical procedures. Our aim is to report the results we obtained treating all thoracic anomalies according to the proposed algorithm in a cohort of adolescents affected by PS.

Methods: Between January 2016 and June 2023, 52 patients with PS were treated using the TBN classification in our institute. Each patient was evaluated by a multidisciplinary team composed of pediatric and plastic surgeons and treated according to the algorithm. Surgical procedures included were minimally invasive pectus excavatum repair, open sternochondroplasty, patch, metal plate, 3-dimensional chest wall prosthesis, fat grafting, tissue expanders, and breast and pectoral implants. Conservative treatments included vacuum bell and corset.

Results: Half of the patients had no thoracic skeletal defect (T1), but all had soft-tissues defects. Nineteen patients required thoracoplasty, all of whom experienced T downstaging after treatment, and 90% were postoperatively reclassified as T1. Conservative treatment for T correction was proposed in 8 patients; 83% underwent fat grafting, and 88% had a breast/pectoral prosthesis implanted. There were no major complications registered. The mean follow-up was 5.9 years.

Conclusions: The proposed algorithm can be considered a useful tool for standardized surgical decision-making in PS. Fat grafting can play a major role in a pediatric setting. A multidisciplinary and minimally invasive approach, whenever possible, should be prioritized in patients younger than 18 years. *(Plast Reconstr Surg Glob Open 2024; 12:e6261; doi: 10.1097/GOX.0000000000006261; Published online 22 November 2024.)*

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INTRODUCTION

Poland syndrome (PS) (OMIM: 173800) is a congenital syndrome (incidence 1/30,000)[1,2] characterized by the partial or complete unilateral absence of the pectoralis major muscle, which can be associated with a wide range of defects in the chest, ipsilateral upper limb, and solid organs.[2,3] Thoracic malformations in PS are characterized by high phenotypic variability. In 2016, our unit proposed the thorax, breast, and nipple-areola complex (TBN) classification as a simple tool for the classification of the whole spectrum of PS thoracic anomalies (Table 1).[4] Based on this classification, an algorithm for PS treatment was proposed (Fig. 1), including both surgical and conservative treatments. The main objective of our study is to report our results and evaluate the applications of the proposed algorithm in adolescents affected by PS.

Disclosure statements are at the end of this article, following the correspondence information.

To our knowledge, no other reports describing an algorithm for thoracic anomalies systematic classification and treatment in PS have been published. Overall, very little information about PS global management in children and adolescents is available.

PATIENTS AND METHODS

From January 2016 to September 2023, 100 patients with PS 11–18 years of age were evaluated at Istituto Gaslini. Patients younger than 12 years and patients who did not complete their surgical reconstructive pathway were excluded from this study. In our institute, PS diagnosis was initially made by clinical detection of complete or partial absence of the pectoralis major muscle; in all cases, the diagnosis was confirmed by musculoskeletal ultrasound. Anthropometric measures [sternal notch to nipple-areola complex (NAC); sternal midline to NAC; NAC to inframammary fold distances; and NAC diameter] were taken to classify every patient according to the TBN classification (Table 1).[4] Ribcage anomalies were clinically defined; paradoxical chest movements were evaluated in cases of rib aplasia. Computed tomography (CT) and magnetic resonance imaging were requested in cases of severe chest deformities. Detailed diagnostic workup for PS in our center has been previously described.[4] Functional impairment, such as effort dyspnea and other respiratory symptoms, was always investigated: echocardiography and spirometry tests were prescribed in T2 and T4 patients when heart and/or lung compression was suspected. Based on clinical and radiological evaluations and TBN classification, our team of pediatric thoracic and plastic surgeons defined the most suitable treatment plan for each patient. Thoracic surgery was proposed for functional and/or cosmetic purposes.

Surgical treatment for T1 patients (no associated skeletal deformities) and for breast (B) and nipple (N) defect correction included fat grafting (FG), tissue expanders, breast and pectoral implants, and 3-dimensional (3D) custom-made pectoralis prosthesis in men. Due to a lack of pectoral muscles, expanders and implants were placed subcutaneously. Tissue expanders were positioned and oriented based on each patient's anthropometric measurements to obtain more tissue expansion where needed (the expander is frequently placed in the superomedial quadrant in an oblique position, to allow the NAC to be subsequently displaced inferiorly, symmetrical to the contralateral). After the process of skin expansion, substitution of the expander with breast or pectoral implant and FG were performed.

For FG, we adopted the "wet" method of fat harvesting: the donor site (abdominal region and/or thighs) was injected with a modified Klein solution (composed of 1 mg Epinephrine, 10 mEq $NaHCO_3$, 200 mg mepivacaine hydrocloride and 50 mg ropivacaine added to 500 mL 0.9% NaCl). Twenty minutes after tumescent infiltration, manual liposuction harvest was performed using a 3-mm diameter liposuction cannula, followed by gravity separation process (20 min) and FG in 3-mL syringes.

Surgical treatment for T2 category consisted of minimally invasive pectus excavatum repair (MIRPE) proposed by Nuss or open sternochondroplasty for patients with pectus carinatum (PC). Patients presenting a compliant chest wall were considered eligible for conservative treatments which included vacuum bell (VB) and corset to improve pectus excavatum (PE) and PC, respectively. Surgical repair of T3, depending on the severity of the defect, consisted in rib cage stabilization using a Goretex patch, metallic plate, or 3D-printed custom-made metallic prosthesis. In the case of complex malformations (T4), surgery was always proposed.

Takeaways

Question: How can we optimize the treatment of the whole spectrum of Poland syndrome (PS) thoracic anomalies?

Findings: Analysis of the results of 52 pediatric patients affected by PS treated according to the thorax, breast, and nipple-areola complex (TBN) classifications-based algorithm. All patients decreased or improved the severity of their malformations and chest symmetry (T downstaging) with a better quality of life after treatment.

Meaning: This is the first attempt to adopt a systematic approach to skeletal deformity correction in PS pediatric/adolescent patients. TBN classification and the proposed algorithm demonstrated their effectiveness and can be useful tools to standardize chest treatment in PS.

Table 1. TBN Classification by Romanini et al

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		NAC
	N1	NAC hypoplasia with dislocation of <2 cm
	N2	NAC hypoplasia with dislocation of >2 cm
	N3	Absent NAC

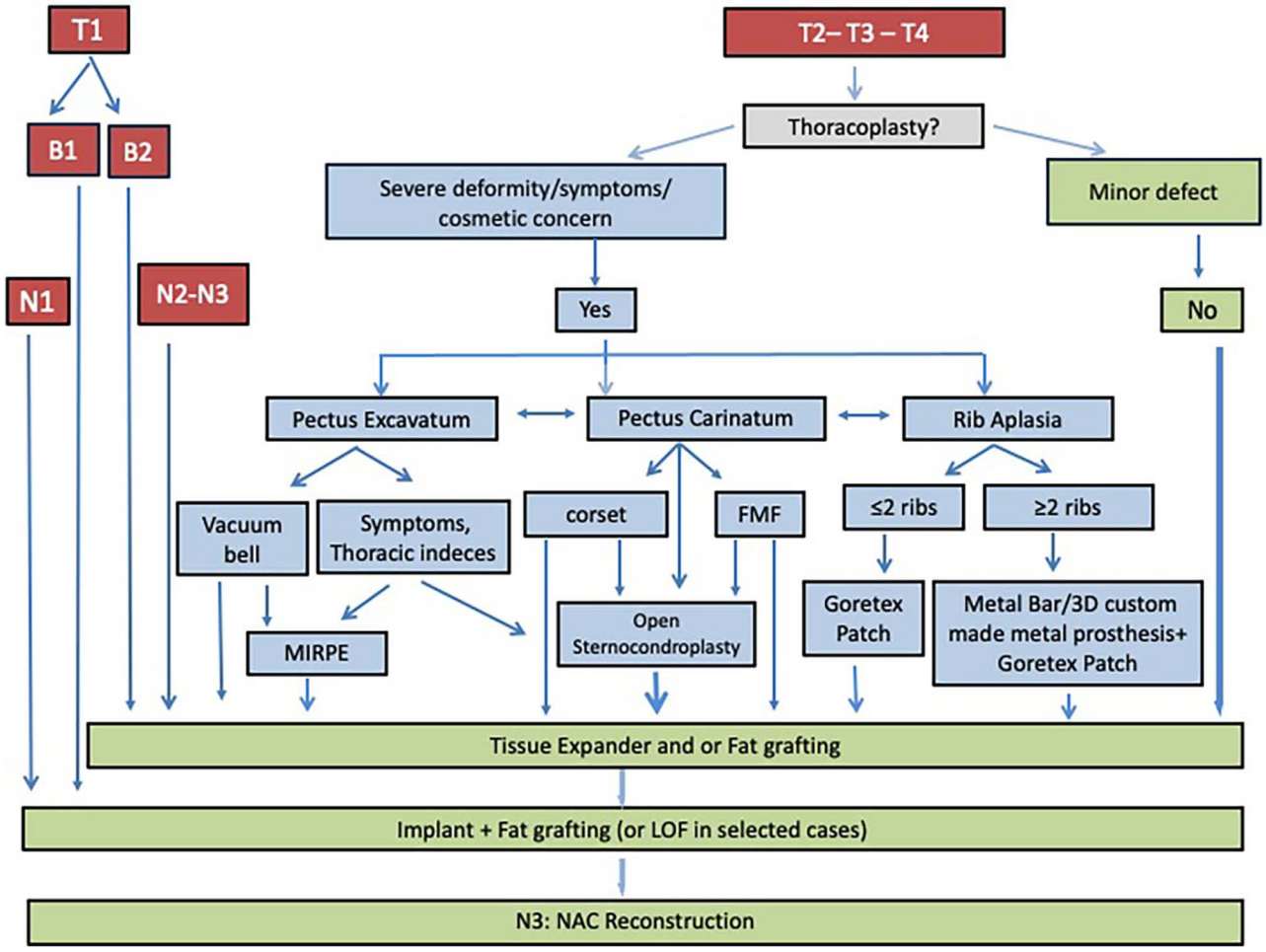

Fig. 1. Treatment algorithm based on TBN classification. FMF, FMF Dynamic System; LOF, laparoscopic omental flap.

Rib cage defects were corrected before soft-tissue and breast defects. In some cases, thoracoplasty and the first plastic surgery step were performed in the same session.

After every procedure (surgical or conservative), in the period between different surgical steps and before any surgery, each patient was clinically evaluated in the outpatient clinic by the same surgical team to reassess the thoracic defect according to the TBN classification. All patients who underwent FG and/or breast or pectoralis prosthesis placement performed an annual ultrasound as follow-up. Patients' satisfaction was determined by asking them to evaluate their chest appearance at the beginning and end of the reconstruction process, using a 1 to 10 rating scale (1 corresponds to very dissatisfied and 10 to very satisfied).

An approval statement for publication was obtained by our regional ethics committee (CER Liguria) on May 12, 2022, authorization n° 636/2022–DB id 12824. Pictures of the chest were taken during physical examination and stored in a password-protected institutional cloud service, and informed consent for publication was obtained, in compliance with the GDPR.

RESULTS

A total of 52 patients with PS completed their reconstruction and were included in this study. The majority of them were men (28 versus 24) and had right-sided PS (37 versus 15). One patient had bilateral PS, T4 on the left side, and T1 on the right side; in this patient, we considered the left side only.

The type of thoracic deformity is shown in Table 2, male and female distribution according to TBN classification in Tables 3 and 4, conservative and surgical treatment for chest wall anomaly in Table 5, and all surgical treatments performed in Table 6.

All 25 patients with a T1 defect underwent surgical breast and/or pectoral and soft-tissue reconstruction. In 3 T1 male patients, FG alone was sufficient to correct soft-tissue defects. Considering the entire cohort, 83% of patients required FG, and each patient underwent a mean of 2 procedures. Fat donor sites considered were abdomen and/or inside and outside of the thighs in women. The injected volume in each session varied based on each patients' fat availability (range: 30–240 mL).

Based on our surgical algorithm, all patients classified as N2 required a tissue expander placement to correct

the NAC position before the breast/pectoral reconstruction, and subsequently, nearly 90% of patients implanted a breast/pectoralis prosthesis. Among surgically treated female patients, 9 had contralateral tuberous breast, and its correction was included in the surgical path to improve breast symmetry and optimize surgical timing.

Complex deformities required a combination of different procedures (Table 6); 3 T2 patients underwent MIRPE (2 with 2 retrosternal bars and 1 with a single bar). A patient with PE and PS is shown in Figure 2. The mean time before bar removal was 3 years.

Table 2. Patient Classification According to Thoracic Anomaly

T	N
T1	
25	
T2	
Pectus excavatum	4
Pectus carinatum	4
Currarino-Silverman syndrome	2
T3	
Rib aplasia < 2	1
Rib aplasia > 2	1
T4	
Pectus excavatum + rib aplasia	1
Pectus carinatum + rib aplasia	7
Pectus carinatum + Pectus excavatum + rib aplasia	7
Total	52

Sternochondroplasty was performed in 14 patients (12 T4 and 2 T2). In all but 3 cases, it was associated with other surgical procedures to correct rib aplasia or sternal depression. Prominent costal cartilages were resected through a sternal approach. The mean number of costal cartilages resected in a single procedure was 2 (range: 1–3). The fragments of the smashed cartilages were placed on a Vicryl mesh to correct sternal depression in 5 T4 patients with associated PE. Through a second incision on the anterior axillary line, rib aplasia was corrected using metal implants fixed to the sternum and/or a Goretex patch. Through the same incision, a subcutaneous pocket was created to implant a breast/pectoral expander or prosthesis for soft-tissue reconstruction (Fig. 3 after FG in the same patient). The position of the expander was chosen based on the anthropometric measures and, more importantly, on the distance from NAC, with the purpose of obtaining NAC maximal symmetry.[4] In 11 patients, breast/pectoral expanders were implanted in the same surgical session as the thoracoplasty (Figs. 2C, 3B). Particular attention is needed to prevent iatrogenic symmastia during dissection in female patients.

Among 17 patients with rib aplasia (2 T3 and 15 T4), 12 underwent thoracoplasty: Goretex patches alone were sufficient for 5 minor defects, 3 patients were treated with metal plate implants (a single bar in 2 cases, 2 bars in 1), and 4 patients were treated with a 3D custom-made metallic prosthesis. Different models of custom-made titanium rib prostheses were used. All metal devices were covered with a Goretex patch. A female patient classified as T4B2N3 is shown in preoperative view in Figure 4A,

Table 3. Male Patients Classified According to T and N Anomalies

	N1	N2	N3	Total
Total	6	21	1	28
T1	5	8	1	14
T2	1	3	0	4
T3	0	1	0	1
T4	0	9	0	9

Table 4. Female Patients Classified According to T, B, and N Anomalies

		N1	N2	N3	Total
B1	T1	0	9	0	9
	T2	0	6	0	6
	T3	0	0	0	0
	T4	0	0	1	1
B2	T1	0	1	1	2
	T2	0	0	0	0
	T3	0	0	1	1
	T4	1	2	2	5
Total		1	18	5	24

Table 5. Conservative and Surgical Treatment in Chest Wall Anomaly (T)

T (n)	Chest Surgical Treatment * (n)	Conservative Treatment * (n)	Conservative and Surgical Treatment * (n)	NO Treatment* (n)	Plastic Surgery (n)
T1 (25)	Not indicated	Not indicated	Not indicated	Not indicated	25
T2 (10)	4	2	1	3	10
T3 (2)	1	—	—	1	2
T4 (15)	9	1	4	1	15

Chest surgical treatment, conservative treatment and conservative and surgical treatment refer to "T" defect.

Table 6. Surgical Treatment T Patients

	T1, n (%)	T2, n (%)	T3, n (%)	T4, n (%)	Total, n (%)
Patients	25 (100)	10 (100)	2(100)	15(100)	52 (100)
MIRPE (Nuss)	0	3 (30)	0	0	3 (6)
Patch	0	0	1 (50)	11 (73)	12 (23)
Sternochondroplasty	0	2 (20)	0	11 (73)	13 (25)
Metal bar	0	1 (10)	0	3 (20)	4 (7)
Custom-made 3D metal prosthesis	0	0	0	4 (27)	4 (7)
FG	17 (68)	10 (100)	2 (100)	14 (93)	43 (83)
Expander	14 (56)	8 (80)	2 (100)	8 (53)	32 (62)
Breast/pectoral prosthesis	22 (88)	8 (80)	2 (100)	14 (93)	46 (88)
Standard	20 (80)	8 (80)	2 (100)	13 (87)	43 (83)
Custom-made	2 (8)	0	0	1 (6)	3 (6)

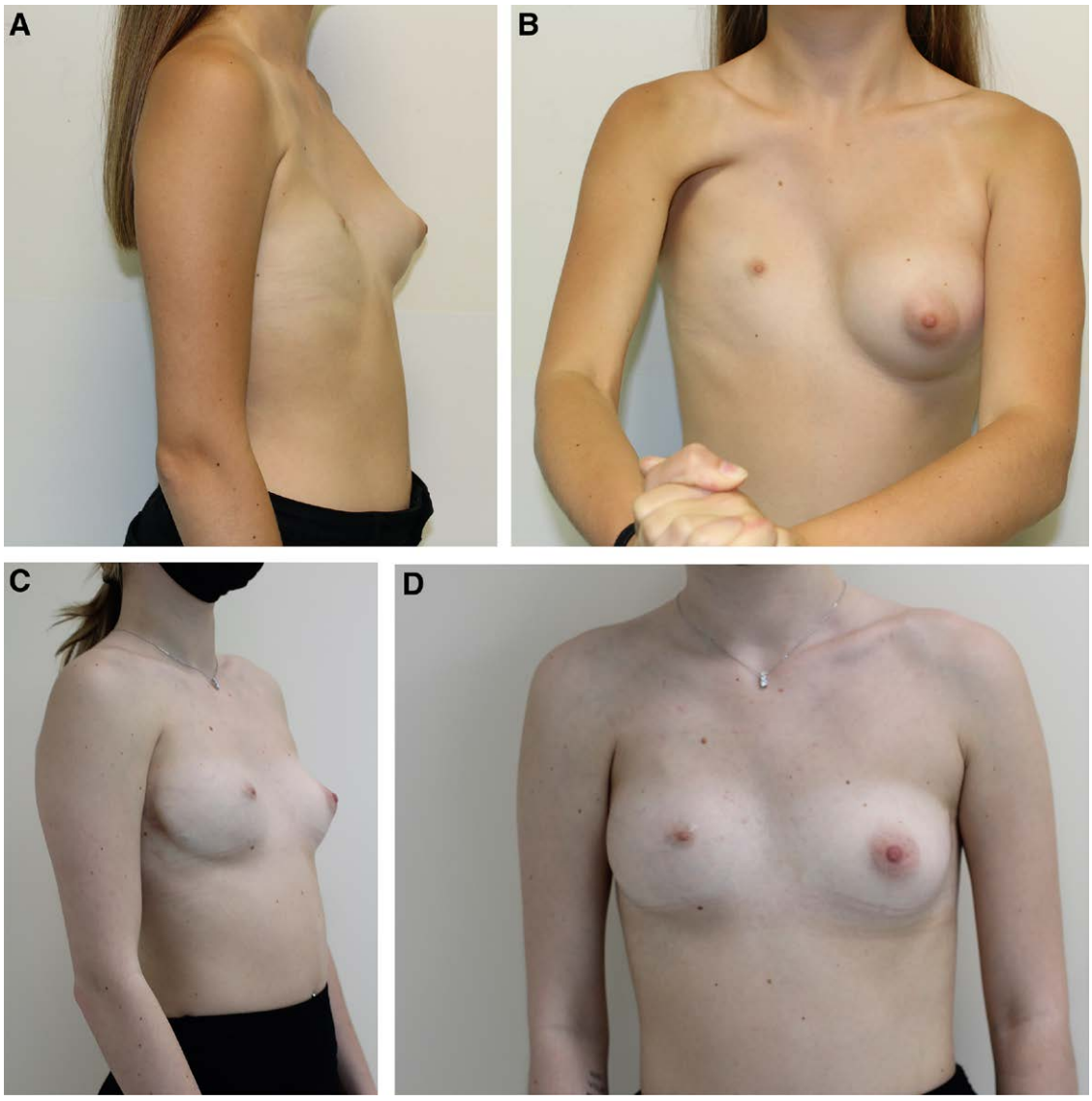

Fig. 2. T2B2N2 patient affected by PE. A-B, Preoperative images. C-D, Postoperative results after multidisciplinary approach: MIRPE (2 bars) and breast expander.

her scanner in Figure 4B, 3D implant in Figure 4C, intraoperative picture showing 3D implant in Figure 4D, and after breast reconstruction in Figure 4E before the nipple-areolar reconstruction.

Regarding conservative treatment, a compression corset and VB were prescribed in 6 patients with PC and in 2 patients with PE, respectively. Seven of 8 patients who received conservative treatment experienced downstaging

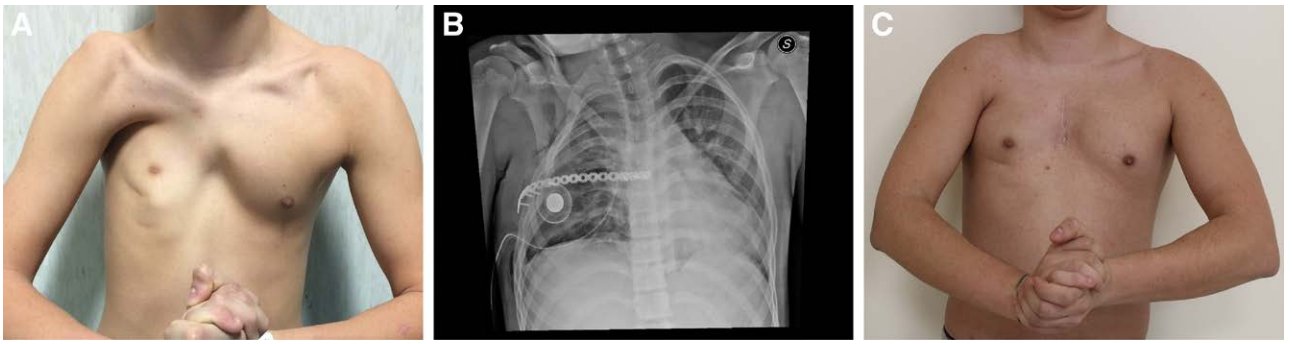

Fig. 3. Treatment of a 15-year-old boy with PS, T4N2 (PC and rib aplasia) treated first conservatively with a corset. A, A 15-year-old boy, T4N2 (PC and rib aplasia) treated first conservatively with a corset and then with surgical thoracoplasty (sternochondroplasty and metal bar) associated with breast expander implant. B, Post thoracoplasty XR. C, Final result after pectoral implant and FG, 17 years old.

of the thoracic defect (T4 to T3 and T2 to T1) and 4 of them (1 treated with VB and 3 treated with corset) did not require any other rib cage surgery.

After treatment (surgical and/or conservative), patients were reclassified as shown in Figure 5. All patients experienced T downstaging after treatment, and 20 of them (90%) were reclassified as T1 (no residual skeletal malformations) after thoracoplasty. Chest symmetry improved in all patients after treatment, and residual minimal breast asymmetry was detected in 5 patients who refused other treatments. All patients had an improvement of thoracic symmetry based on the anthropometric measures. Of the 39 patients initially classified as N2 (21 men and 18 women), 37 were reclassified as N1 at the end of breast/pectoral reconstruction; all 37 patients initially had the affected NAC displaced cranially, more than 8 cm compared with the contralateral and no contralateral mastopexy were performed. Each patient retained their own NAC.

The mean follow-up was 5.9 years (range, 3.3–7.6 y). Five patients presented an early complication: 1 experienced a pneumothorax after thoracoplasty requiring chest drainage, 2 presented with a hematoma, 1 with deep vein thrombosis, and 1 patient had a wound dehiscence treated with vacuum-assisted closure therapy. Late complications included capsular contracture (2), symmastia (1), and fat cyst in the subcutaneous tissue (5). The mean scores obtained when asking our patient to judge their chest appearance at the beginning and at the end of the reconstruction process using the abovementioned scale were 3.5 and 8.5, respectively.

DISCUSSION

Thoracic defects (both skeletal and soft tissue) are highly variable with PS, so it can be difficult to define a standardized and systematic treatment. Although soft-tissue reconstruction strategies in PS have been widely described in the literature,[5–8] little data on global correction of PS related thoracic malformation are available, particularly in more severe cases. In the past, ribs translocation from the contralateral to the affected side has been proposed to correct severe defect.[9,10] However, rib translocation is an invasive procedure causing damage to the healthy donor site, and it has been abandoned by most centers, as suggested by the lack of reports in recent years. In 2015, Majdak-Paredes et al[11] published a reconstruction algorithm for complex forms of PS. They proposed custom-made silicone prostheses to mask severe skeletal chest wall deformities. The adoption of specific surgical techniques, such as MIRPE, has been described in the literature as isolated case reports and has not been included in a systematic surgical approach.[12,13] Recently, consensus-based recommendations for medical management of PS were published.[14] However, at the present time, no surgical guidelines or recommendations have been reported in the literature regarding the management of thoracic defects in PS, and little information on PS treatment in children or teenagers is available. In 2016, Romanini et al[4] proposed an innovative algorithm for chest wall treatment in PS patients based on the TBN classification, which has been adopted in our institute and applied in pediatric and adolescent settings. As reported above, the results are extremely encouraging. Treatment of skeletal deformities performed as a first step allows harmonization between the right and left sides of the thorax, and the following breast/pectoral reconstructive steps enable the possibility of achieving good symmetry and cosmetic result.

Thoracic surgery in PS has both a cosmetic and functional purpose. Multidisciplinary evaluation (thoracic and plastic surgeons) is essential to define the most suitable surgical pathway for each patient Ribcage deformity correction was identified as the initial step, which is preparatory to soft tissue and pectoral or breast reconstruction. In regards to the T category, in cases of severe sternum and rib malformation (T4), thoracoplasty is required; T1 never needs thoracoplasty, whereas T2 and T3 could benefit from it. In some cases, thoracoplasty and the first plastic surgery step could be performed in the same session, which reduces the number of surgical interventions (Figs. 2, 3, 4B). Regarding breast defects (female patients), B1 patients can benefit from breast reconstruction and/or breast symmetry improvement, whereas B2 patients (amastia) always need breast reconstruction. NAC dislocation greater than 2 cm (N2)

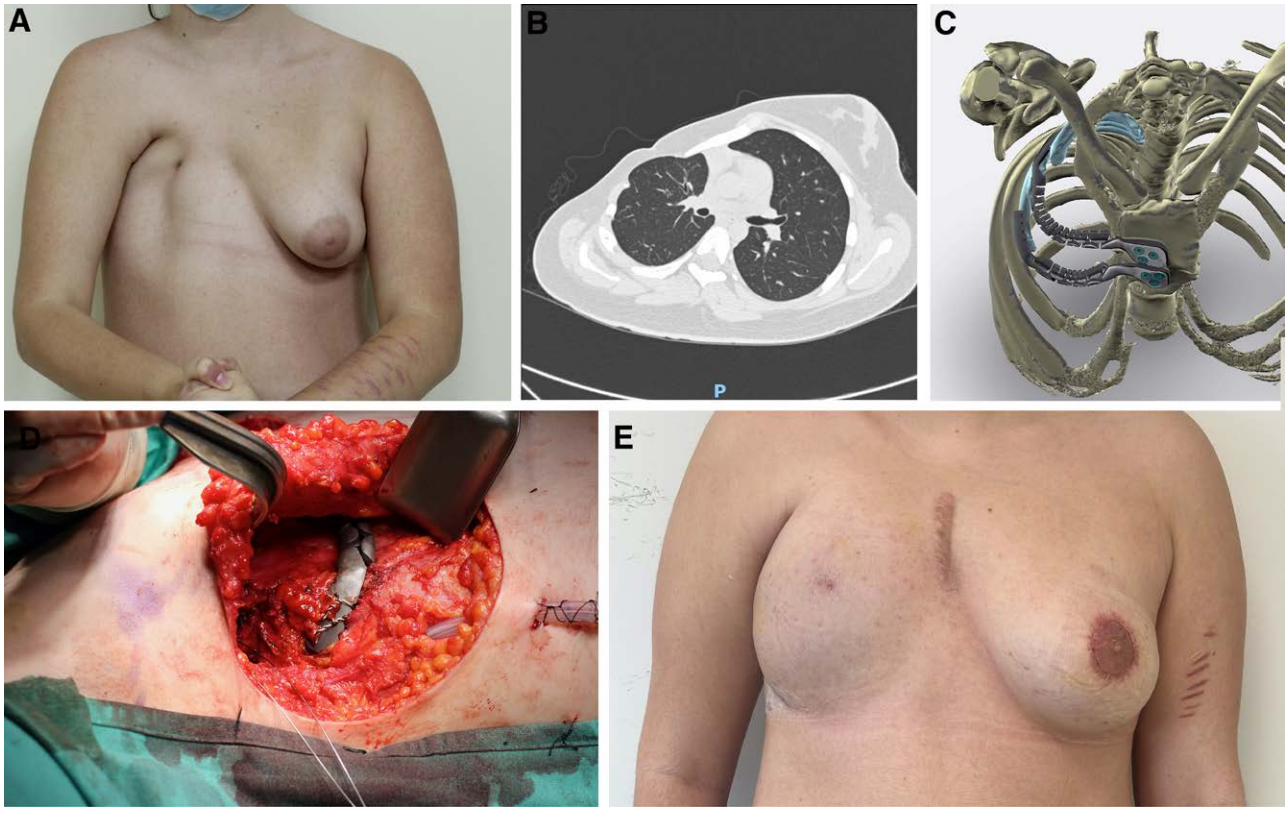

Fig. 4. T4 complex thoracic malformation T4B2N3 16 years old: (A) preoperative CT scan (B) preoperative image of the patient contracting the pectoralis muscles; (C) 3D CT scan and design of the prosthesis; (D) intraoperative picture of the 3D chest wall implant; and (E) postoperative result after complex thoracoplasty (3D custom-made prosthesis, Goretex patch, breast expander, and FG).

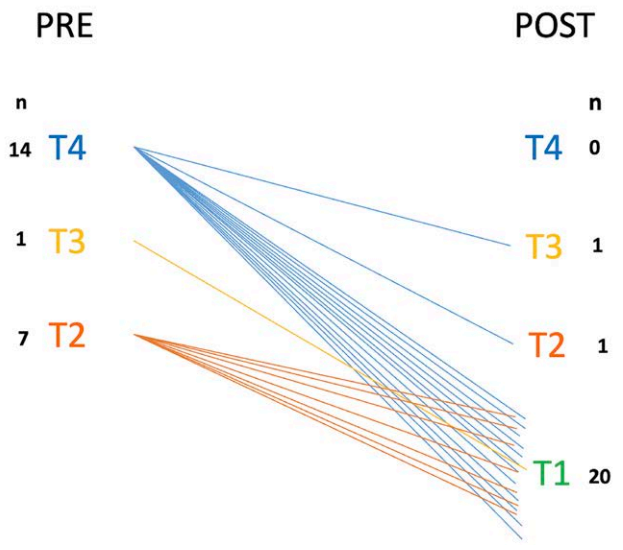

Fig. 5. Graph showing the reduction in the severity of chest wall malformation on patients with PS after surgery.

requires, both in female and male patients, a tissue expander placement with the purpose of improving NAC symmetry before breast/pectoral reconstruction with a breast/pectoral implant.

FG plays a major role: whenever feasible, we proposed FG to improve skin trophism, increase subcutaneous volume, reshape the axillary fold, correct residual volume defect after prosthesis implantation, smooth the implant's profile, and treat capsular contracture. For these purposes, FG is performed before, during, and after expander/implant placement. In some cases, just FG was used to correct volume asymmetry and improve NAC position, avoiding prosthetic implants. Any contralateral defect (eg, gynecomastia, tuberous breast) was investigated and its correction included in each patient's surgical plan.

Some authors have proposed custom-made silicone prostheses[11,12] as a primary surgical option in patients affected by severe chest wall deformities. In our opinion, for these patients, it is preferable to first fix the ribcage defect and the possible related complications (such as cardiac compression and paradoxical respiration) and subsequently to correct soft-tissue defects. Our group uses custom-made silicone pectoral implants to reshape the pectoralis muscle in cases that are predisposed to less favorable outcomes with the standard pectoral implant. In our experience, the TBN classification proved to be a dynamic concept: (1) the same type of "T" can include patients with different degrees of defect (patients with mild or severe PE or PC are all classified as T2, but their treatment differs according to severity of the malformation)

and (2) T4 patients can become T1 and the "N" category can improve to N1.

In children with very elastic chest walls, conservative treatments could be proposed as a first choice. In our experience, conservative treatments alone can lead to "T" downstaging (Fig. 5).[15–18]

Three-dimensional custom-made titanium prosthesis for chest wall malformation treatment is a rising procedure in more severe cases[2,19–21] With the current technology, the prosthesis is easily designed based on a simple preoperative CT scan (Fig. 2). Preliminary results are promising, but our experience is still limited to few cases.

Latissimus dorsi muscle flap transfer has been traditionally considered in the literature as a safe and efficient procedure to correct chest wall defects in PS, but minimally invasive alternatives were recently described.[22,23] However, we decided to exclude this approach from our treatment algorithm for the following reasons: (1) latissimus dorsi could be absent/hypoplastic in PS[24]; (2) this technique does not correct possible skeletal defects; (3) we prefer to avoid any iatrogenic upper limb muscular deficit in a healthy shoulder, especially during the growing age; (4) latissimus dorsi does not replace the function of pectoralis muscle and can become atrophic after the transfer; and (5) alternative techniques associated with lower morbidity in children are now available, particularly fat transfer and implants.

Despite some patients requiring high-complexity surgery, we observed a small number of complications. A multidisciplinary approach and the centralization of patients in a high-volume center can improve the quality of care.

Another matter of discussion about PS surgical treatment is the age at which it is advisable to start the correction. Generally, adult thoracic surgeons and plastic surgeons prefer to wait until the end of puberty, to avoid intervening in a growing chest. In our opinion, except for specific cases, adolescence is the preferred time to start the surgical pathway, because chest wall and breast development are almost complete, and patients start experiencing high levels of psychological stress for their malformation. It has been demonstrated that patients will eventually have better body image and quality of life when chest reconstruction is started before the patient is 18 years old.[25] All female patients treated during adolescence were classified as Tanner IV, and surgery was performed at least 2 years after menarche. Currently, no guidelines are available regarding psychological support for patients with PS; however, in our opinion, the decision to undergo surgery should be taken with the help of a psychologist and in full awareness.[25]

Considering the rarity of PS, the main strength of this study is to report, for the first time in the literature, the results of a relatively large cohort of pediatric/adolescent patients with PS treated according to a standardized algorithm, considering the entire spectrum of thoracic defects. Nonetheless, this research has some limitations. First, patients' outcomes after ribcage surgery were assessed solely on clinical evaluation due to the fact that they usually do not repeat CT or magnetic resonance imaging scans after ribcage surgical treatment if not necessary. Second, patient satisfaction was not thoroughly assessed. As previously mentioned, patients were asked to evaluate their chest at the beginning and end of reconstruction using a 1 to 10 auto-rating scale, which resulted in a substantial improvement.

In conclusion, this is the first attempt at adopting a systematic approach to skeletal deformity correction in PS pediatric patients. Our results demonstrate that the TBN classification and proposed algorithm can be useful tools to standardize treatment in children and adolescents affected by PS. In the pediatric setting, a multidisciplinary and minimally invasive approach, whenever possible, is to be preferred. Reconstructive surgery is best started during adolescence.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

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ETHICAL APPROVAL

An approval statement for publication was obtained by our regional Ethics Committee (CER Liguria) on May 12, 2022, authorization no. 636/2022–DB ID 12824.

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