Chest wall reconstruction in children and adolescents: recent personal experience from a case series and narrative review

Paolo Scanagatta[1,2]^, Casimiro Eugenio Giorgetta[1], Eugenio Ravalli[1], Francesco Inzirillo[1]^, Luca Colombo[3], Stefano Sestini[4]^, Sara Cagnetti[1], Giuseppe Naldi[1]^

[1]Division of Thoracic Surgery, ASST Valtellina e Alto Lario, "Eugenio Morelli" Hospital, Sondalo, Italy; [2]School of Medicine, University of Milan, Milan, Italy; [3]Pediatric Orthopedics and Traumatology, "Vittore Buzzi" Children's Hospital, Milan, Italy; [4]Division of Thoracic Surgery, ASST Mantova, Mantova, Italy

Contributions: (I) Conception and design: P Scanagatta, G Naldi; (II) Administrative support: P Scanagatta; (III) Provision of study materials or patients: P Scanagatta, L Colombo, S Sestini; (IV) Collection and assembly of data: P Scanagatta, L Colombo, S Sestini, G Naldi; (V) Data analysis and interpretation: P Scanagatta, G Naldi; (VI) Manuscript writing: All authors; (VII) Final approval of manuscript: All authors.

Correspondence to: Paolo Scanagatta, MD. Director, Department of Thoracic Surgery, ASST Valtellina e Alto Lario "Eugenio Morelli" Hospital, Via Zubiani 33, 23035 Sondalo, Italy. Email: paolo.scanagatta@asst-val.it or paoscan@hotmail.com.

Background: Resection of chest wall tumors is a key step in multimodal treatment. In children and adolescents, planning a proper reconstruction is challenging, as the future growth of the chest wall should be foreseen and considered.

Case Description: We present the satisfactory preliminary results of a case series. This is the first study regarding an original semi-rigid reconstruction technique following chest wall resection in children and adolescents operated for chest-wall tumors. We present the recent experience of 5 patients (age 12–18 years), treated by chest wall resection and reconstruction using a Polypropylene knitted mesh, technically adapted to obtain a semi-rigid prosthesis. A narrative review was performed through an electronic search for relevant studies from January 1990 to April 2022 in the following databases: PubMed, Science Direct, Embase, Scopus and Cochrane. Our reconstruction technique warranted good aesthetic and functional results in the short and medium term (medium follow up 27.6 months; range, 12–52 months). Currently we have not observed significant deformation of chest wall related to the surgical intervention.

Conclusions: The use of a semi-rigid reconstruction technique following chest wall resection in patient during the growing age could provide satisfactory results without pulmonary impairment, long-term deformation and scoliosis-development, but long-term follow-up is still ongoing.

Keywords: Chest wall resection; childhood tumors; pediatric tumors; pediatric thoracic surgery; case series

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Introduction

Malignant chest wall tumors in children and adolescents are rare and comprehend a heterogeneous variety of histopathological entities with different biological behavior (1,2).

Management of these tumors needs an aggressive approach, and a multidisciplinary treatment is planned according to the malignancy of the tumor (3). For every patient, a different treatment-schedule is programmed, and it may include induction chemotherapy for reduction

^ ORCID: Paolo Scanagatta, 0000-0002-1619-6453; Francesco Inzirillo, 0000-0002-1661-3093; Stefano Sestini, 0000-0003-2274-9293; Giuseppe Naldi, 0000-0001-8582-1606.

Table 1 Clinical characteristics of patients

Case	Gender (F/M)	Age (years)	Histology	Year of surgery	Resection type	Length of surgery (min)	Time to discharge (POD)	Follow-up (months)
Case 1	M	18	CS	2019	Sternal body	220	13	30
Case 2	M	12	OS	2017	2nd, 3rd and 4th rib	120	7	12†
Case 3	F	18	SS	2020	2nd and 3rd rib + left upper lobectomy	180	8	23
Case 4	M	14	GCT	2018	6th and 7th rib + 6th, 7th and 8th intercostal space	130	5	52
Case 5	M	17	SS	2020	8th, 9th and 10th rib + diaphragm	200	16	21
Mean		15.8				170	9.8	27.6

†, dead of disease. F, female; M, male; CS, chondrosarcoma; OS, osteosarcoma; SS, synovial sarcoma; GCT, giant cell tumor; POD, postoperative days.

of local disease, followed by surgical resection and reconstruction, radiotherapy for local control and adjuvant chemotherapy to prevent distant metastases.

Nowadays the improvement in surgical techniques, pharmacological therapy, and critical care, opens the possibility to perform more aggressive and extensive surgical resections. In fact, the asportation of a substantial portion of the chest wall often necessitates the use of prosthetic materials to restore the thoracic shape and rib-cage functionality. Although it could be complex in adults, in children this necessity represents a greater challenge. The future growth must be considered and impact significantly on the strategy adopted and materials used, to warrant protection of vital organs and maintain the physiologic chest excursion in the future stages of patient life (4).

This paper presents few cases recently treated using a multidisciplinary approach, describing a modified, original "semi-rigid" technique of reconstruction of large chest-wall defects after surgical resection. The aim of this study is to understand the preliminary findings of this type of reconstruction, following the hypothesis it could be useful for reducing the subsequent onset of scoliosis or rib cage deformity.

Case presentation

In this paper we retrospectively reviewed a non-consecutive series of 5 patients who underwent surgery for chest wall tumors of childhood and adolescent age, during a 5-year period (*Table 1*).

The patients were treated by the same oncological and surgical team in a single comprehensive cancer-center, from 2017 and 2020; patient records and data were recorded and kept up-to-date through outpatient service and phone call from the time of discharge through May 2022.

All the patients were diagnosed by Tru-Cut biopsy or surgical biopsy and included in multimodal treatment schedule. A complete preoperative staging was performed, using computed tomography scan, magnetic resonance imaging, and in some cases bone scintigraphy and/or fluorodeoxyglucose positron emission tomography (FDG-PET/CT) to evaluate the size of the tumor and its local extension in terms of involvement of contiguous structures.

Every case was discussed by our multidisciplinary team board, which includes surgeons, oncologists, radiation therapists, histopathologists, psychologists.

The operation consisted in resection of the tumor with radical purpose achieving large surgical margins and the removal of all adjacent involved structures, followed by chest wall reconstruction. After having previous experiences with rigid and semi-rigid prosthesis, which are a gold standard in adult patients, but having experienced some chest wall deformation due to the physiological growth in pediatric population, the author (PS) decided to maintain the high standard of radicality, trying to reduce the stiffness of the prosthetic materials and modifying the technique of the reconstructive procedure, in the aim of minimizing long term complications such as scoliosis and rib cage deformity.

The defect obtained from the resection were repaired with mesh positioning, using four layers of Knitted Polypropylene mesh (a double-layer of a folded mesh); in

Table 2 Relevant articles on semi-rigid chest wall reconstruction

Author, year	Medium age (y)	Number of patients	Prosthesis	Follow-up	Scoliosis
Tuggle DW *et al*. 2004, (5)	7	4	BP*	12.5 months	n/a
Soyer T *et al*. 2006, (6)	7.6	17	Various	n/a	6%
Lin SR *et al*. 2012, (7)	15.7	5	Permacol	23 months	60%
Oliveira C. *et al*. 2012, (8)	2.5 months	3	Surgisis	32 months	66%
Dingemann C *et al*. 2012, (9)	10.6	8	Various	37.5 months	33%**
Makarawo TP *et al*. 2015, (10)	13	1	BP	n/a	n/a
Guillén G *et al*. 2017, (11)	10.6	8	BP	39.6 months	37%
Lopez C *et al*. 2017, (12)	14	44	Various	n/a	11%
Saltsman JA *et al*. 2021, (13)	15.6	76	Various	n/a	25%
Basharkhah A *et al*. 2021, (14)	10.9	13	Various	8.8 years	15%
Scanagatta P *et al*. 2022, (present case series)	15.8	5	DFKPM***	27.6 months	0%

*, bioresorbable plate; **, 2/6 patient was considered since 2 patients had insufficient follow-up due to early complications leading to death; ***, DFKPM: double layer of folded Knitted Polypropylene mesh. n/a, data or information not available.

two cases a local (latissimum dorsi) muscle transposition was performed.

Then, a narrative review of the literature was performed through an electronic search for relevant studies from January 1990 to April 2022 in the following databases: PubMed, Science Direct, Embase, Scopus and Cochrane. The results are summarized in *Table 2* and analyzed in the discussion section.

The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). Written informed consents were obtained from the patients for publication of this case series and accompanying images. For patients under 18 years informed consents were obtained from the parents. Copies of the written consent are available for review by the editorial office of this journal.

This is a retrospective study on patients operated following usual clinical indications, using registered and authorized materials and instruments; the peculiar variations of the surgical technique are merely technical: they do not affect the scope of the surgical procedure and they do not rise any ethical concern. Thus, ethics board approval is waived.

Case 1

An 18-year-old male was referred for a growing mass of the sternum; the chest CT scan revealed a well-circumscribed lesion measuring 6 cm × 7.5 cm of the body of the sternum. We performed a trough-cut biopsy that described the lesion as a chondrosarcoma G1. The PET-FDG demonstrated the sternal mass as the only localization, without local or distant metastases; the multidisciplinary team board decided for surgical resection. We performed a resection of the sternum en-bloc with bilateral 3rd, 4th, 5th, 6th rib and costal arch medial aspect, along with overlaying muscles and skin (*Figure 1*). Reconstruction was performed with two independent layers of folded non absorbable Knitted Polypropylene mesh (MicroVal 2D Mesh, Saint-Just-Malmont, France) fixed by Polypropylene 1 stiches. A single chest tube was positioned in the right pleural space. To complete the reconstruction a muscular-skin flap of latissimus dorsi was performed. The patient was discharged after 13 days without any major complication. The respiratory function and chest mechanic was normal. After 30 months the patient is well and disease-free.

Case 2

A 12-year-old male affected by metastatic osteosarcoma originating from the left humerus. The patient underwent chemotherapy, surgery of the primary tumor, sequential bilateral pulmonary metastasectomy. After an 8-month disease free interval he developed a metachronous metastasis of the anterior arch of the left 3rd rib. After three months of second-line chemotherapy he underwent to resection of the 2nd–3rd and 4th rib along with surrounding soft

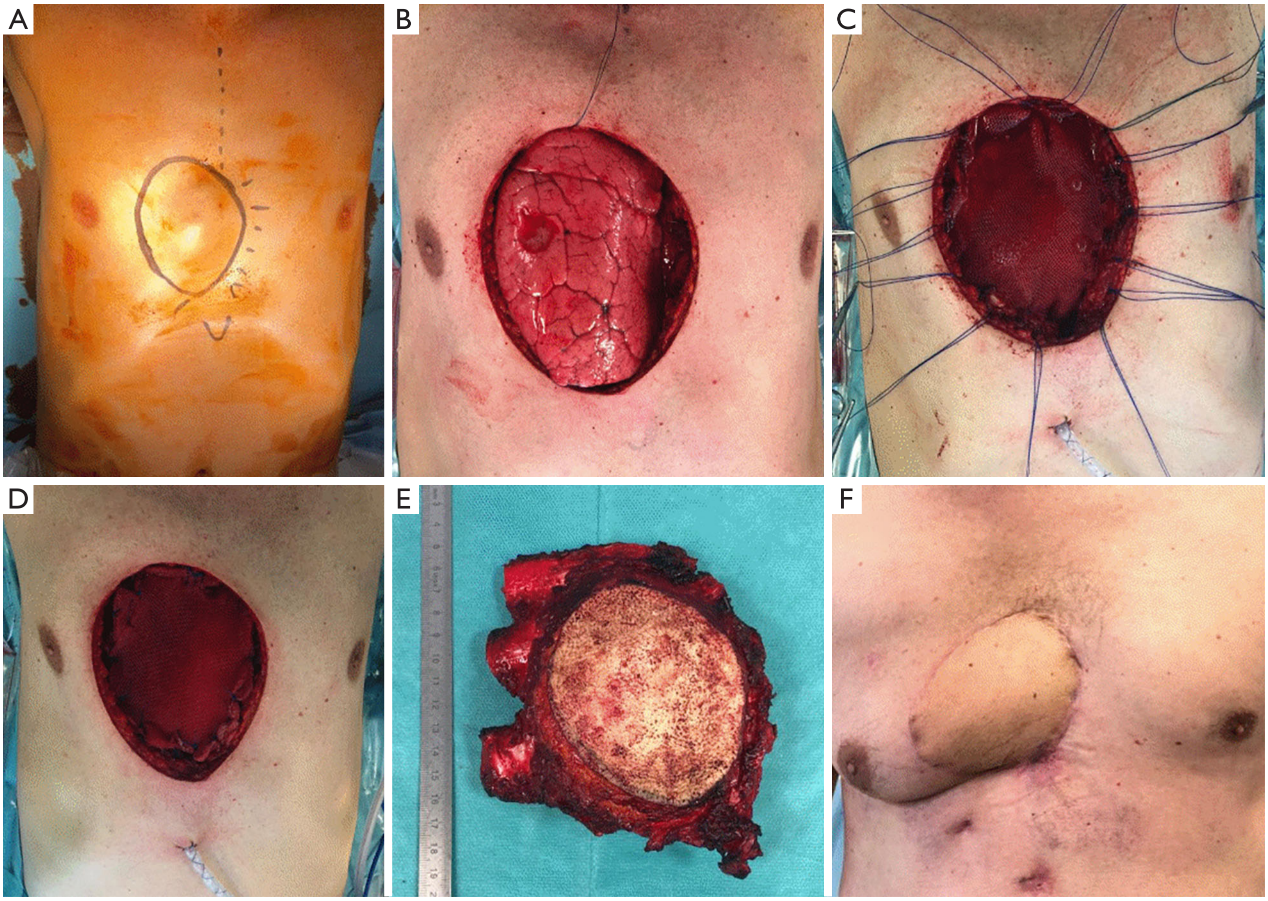

Figure 1 An 18-year-old male affected by a chondrosarcoma G1 of the sternum (more details in the text, Case 1). (A) Patient positioned on the operatory table and draw of the starting incision. (B) Patient after the complete resection of sternal body, the right lung herniated due to ventilation. (C) Reconstruction phase, fixation of inner layer of folded mesh, stitch wires preserved for fixation of the outer layer. (D) Outer layer of folded mesh positioned, ready for transposition of latissimus dorsi pedicled flap. (E) Detail of specimen. (F) Patient at 30-day follow-up.

tissue (skin and pectoralis major muscle were preserved). Reconstruction was performed using two independent layers of folded non absorbable Knitted Polypropylene mesh (MicroVal 2D) fixed by Polypropylene 1 stiches. Postoperative period was uneventful (the patient was discharged on seventh postoperative day). After 40 days from surgery adjuvant sequential Radiotherapy (54 Gy in 20 fractions) was administered. The patient had a pulmonary and skeletal recurrence after 4 months, then he was treated with further chemotherapy and radiotherapy but died with disease after 12 months.

Case 3

An 18-year-old female presented to the emergency room of secondary hospital for a growing mass located deeply under the left pectoral muscle and breast. After a CT scan showing a tumoral mass arising from the 2nd and 3rd rib and involving the left lung deeply to the hilum. We executed a surgical biopsy showing localization of a Synovial sarcoma. We presented the case at the multidisciplinary board, and she performed induction chemotherapy for 4 months obtaining a good partial-response. The re-staging scan showed partial response and reduction of the tissue particularly in the lung (the left lower lobe was no longer involved). We performed a resection of the anterior aspects of 2nd and 3rd rib along with first and third intercostal spaces and extrapleural left upper lobectomy. Reconstruction was performed with folded Knitted Polypropylene mesh (MicroVal 2D), fixed on the 1st left rib and passed and turned over the 4th rib (both

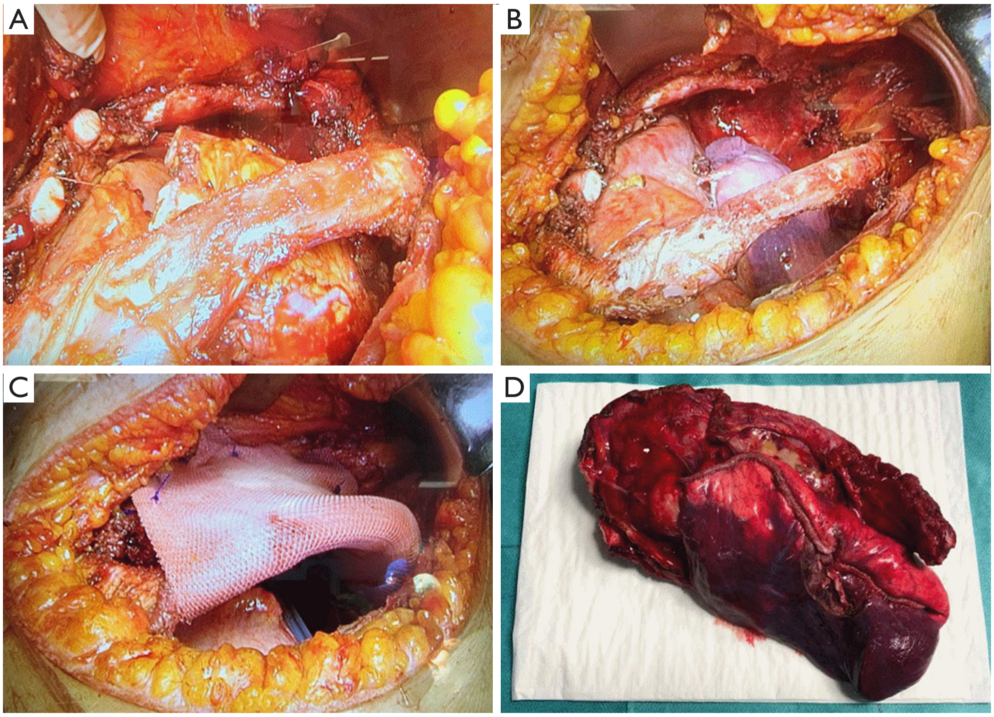

Figure 2 An 18-year-old female affected by a Synovial sarcoma of the anterior left chest wall, after induction treatment (more details in the text, Case 3). (A,B) Completed chest wall resection of 2nd and 3rd rib along with left upper lobectomy (before and after asportation of specimen from the chest cavity). (C) Reconstruction complete; the folded mesh is fixed by stiches to 1st rib and chest wall, the lower portion has been turned around the 4th rib. (D) Detail of specimen.

ribs were freed from soft-tissue, according to the "bird-cage technique"), using it to separate the two layers (*Figure 2*). The postoperative stay were 8 days without complication. The respiratory function and chest mechanic was normal. The patient is well and without disease after 23 months from surgery.

Case 4

A 14-year-old male was referred with diagnosis of giant cell tumor of the anterior arch of the 7th right rib. The multidisciplinary team confirmed surgical approach as the first treatment. A wide resection of the rib was performed, along with the soft tissue of the 6th and 8th intercostal space and the lateral aspect of the 6th rib. Reconstruction was performed with folded Knitted Polypropylene mesh (MicroVal 2D), fixed on the right 5th rib and passed and turned over the 8th rib (both ribs were freed from soft-tissue, according to the "bird-cage technique") (15).

The patient was discharged after 5 days, without any complication. The patient is disease-free 52 months after surgery, without any evident chest wall deformation.

Case 5

A 17-year-old male with a story of a primitive synovial sarcoma of the left hemithorax treated with induction chemotherapy. After restaging we observe a good partial response, with residual disease located in the left lateral costo-diaphragmatic recess. After multidisciplinary team discussion we performed a resection of lateral aspects of the 8th, 9th and 10th rib along with a substantial portion of the diaphragm (*Figure 3*). Reconstruction was performed with folded knitted Polypropylene mesh (MicroVal 2D) fixed by Polypropylene 1 stitches: the upper margin passed and turned around the 7th rib, the lower part was fixed posteriorly and anteriorly to the residual aspects of 8th, 9th and 10th rib, and to the entire 11st left rib and diaphragmatic margins inferiorly, the diaphragm was reconstructed using a portion of inner layer of the folded prosthesis fixed by Polypropylene 1 and Maxon 1 stitches. Histopathologic examination confirmed a wide-margin free surgery. The

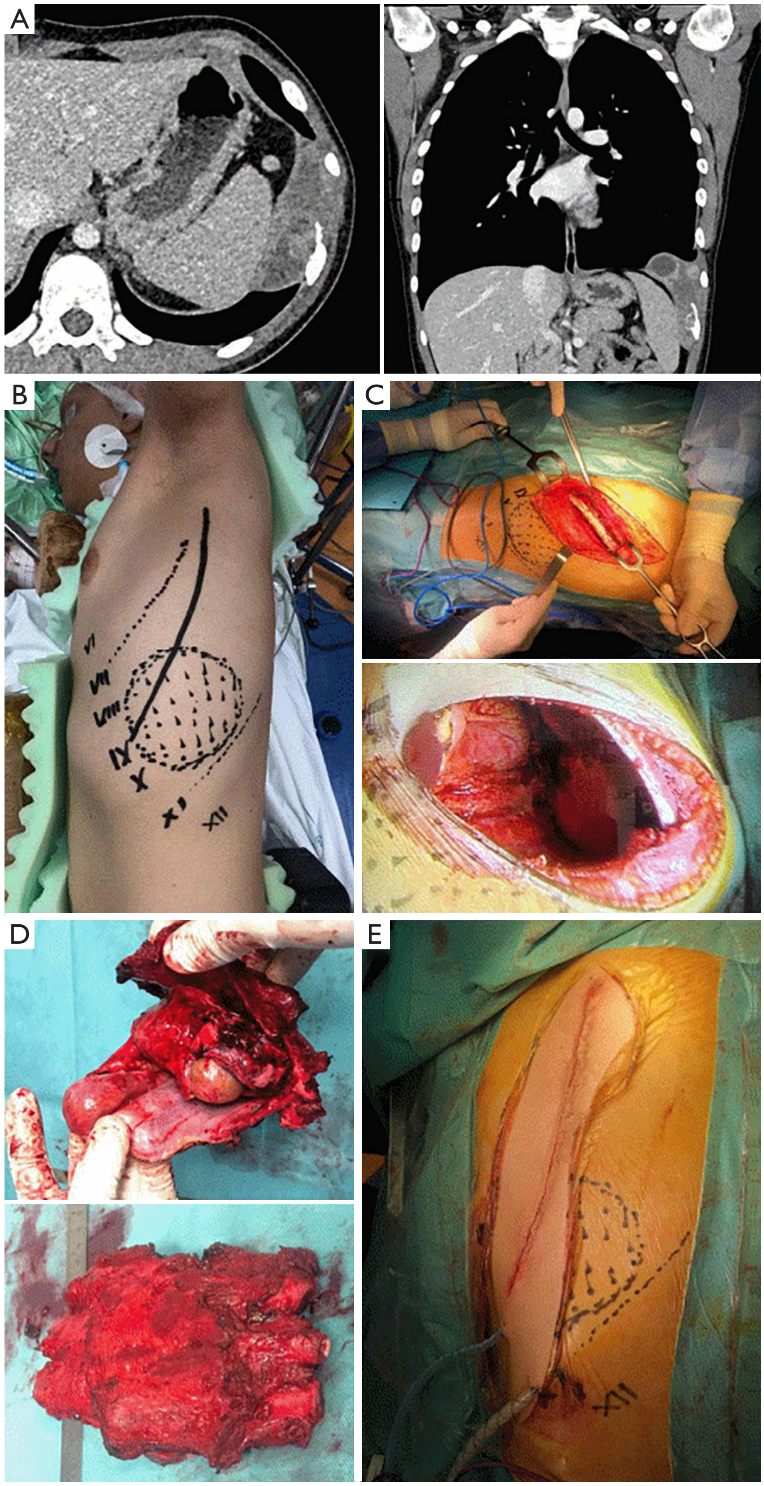

Figure 3 A 17-year-old male with a primitive synovial sarcoma of the left hemithorax treated with induction chemotherapy (more details in the text, Case 5). (A) Two images from preoperatory CT scan showing the tumoral mass in lateral costodiaphragmatic recess. (B) Patient preoperative positioning: the starting incision and mass location has been sketched on the skin. (C) Two images of surgical intervention, before and after chest wall resection. (D) Detail of specimen, resection of 8th, 9th and 10th rib along with diaphragm (the costodiaphragmatic recess). (E) End of the procedure. CT, computed tomography.

postoperative stay was uneventful, and the patient was discharged on 16th postoperative day. The respiratory function and chest mechanic were normal, and the patient is well after 21 months from surgery.

Discussion

Literature on chest wall reconstruction in children and adolescents has grown in the last few decades but remain scarce, and, due to the rarity of the pathologies, there are very few series reporting more than a dozen of patients.

Since the first description of a chest-wall reconstruction in 1880, multiple types of prosthesis have been used, but after more than 140 years there seems to be no consensus on the ideal material for pediatric population, which could be considered "per se" a challenging and demanding population (3,16).

The last two decades had seen a progressive shift from rigid and metallic prosthesis toward more flexible materials, mostly for the reports of progressive deformations and scoliosis after surgical resection and reconstruction of the chest wall in children and adolescents (9,11,13,17).

Dingemann *et al.* (9) demonstrated in their experience of 8 patients reconstructed mostly using flexible meshes, that surgical reconstruction after resection of malignant thoracic wall tumors using non-rigid prosthetic material is safe and effective in children and adolescents, and the use of rigid prosthetic material provides stability, but there is a risk of dislocation.

Scoliosis represents a long-term complication after chest wall reconstruction and should be monitored in routine follow-up: the patients should undergo regular orthopaedic surveillance postoperatively (13,17).

The largest study in literature (13) evaluated 76 patients treated with various combinations of reconstructive materials, suggesting that 1 out of 4 patients could develop a progressive scoliosis after surgery.

In fact, Glotzbecker *et al.* (17) in their case-series, described 11 patients developing progressive scoliosis after chest wall resection, 9 of them following semi-rigid reconstruction with Gore-Tex (which is quite a flexible, but anelastic material). The authors themselves stated that Gore-Tex may have an influence on the development of scoliosis, but no conclusions can be drawn because larger, more complicated resections are more likely to require a Gore-Tex reconstruction.

Recently some authors reported their experience with bioresorbable plate, which could be a good addition to other available methods to reconstruct the chest wall in children, even if further studies are needed (5,10,11). Relevant papers have been summarized in *Table 2* (5-14).

All these studies report the surgical experience with different type of reconstruction, from rigid to semi-rigid to direct closure, and in fact, the diversity of procedures, location and degree of resection performed in this population does confound the results. Moreover, several patients do not have follow-up to maturity, which is an important endpoint when considering their overall risk in developing scoliosis. Saltsman *et al.* (13) concluded that "*Age, type of chest wall reconstruction, and tumor size are not associated with scoliosis development*", but one can argue that population, type of resection and reconstructions vary too much to allow a proper analysis of possible independent risk factor.

Again, the finding of Glotzbecker *et al.* (17) is particularly interesting: "*… Seven of the eight patients with resections that included a rib superior to the sixth rib developed scoliosis, while four of 13 with resections below the sixth rib developed scoliosis…*". This "upper chest wall resection" was confirmed an independent factor to develop scoliosis with logistic regression analysis. The mechanism could be related to the force applied to the column, which increases to the side opposite to the chest-wall resection, particularly in the upper portion of the thorax.

In fact, the forces applied to the prosthesis is loaded to the column and the risk of developing scoliosis has been described both in clinical and in experimental settings (18,19) and we also observed some challenging cases in our previous experience with stiffer semi-rigid prosthesis, even with association of prosthetic displacement (an "extreme" example is shown in *Figure 4*).

Our case-series involves a pediatric population between 12 and 18 years, a period of life which body is growing, so implanting a completely rigid structure could lead to chest-wall deformation.

A solution should consider a material which have to be "stable but not rigid" as well as "flexible but not limp". The use of a double layer of folded Knitted Polypropylene mesh granted a rigidity sufficient to avoid paradoxical respiration (further reduced by the double layer structure) without impairing the growth of the chest-wall in the medium and long term and applying a four-layer mesh halved by a subcentimetric interspace.

Probably, the space between the two layers allows the independent movement of each stratum, just like an inner tube inside a tire. The mesh used is made of Knitted Polypropylene arranged to constitute a net, resulting very

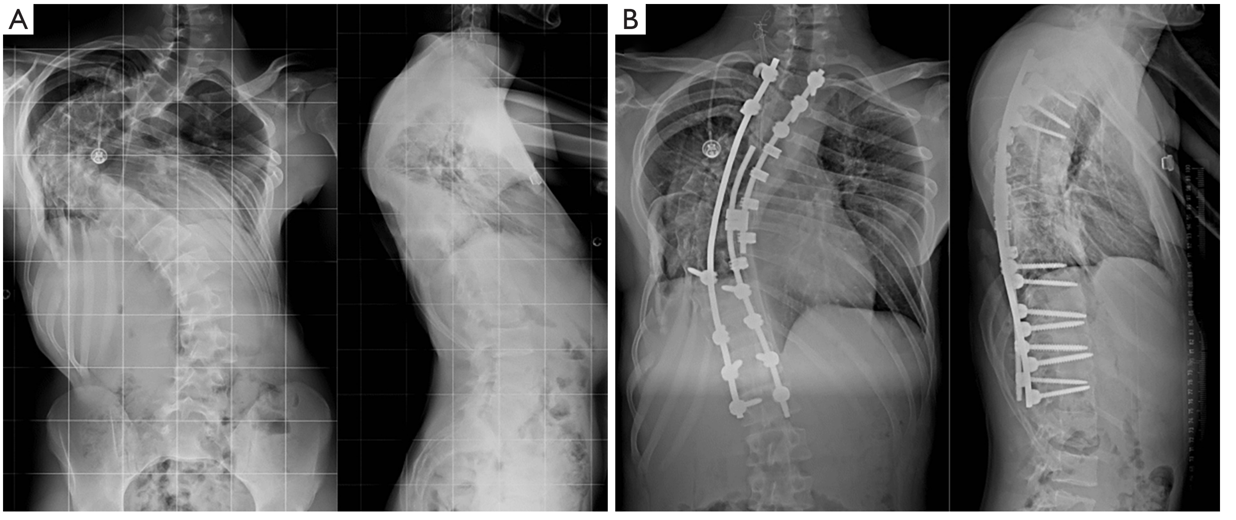

Figure 4 A case of severe scoliosis developed in a 17-year-old male three years after a 4-rib (6th to 9th) right chest-wall resection and semi-rigid Marlex-methacrylate reconstruction for Ewing's sarcoma. The patient presented ongoing dyspnea after 36 months from prosthetic chest-wall reconstruction. (A) Column X-ray: the radiogram shows severe scoliosis with chest wall deformation. (B) Radiograms after vertebral stabilization. The patient had a good cosmetic result and gained 5 cm in height.

permeable to corporeal fluids, and allowing immunity cells to move between the layer, reducing the possibility of infection, and the formation of seroma even without a superficial drainage tube.

This kind of structure make the fixation quite easy, the stitches used to fixate the inner folded layer, can be utilized for the outer layer even without the needle. We suggest fixing the prosthesis all around the defect (as illustrated in case 1) to reduce the tension applied to every single stitch. When the resection involves complete ribs, the folded mesh allows even to fix the prosthesis to a single edge, bending the mesh around a properly freed rib (as illustrated in case 3). This technique allows a perfect distribution of forces between the inner and outer layer, balancing tension and reducing parietal distress.

The strengths of our study are mainly the use of a stable, elastic and flexible material, which could adapt to the patients' growing, which is an important and often underestimated open issue in children and adolescents who need a chest wall resection, mainly for aggressive tumors. The first results are encouraging, not having observed scoliosis or chest wall deformation at the moment. The limitations are the small number of this retrospective, single center, non-consecutive case series, with a heterogenous population and quite different cases treated. Obviously we are aware of the existence of a selection bias in this study, but this is intrinsically a patient tailored surgery. In the short period this type of surgery seems to be feasible, and we do not observe major postoperative complications in the five patients treated up to now. A large series with a longer follow-up must be obtained to consolidate long-term results. Another limitation is related to the extent of the resection, being this kind of reconstruction indicated for defect up to four ribs or resection of sternal body, and also with the possibility of repair a diaphragmatic defect; however, for greater resection it should be associated to a rigid material (homologous or heterologous bone graft, titanium, resin, synthetic material) to warrant stability and protection of the reconstructed chest wall.

Conclusions

Pediatric patients need a peculiar attention, planning the reconstruction strategy has a key role in future growth. This original surgical technique using a "double folded, double layer non-absorbable, permeable mesh", seems to be a promising technique if compared to other rigid and semi-rigid type of reconstructions.

The flexibility of the prosthesis reduces the tension applied to the residual chest wall allowing a better distribution of forces and decreasing the stress on the column, and we are confident that this should provide a reduction in the risk of scoliosis in the long term.

The thickness of the chest wall reconstructed this way simulates the normal one and allows a better support, particularly inward, avoiding paradoxical movement during inspiration by reducing the negative pressure applied to muscular portion of the chest wall, reducing the impairment of respiratory functionality: this will be studied through a planned dynamic magnetic resonance during normal and forced respiration, in the hypothesis that the positioning of an inner double layer and an outlet double layer, separated by a few millimeter space, consents a more physiological movement with every chest expansion.

The permeability of the material allows a better movement of fluids and immunological cells between every different layer, potentially reducing the occurrence of postoperative infections.

We currently consider this technique an option for pediatric reconstruction involving resection of sternum and chest-wall when 2 to 4 ribs must be removed, even if planning such a reconstruction should be carefully studied case by case, in a tailored fashion.

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Footnote

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://asj.amegroups.com/article/view/10.21037/asj-22-22/coif). PS serves as an unpaid associate Editor-in-Chief of *Pediatric Medicine* (AME Publishing Company) from July 2022 to June 2024. The series "New Materials for Reconstruction in Thoracic Surgery" was commissioned by the editorial office without any funding or sponsorship. PS reports personal fees from Baxter International, outside the submitted work. The authors have no other conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). This is a retrospective study on patients operated following usual clinical indications, using registered and authorized materials and instruments; the peculiar variations of the surgical technique are merely technical: they do not affect the scope of the surgical procedure and they do not rise any ethical concern. Thus, ethics board approval is thus waived. Written informed consents were obtained from the patients for publication of this case series and accompanying images. For patients under 18 years informed consents were obtained from the parents. Copies of the written consent are available for review by the editorial office of this journal.

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