Pediatric Surgery International (2024) 40:53
https://doi.org/10.1007/s00383-024-05635-x

ORIGINAL ARTICLE

Congenital lung malformations: a nationwide survey on management aspects by the Italian Society of Pediatric Surgery

Luca Pio[1,2] · Valerio Gentilino[3] · Francesco Macchini[4] · Alberto Attilio Scarpa[5] · Roberto Lo Piccolo[6] · Andrea Conforti[7] · Alberto Ratta[8] · Riccardo Guanà[9] · Francesco Molinaro[10] · Sara Costanzo[11] · Giovanna Riccipetitoni[12,13] · Gabriele Lisi[14] · Paola Midrio[15,16] · Francesca Tocchioni[17] · Giovannii Cobellis[18] · Andrea Volpe[19] · Elisa Zolpi[20] · Anna Morandi[21] · Enrico Ciardini[22] · Claudio Vella[23] · Maria Giovanna Grella[24] · Maria Sergio[25] · Edoardo Guida[26] · Lorenzo Nanni[27] · Silvia Ceccanti[28] · Vincenzo Di Benedetto[29] · Maurizio Cheli[30] · Alfredo Garzi[31] · Maria Nobili[32] · Valeria Gabriele[33] · Giovanni Boroni[34] · Filippo Incerti[35] · Nicola Zampieri[36] · Sebastiano Cacciaguerra[37] · Pier Luca Ceccarelli[38] · Maria Escolino[39] · Vito Briganti[40] · Davide Gori[41] · Ciro Esposito[39] · Piergiorgio Gamba[19,42] · Fabrizio Gennari[9] · Alessandro Inserra[43,44] · Patrizia Dall'Igna[45,46] · Carmelo Romeo[47] · Pietro Bagolan[7,44] · Cosimo Bleve[20] · Fabio Chiarenza[20] · Francesco Morini[17] · Gloria Pelizzo[11] · Michele Torre[1]

Accepted: 7 January 2024

Abstract

Introduction Over the years, congenital lung malformations (CLM) management remains a controversial topic in pediatric thoracic surgery. The Italian Society of Pediatric Surgery performed a national survey to study the current management variability among centers, trying to define national guidelines and a standardized approach of children with congenital lung malformations.

Methods Following a National Society approval, an electronic survey including 35 items on post-natal management was designed, focusing on surgical, anesthesiology, radiology and pneumology aspects. The survey was conducted contacting all pediatric surgical units performing thoracic surgery.

Results 39 pediatric surgery units (97.5%) participated in the study. 13 centers (33.3%) were classified as high-volume (Group A), while 26 centers (66.7%) were low volume (Group B). Variances in diagnostic imaging protocols were observed, with Group A performing fewer CT scans compared to Group B ($p=0.012$). Surgical indications favored operative approaches for asymptomatic CLM and pulmonary sequestrations in both groups, while a wait-and-see approach was common for congenital lobar emphysema. Surgical timing for asymptomatic CLM differed significantly, with most high-volume centers operating on patients younger than 12 months ($p=0.02$). Thoracoscopy was the preferred approach for asymptomatic CLM in most of centers, while postoperative long-term follow-up was not performed in most of the centers.

Conclusion Thoracoscopic approach seems uniform in asymptomatic CLM patients and variable in symptomatic children. Lack of uniformity in surgical timing and preoperative imaging assessment has been identified as key areas to establish a common national pattern of care for CLM.

Keywords Pediatric thoracic surgery · CPAM · Congenital lung malformations · MIS · Thoracoscopic surgery · CLM

Introduction

Congenital lung malformations (CLM) pose significant challenges for pediatric surgeons and are considered one of the most pertinent thoracic diseases. The estimated incidence of congenital laryngeal malformation (CLM) has been recently revised to 1 in 7200 live births, however, some studies suggest that the actual incidence might be higher due to limitations in previous epidemiological studies caused by prenatal ultrasound technology [1, 2]. The rarity of CLM makes it difficult to gather scientific evidence regarding surgical management, including indications, timing, and approach, leading to a lack of national and international guidelines. Collaborative efforts across multiple centers have been made to generate scientific evidence on CLM management, aiming to define core outcomes and enhance our understanding of

Extended author information available on the last page of the article

Published online: 10 February 2024

the underlying pathophysiological processes [3, 4]. However, recent studies have revealed a lack of uniformity in management practices within the international pediatric surgery community [5]. Among the various approaches to CLM management, thoracoscopic techniques have gained popularity over the years due to accumulating evidence supporting their advantages in terms of musculoskeletal sequelae and postoperative outcomes compared to traditional thoracotomy [6, 7]. Nonetheless, our knowledge about national implementations of thoracoscopic procedures, as well as the pre- and postoperative settings that influence and shape the care patterns of minimally invasive CLM approaches, remains limited.

To address this gap, the Italian Society of Pediatric Surgery conducted this study to evaluate the existing variability in CLM management among different centers in Italy. The aim was to analyze the current discrepancies in national guidelines and explore the implications for the development of minimally invasive surgical techniques on a national level.

Methods

This study utilized Google Forms® (Google LLC Mountain View, California, USA) as it is designed to enable online completion, receiving approval from the review board of the Italian Society of Pediatric Surgery directorate.

A nationwide survey was conducted, targeting the surgical unit directors of all Italian centers specialized in thoracic surgery. The survey collected responses from the professionals in charge for thoracic surgery procedures at each respective center.

The survey has been kept open for a duration of three months (August to October 2022), with the reported data collected in November 2022.

The questionnaire consisted of three parts. The first part comprised general questions about the institutional setting and preoperative management of CLM (Table 1), the second part comprised perioperative management questions (Table 2), and the third part comprised postoperative follow-up settings questions (Table 3).

To analyze the nationwide variability in CLM management, a comparison was made between high-volume centers (Group A) and low-volume centers (Group B). The criterion for defining low-volume centers was a minimum of four procedures per year, equating to one procedure for each quarter. This threshold was based on data published by the European Pediatric Surgeons Association in 2018, which indicated that most surgeons performed fewer than five procedures per year [5].

Categorical variables were summarized as frequencies and percentages. A Pearson Chi2 test was used to analyze correlation of the different variables with the dichotomous volume variable. The significance level was set to $p < 0.05$.

Results

A total of 39 pediatric surgery units, representing 97.5% of the 40 Italian centers specializing in thoracic surgery for children, participated in the study.

Among these centers, 13 (33.3%) were classified as high-volume centers (Group A), where at least four procedures per year were performed, while the remaining 26 pediatric surgical units (66.7%) were considered low-volume centers and categorized as Group B.

The analysis of preoperative management revealed differences in the diagnostic imaging protocols between the two groups. Group A conducted a single computed tomography (CT) scan, whereas Group B performed twice as many CT scans ($p = 0.012$). Fetal magnetic resonance imaging (MRI) was conducted in 79.5% of the centers, regardless of their case volume, while preoperative MRI assessment was performed in 46.2% of centers (Table 1).

Surgical indications favored operative approaches for asymptomatic congenital cystic lung malformations, hybrid forms, extra lobar, and intralobar sequestration, regardless of the case volume in both high and low-volume centers. However, a wait-and-see approach was commonly adopted for cases of congenital lobar emphysema, with surgery only considered if symptoms were present.

Significant differences were observed in surgical timing for asymptomatic congenital cystic lung malformations. Most high-volume centers operated on patients younger than 12 months ($p = 0.02$). No differences were found in terms of surgical team composition and the surgical environment, as the majority of CLM cases were managed by pediatric surgeons (74.4%) in general hospital settings (61.5%) (Table 1).

The analysis of perioperative management revealed a national consensus among the centers, regardless of their case volume. Thoracoscopy was the preferred approach for asymptomatic CLM in the majority of centers (64.1%), while there was no standardized approach for symptomatic patients in 66.7% of centers. Lobectomy was considered the primary choice for CLM resection in 71.8% of centers. Most centers performed CLM surgeries under single lung ventilation (43.6%), followed by selective lung intubation (33.3%) and bipulmonary ventilation (23.1%). Postoperative recovery took place in pediatric surgical wards in 59% of centers, while 41% of children spent the immediate postoperative period in intensive care units (Table 2). Postoperative follow-up involved systematic imaging (CT or MRI) in 59% of centers, and pulmonary function tests were conducted in 76.9% of centers. However, the transition of care and long-term follow-up was not consistently provided by most centers (87.2%). There were

Table 1 Preoperative management

	High-volume centers ($N=13$) *n* (*n/N%*)	Low-volume centers ($N=26$) *n* (*n/N%*)	Total centers ($N=39$)	*P*-value
Fetal MRI				0.16
Yes	12 (92.3%)	19 (73.1%)	31 (79.5%)	
No	1 (7.7%)	7 (26.9%)	8 (20.5)	
Preoperative MRI				0.46
Yes	5 (38.5)	13 (50%)	18 (46.2%)	
No	8 (61.5%)	13 (50%)	21 (53.8%	
Number of preoperative CT				0.04
1	12/12 (100%)[a]	16/25 (64%)[b]	28/37 (75.7%)	
2	0/13 (0%)	9/25 (36%)	9/38 (24.3%)	
Surgical indication for extralobar sequestration				0.30
Surgery for all patients	13 (100%)	24 (92.3%)	37 (94.9%)	
Wait and surgery if symptoms	–	2 (7.7%)	2 (5.1%)	
Surgical indication for intralobar sequestration				0.20
Surgery for all patients	13 (100%)	23 (88.5%)	36 (92.3%)	
Wait and surgery if symptoms	–	3 (11.5%)	3 (7.7%)	
Surgical indication for hybrid pulmonary malformations				0.47
Surgery for all patients	13 (100%)	25 (96.2%)	38 (97.4%)	
Wait and surgery if symptoms	–	1 (3.8%)	1 (2.6%)	
Surgical indication for congenital Emphysema				1
Surgery for all patients	4 (30.8%)	8 (20.0%)	12 (30.8)	
Wait and surgery if symptoms	9 (69.2%)	18 (73.3%)	27 (69.2%)	
Surgical indication for asymptomatic congenital cystic lung malformations				0.20
Surgery for all patients	13 (100%)	23 (88.5%)	36 (92.3%)	
Wait and surgery if symptoms	–	3 (11.5%)	3 (7.7%)	
Surgical timing for asymptomatic congenital cystic lung malformations				0.02
< 12 months	12/13 (92.3%)	13/23 (56.5%)	25/36 (69.4)	
≥ 12 months	1/13 (7.7%)	10/23(43.5%)	11/36 (30.6%)	
Surgical team				0.79
Pediatric surgeons	10 (76.9%)	19 (73.1%)	29 (74.4%)	
Pediatric surgeon and adult thoracic surgeons	3 (23.1%)	7 (26.9%)	10 (25.6%)	
Surgical environment				0.48
Pediatric hospital	6 (11.1%)	9 (20.0%)	15 (38.5%)	
General hospital	7 (5.6%)	17 (6.7%)	24 (61.5%)	

CT computed tomography, *CLM* congenital lung malformation

[a]1 High-volume center did not perform preoperative CT scan

[b]1 Low-volume center did not perform preoperative CT scan

no differences observed between the two groups regarding postoperative follow-up (Table 3).

Discussion

Congenital pulmonary airway malformations (CLM) pose significant challenges in pediatric thoracic surgery. This study aimed to evaluate the current management variability of CLM in Italian centers and analyze its implications for the development of minimally invasive surgery at a national level. The results provide valuable insights into existing practices, highlighting both areas of discrepancy and uniformity among different centers.

A high response rate of 97.5% indicates strong participation and enhances the representativeness of the findings. The analysis compared high-volume and low-volume centers to assess any variations based on case volume. Considering the

Table 2 Perioperative management

	High volume (*N* = 13) *n* (*n*/*N* %)	Low volume (*N* = 26) *n* (*n*/*N* %)	Total (*N* = 39)	*P*-value
Surgical approach for asymptomatic CLM				0.81
Thoracoscopy, as a standardized approach	8 (61.5%)	17 (65.4%)	25 (64.1%)	
Thoracotomy or thoracoscopic, without a standardized approach	5 (38.5%)	9 (34.6%)	14 (35.9%)	
Surgical approach for symptomatic CLM				0.39
Thoracoscopic, as a standardized approach	3 (23.1%)	7 (27%)	10 (25.6%)	
Thoracotomic, as a standardized approach	–	3 (11.5%)	3 (7.7%)	
Thoracotomic or thoracoscopic, without a standardized approach	10 (76.9%)	16 (61.5%)	26 (66.7%)	
Type of surgery				0.61
Lobectomy, as a standardized approach	10 (76.9%)	18 (69.2%)	28 (71.8%)	
Lobectomy or wedge resection, without a standardized approach	3 (23.1%)	8 (30.8%)	11 (28.2)	
Type of ventilation				0.07
Bipulmonary ventilation	2 (15.4%)	7 (26.9%)	9 (23.1%)	
Selective pulmonary intubation	2 (15.4%)	11 (42.3%)	13 (33.3%)	
Endobronchial blocker	9 (69.2%)	8 (30.8%)	17 (43.6%)	
Postoperative recovery				0.11
Intensive care unit	3 (11.1%)	13 (50%)	16 (41%)	
Pediatric surgery ward	10 (77.8%)	13 (50%)	23 (59%)	

CT computed tomography, *CLM* congenital lung malformation

Table 3 Postoperative management

	High volume (*N* = 13) *n* (*n*/*N* %)	Low volume (*N* = 26) *n* (*n*/*N* %)	Total (*N* = 39)	*P*-value
Postoperative imaging (CT/MRI)				0.82
Systematically performed	8 (61.5%)	15 (57.7%)	23 (59%)	
Not performed	5 (38.5%)	11 (42.3%)	16 (41%)	
Postoperative pulmonary function tests				0.11
Systematically performed	12 (92.3%)	18 (69.2%)	30 (76.9%)	
Not performed	1 (7.7%)	8 (30.8%)	9 (23.1%)	
Transition care program				0.17
Provided with adult pneumologist	3 (23.1%)	2 (7.7%)	5 (12.8%)	
Not established	10 (76.9%)	24 (92.3%)	34 (87.2)	

CT computed tomography, *MRI* magnetic resonance imaging

epidemiology of CLM and the number of live births in Italy in 2022, which was 393,000 (http://dati.istat.it/?lang=en), the estimated annual number of CLM cases is 54. Therefore, a threshold was set using the largest European survey that reported most surgeons performing fewer than 5 procedures per year as a reliable cutoff, with a minimum of one procedure per quarter to distinguish low and high-volume centers [5].

Prenatal practices revealed that fetal magnetic resonance imaging (MRI) was performed in most centers, regardless of case volume, indicating its widespread use in diagnosing CLM prenatally. However, differences were observed in the diagnostic imaging setup for preoperative management. High-volume centers tended to perform a single computed tomography (CT) scan, while low-volume centers conducted two scans. In addition, post-natal MRI resulted still under employed, with less than half of centers considering this imaging technique as additional preoperative study for CLM. Nonetheless, a single CT scan acquisition remained the most reported choice by most internal centers, although little is known about the preoperative imaging assessment of CLM [6]. CT scan remains the imaging modality of choice for CLM, but the increasing studies highlighting the importance of pediatric radioprotection suggest limiting the number of ionizing radiation imaging [7–9].

Surgical indications showed broad agreement among both low-volume and high-volume centers, confirming internationally adopted indications [10–13]. The operative approach favored asymptomatic congenital cystic lung malformations, hybrid forms, extra lobar, and intralobar sequestration in both groups, with a minority of centers underwent a watchful waiting approach. However, a wait-and-see approach was more commonly adopted for congenital lobar emphysema, with surgery considered only in the presence of symptoms. This highlights the importance of clinical judgment and individualized decision-making based on specific CLM characteristics.

Surgical timing for asymptomatic CLM showed significant differences between high-volume and low-volume centers. High-volume centers tended to operate on patients younger than 12 months, potentially reflecting a preference for early intervention to address the condition. These results confirm the current trend of reducing the age at surgery for CLM due to the higher risk of infection over time and the related increase in surgical complications [14–17]. The composition of the surgical team and the surgical environment did not significantly vary between the two groups, with pediatric surgeons being the primary operators in a general hospital setting. Integrated practice units between pediatric and adult surgeons are increasingly adopted in the USA, sharing common pathways to overcome the management limits of small numbers of rare conditions [18]. The findings confirmed an increased integrated pattern of care for CLM, with adult thoracic surgeons participating in a quarter of the centers and the prevalence of pediatric surgical units in general hospitals. However, the value of quality improvement in the integrated pattern of care has not yet been established and requires further investigation.

In perioperative management, there was a nationwide agreement among centers, regardless of case volume, regarding the preferred approach for asymptomatic CLM, with thoracoscopy being the favored method in most centers. Recent structured comparative studies have demonstrated the superiority of the thoracoscopic approach over a thoracotomy in terms of pulmonary function and musculoskeletal sequelae, confirming the importance of adopting this technique [19–21]. However, for symptomatic patients, there is no standardized approach, suggesting a need for further consensus and guidelines in this regard. Most centers prefer lobectomy as the primary surgical procedure for CLM resection, indicating its established position. The nationwide agreement on lobectomy reflects the widespread adoption of this technique over pulmonary-sparing techniques like segmentectomy and wedge resection, as it carries a lower risk of residual disease and allows for the detection of distant lobar locations not identified during preoperative imaging [22, 23].

The utilization of single lung ventilation during surgical procedures is widespread in most centers, aligning with the current international practice in anesthesiology. It is followed by selective lung intubation and bipulmonary ventilation [24]. Although there is variability in ventilation techniques, the utilization of bronchial blockers appears to be the preferred method, with a slightly higher frequency in high-volume centers. This trend reflects the increasing popularity of minimally invasive techniques such as thoracoscopy, which often necessitates deflating one lung to ensure optimal visualization and access.

Regarding postoperative recovery settings, there is a preference for patients to stay in the surgical ward rather than the intensive care unit. This approach is supported by the increasing adoption of enhanced recovery protocols in children, including those who have undergone thoracic procedures [25, 26]. Postoperative follow-up practices showed that most centers conducted systematic imaging (CT or MRI) to ensure proper monitoring of surgical outcomes. In addition, pulmonary function tests were performed in most centers, indicating a comprehensive evaluation of respiratory function. However, there was inconsistency in the transition of care and long-term follow-up, highlighting a potential gap in the continuity of care for CLM patients. Pediatric surgery is one of the youngest surgical specialties, and the understanding of CLM pathophysiology and surgical treatment was introduced less than 50 years ago. Thus, similar to other pediatric congenital diseases, there is an increasing need for long-term follow-up to better understand the incidence of functional sequelae resulting from CLM management [27, 28].

The findings of this study provide valuable insights into the current management practices for CLM in Italian centers, with a consensus adoption of minimally invasive surgery for asymptomatic CLM. The observed variations in the overall pattern of care, including diagnostic imaging, surgical indications, surgical timing, and perioperative management, indicate the need for standardized guidelines and consensus to ensure uniformity in care delivery. The establishment of a national common pattern of care and networking has been shown to reduce the gap in quality of care [29, 30]. This study highlights the need to reduce preoperative CT imaging assessment and the age at the surgery to overcome the variability in the pattern of care among low and high-volume centers.

Our study has certain limitations. It focused on the surgical management aspects of CLM and did not compare surgical outcomes between low and high-volume centers. We assumed that surgical volume is associated with better quality of care, as described by Park and colleagues [31]. Therefore, further research is needed to examine the effects of a unified national management approach, considering the variations in surgical case volume. In addition, our findings

cannot be considered conclusive evidence of a nationwide agreement. They rather serve as a foundation for a future national Delphi study that will be necessary to establish evidence-based national guidelines.

Conclusion

The nationwide survey revealed a widespread acceptance of minimally invasive surgery for asymptomatic CLM. The analysis of management variability identified preoperative imaging protocols and surgical timing as key areas for improvement to establish a consistent national standard of care.

Author contributions L.P: conceptualization, writing original draft. G.V, AAS, RLP, AC, AR: methodology. FM, SC, RG, FT, EZ, GC, CV, EG, LN, SC, VDB, GL, MC, AM, AV, EC, SC: data curation. DG: statistical analysis. GR, CE, PD, PB, CB, PG: validation. PM, PB, GP, MT, FC, CE: supervision. All the authors reviewed the manuscript.

Funding Nothing to disclose.

Declarations

Conflict of interest The authors have no conflicts to declare.

Previous communications This work has been previously presented at the 2023 IPEG-ESPES congress.

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Authors and Affiliations

Luca Pio[1,2] · Valerio Gentilino[3] · Francesco Macchini[4] · Alberto Attilio Scarpa[5] · Roberto Lo Piccolo[6] · Andrea Conforti[7] · Alberto Ratta[8] · Riccardo Guanà[9] · Francesco Molinaro[10] · Sara Costanzo[11] · Giovanna Riccipetitoni[12,13] · Gabriele Lisi[14] · Paola Midrio[15,16] · Francesca Tocchioni[17] · Giovannii Cobellis[18] · Andrea Volpe[19] · Elisa Zolpi[20] · Anna Morandi[21] · Enrico Ciardini[22] · Claudio Vella[23] · Maria Giovanna Grella[24] · Maria Sergio[25] · Edoardo Guida[26] · Lorenzo Nanni[27] · Silvia Ceccanti[28] · Vincenzo Di Benedetto[29] · Maurizio Cheli[30] · Alfredo Garzi[31] · Maria Nobili[32] · Valeria Gabriele[33] · Giovanni Boroni[34] · Filippo Incerti[35] · Nicola Zampieri[36] · Sebastiano Cacciaguerra[37] · Pier Luca Ceccarelli[38] · Maria Escolino[39] · Vito Briganti[40] · Davide Gori[41] · Ciro Esposito[39] · Piergiorgio Gamba[19,42] · Fabrizio Gennari[9] · Alessandro Inserra[43,44] · Patrizia Dall'Igna[45,46] · Carmelo Romeo[47] · Pietro Bagolan[7,44] · Cosimo Bleve[20] · Fabio Chiarenza[20] · Francesco Morini[17] · Gloria Pelizzo[11] · Michele Torre[1]

✉ Luca Pio
luca.pio@stjude.org

[1] Pediatric Thoracic and Airway Surgery, IRCCS Giannina Gaslini, Genoa, Italy

[2] Department of Surgery, MS133, St. Jude Children's Research Hospital, 262 Danny Thomas Place, Memphis, TN 38105-3678, USA

[3] Unit of Pediatric Surgery, Woman and Child Department, Filippo del Ponte Hospital—ASST Sette Laghi, Varese, Italy

[4] ASST Grande Ospedale Metropolitano Niguarda, Milan, Italy

[5] Pediatric Surgery Unit, Pietro Barilla Children's Hospital, Parma, Italy

[6] Department of Emergency, Critical Area and Pediatric Surgery, Meyer University Children's Hospital, University of Florence, Florence, Italy

[7] Neonatal Surgery Unit, Department of Medical and Surgical Neonatology, Bambino Gesù Children Hospital—Research Institute, Rome, Italy

[8] Pediatric Surgery Unit, Infermi Hospital, Rimini, Italy

[9] Department of Pediatric General Surgery, Regina Margherita Children's Hospital, Azienda Ospedaliero-Universitaria Città Della Salute E Della Scienza, Turin, Italy

[10] Division of Pediatric Surgery, Department of Medical, Surgical and Neurological Sciences, University of Siena, Siena, Italy

[11] Pediatric Surgery Department, Vittore Buzzi" Children's Hospital, Milan, Italy

[12] Department of Clinical, Surgical, Diagnostic and Pediatric Sciences, University of Pavia, Pavia, Italy

[13] Pediatric Surgery Unit, Department of Maternal and Child Health, Fondazione IRCCS Policlinico San Matteo, Pavia, Italy

[14] Department of Pediatric Surgery "Spirito Santo", Hospital of Pescara "G. D'annunzio", University Chieti-Pescara, Pescara, Italy

[15] Pediatric Surgery, Cà Foncello Hospital, Treviso, Italy

[16] Pediatric Surgery, University of Padua, Padua, Italy

[17] Department of Pediatric and Neonatal Surgery, Meyer Children's Hospital, Florence, Italy

[18] Unit of Pediatric Surgery, Salesi Children's Hospital, Marche Polytechnic University, Ancona, Italy

[19] Pediatric Surgery Unit, Women's and Children's Health Department, University Hospital of Padua, Padua, Italy

[20] Department of Pediatric Surgery and Pediatric Minimally Invasive Surgery and New Technologies, San Bortolo Hospital, Vicenza, Italy

[21] Department of Pediatric Surgery, Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico, Milan, Italy

[22] Chirurgia Pediatrica Ospedale S.Chiara, Trento, Italy

[23] Azienda Ospedaliero-Universitaria di Ferrara, Ferrara, Italy

[24] Department of Pediatric Surgery, Santobono Pausilipon Children's Hospital, Naples, Italy

[25] Policlinico Universitario di Palermo, Palermo, Italy

[26] Department of Pediatric Surgery, Institute for Maternal and Child Health, IRCCS Burlo Garofolo, Trieste, Italy

[27] Fondazione Policlinico Universitario "A. Gemelli" IRCCS, Rome, Italy

[28] Pediatric Surgery Unit, Department of Maternal Infantile and Urological Sciences, Sapienza University of Rome, Azienda Ospedaliero-Universitaria Policlinico Umberto I, Rome, Italy

[29] Pediatric Surgery Unit, University Hospital of Catania, Catania, Italy

[30] Department of Paediatric Surgery, Papa Giovanni XXIII Hospital, Bergamo, Italy

[31] Dipartimento di Chirurgia Pediatrica e Robotica, Università di Salerno, Salerno, Italy

[32] Pediatric Surgery Unit, University of Foggia, Foggia, Italy

[33] AOU Maggiore Della Carità, Novara, Italy

[34] Department of Pediatric Surgery, Azienda Ospedaliera Spedali Civili, Brescia, Italy

[35] Department of Pediatric Surgery, Umberto Bosio Center for Digestive Diseases, The Children Hospital AO SS Antonio E Biagio E Cesare Arrigo, Alessandria, Italy

[36] Woman and Child Hospital, Department of Surgery, Dentistry, Paediatrics and Gynaecology, University of Verona, Verona, Italy

[37] Deparment of Pediatric Surgery, Ospedale Garibaldi, Catania, Italy

[38] Pediatric Surgery Department, Policlinico di Modena, Modena, Italy

[39] Department of Translational Medical Sciences, Federico II University, Naples, Italy

[40] Department of Pediatric Surgery and Urology Unit, San Camillo Forlanini Hospital, Rome, Italy

[41] Department of Biomedical and Neuromotor Sciences, University of Bologna, 40126 Bologna, Italy

[42] Pediatric Surgery, Department of Women's and Children's Health, Padua University, Padua, Italy

[43] General Surgery Unit, Department of Surgery, Bambino Gesù Children's Hospital - IRCCS, Rome, Italy

[44] Department of Systems Medicine, University of Tor Vergata, Rome, Italy

[45] Department of Emergencies and Organ, Transplantation Azienda Ospedaliero-Universitaria Consorziale Ospedale Pediatrico Giovanni XXIII, Bari, Italy

[46] University of Bari, Bari, Italy

[47] Unit of Pediatric Surgery, Department of Human Pathology of Adult and Childhood "Gaetano Barresi", University of Messina, Messina, Italy