EARLY SURGICAL APPROACH OF ASYMPTOMATIC CHILDREN WITH CONGENITAL PULMONARY AIRWAY MALFORMATION: AN UPDATE REVIEW

ABORDAGEM CIRÚRGICA PRECOCE DE CRIANÇAS ASSINTOMÁTICAS COM MALFORMAÇÃO CONGÉNITA DAS VIAS AÉREAS PULMONARES: UMA REVISÃO ATUALIZADA

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ABSTRACT

The objective of this integrative literature review is to discuss the perspectives regarding the early surgical approach of asymptomatic children with congenital pulmonary airway malformation. The review was conducted by performing electronic searches from May to July 2023, in the PUBMED database – National Library of Medicine. Studies published from January 2013 to January 2023, clinical trials, retrospective studies, case studies, series and cases and mixed studies available in English were included. Elementary information was collected from the articles selected after the filters, such as study design (main methodological aspects), key findings and conclusion (based on the problem question that guided this study). Initially, 125 articles were identified from the search in the selected database. After the analysis regarding the application of the inclusion and exclusion criteria, 9 manuscripts remained, all of them of the retrospective type. In conclusion, although there is currently a divergence of opinions regarding the performance of an early surgical procedure in children diagnosed with asymptomatic congenital pulmonary airways malformation, current studies point to the procedure as they consider several factors, including patient safety.

KEYWORDS: Cystic adenomatoid malformation of lung; surgery; asymptomatic diseases; pediatrics.

RESUMO

O objetivo desta revisão integrativa da literatura é discutir as perspectivas em relação à abordagem cirúrgica precoce de crianças assintomáticas com malformação congénita das vias aéreas pulmonares. A revisão foi realizada por meio de buscas eletrônicas no período de maio a julho de 2023, na base de dados PUBMED - National Library of Medicine. Foram incluídos estudos publicados de janeiro de 2013 a janeiro de 2023, ensaios clínicos, estudos retrospectivos, estudos de caso, séries e casos e estudos mistos disponíveis em inglês. Dos artigos selecionados após os filtros foram recolhidas informações elementares, tais como: desenho do estudo (principais aspectos metodológicos), principais resultados e conclusão (com base na questão-problema que orientou este estudo). Inicialmente, foram identificados 125 artigos a partir da busca na base de dados selecionada. Após a análise quanto à aplicação dos critérios de inclusão e exclusão, restaram 9 manuscritos, todos do tipo retrospectivo. Concluindo, apesar de atualmente haver divergência de opiniões quanto à realização de procedimento cirúrgico precoce em crianças com diagnóstico de malformação congênita assintomática das vias aéreas pulmonares, estudos atuais apontam para o procedimento por considerarem diversos fatores, dentre eles a segurança do paciente.

PALAVRAS-CHAVE: Malformação Adenomatoide cística congênita do pulmão; cirurgia; doenças assintomáticas; pediatria.

1. INTRODUCTION

The Congenital Pulmonary Airway Malformation (CPAM), previously known as Congenital Cystic Adenomatoid Malformation (CCAM)[1], results from abnormalities in the morphogenesis of the lower airways. Although rare and with etiology still poorly understood, it is the most common congenital pulmonary lesion, whose diagnosis has increased with

the improvement of prenatal ultrasound techniques[2,3].

Its first definition dates to 1949 when Ch'in and Tang described CPAM as characterized by the replacement of lung tissue by cystic tissue (dysfunctional)[4]. Currently, it is known that CPAM accounts for about 95% of a group of pathologies called Congenital Cystic Lung Lesions (LLCCs)[5].

It is important to note that previous epidemiological analyses estimated the incidence of CPAM at 1/25,000 to 1/35,000 pregnancies[6]. However, with advances in prenatal ultrasound diagnostic procedures, more recent studies indicate that its incidence has reached a level of 1/3,000 pregnancies. This is because diagnoses of LLCCs, made by prenatal ultrasound, are accurate, with a sensitivity in the order of 90%[7,8]. It is worth noting that both ultrasound and X-ray are not able to define the size and nature of congenital lung malformations after birth, with chest CT being indicated for this purpose. Lesion classification is only possible through histological analysis (types 0-IV by Stocker)[9].

In diagnostic terms, the clinical manifestations of CPAM are diverse, as evidenced by the possibility of identifying it prenatally through ultrasound procedures, or even remaining asymptomatic until adulthood. Recent studies show that approximately 25% of newborns with CPAM identified prenatally are symptomatic at birth, and the severity of the condition is directly associated with the increasing size of the lesions and the presence of other morphological complications (ascites, mediastinal deviation, etc.)[2,10].

Considering that the vast majority of CPAM cases remain asymptomatic, current studies still present contradictory findings regarding the surgical approach for this patient population[11]. Thus, the aim of this study was to discuss perspectives on early surgical intervention for asymptomatic CPAM patients.

2. MATHERIAL AND METHODS

This is an integrative review study built based on the PRISMA extension applied to integrative and scoping reviews[12], using the guiding instrument by Souza *et al.* (2010)[13], following the construction steps proposed by Whittemore and Knafl (2005)[14]: a) problem identification, b) literature search, c) data evaluation, d) data analysis, and e) presentation.

Problem Identification

For a better formulation of the research question, the PICO acronym was used as guidance. P (patients) - asymptomatic pediatric patients diagnosed with CPAM; I (intervention) - early surgical intervention; C (comparator) - groups without intervention, clinical/conservative management; O (outcomes) - evaluation of surgical intervention in asymptomatic pediatric patients and its impact on prognosis improvement and/or quality of life.

Thus, the guiding research question for the review was formulated as follows: Does early surgical intervention in asymptomatic children with CPAM lead to better prognosis?

Literature Search

Electronic searches were conducted from May to July 2023, in the PUBMED - National Library of Medicine database. The following terms, duly registered in the Medical Subject Headings (MESH), were used for the search: a) Cystic Adenomatoid Malformation of Lung, Congenital, b) Surgery, c) Asymptomatic. Boolean operators AND and OR were used in the advanced search stage to formulate the search key, resulting in the following key: ((Cystic Adenomatoid Malformation of Lung, Congenital [Text Word]) AND (Surgery [Text Word])) AND (asymptomatic [Text Word]) Filters: from 2013 - 2023.

In this study, manuscripts meeting the following criteria were included: a) Studies published from January 2013 to January 2023, b) Clinical trials, retrospective studies, case studies, case series, mixed-method studies, c) available in the English language. Manuscripts were excluded if they: a) were editorials, comments, or review studies, b) had samples with other associated underlying diseases, c) involved surgical intervention in patients outside the pediatric age group.

Data Evaluation and Analysis

Given the study's characteristics, these two steps were worked on dynamically and inseparably. For this, the provider "Rayyan Systems Inc." was used as a resource. The screening and analysis process of the manuscripts to be included in the sample was carried out simultaneously by two of the researchers involved and followed the steps outlined in the PRISMA extension applied to integrative and scoping reviews[12] (Figure 1). From the articles selected after the filters, elementary information was collected, such as study design (main methodological aspects), determinant findings, and conclusion (based on the problem question).

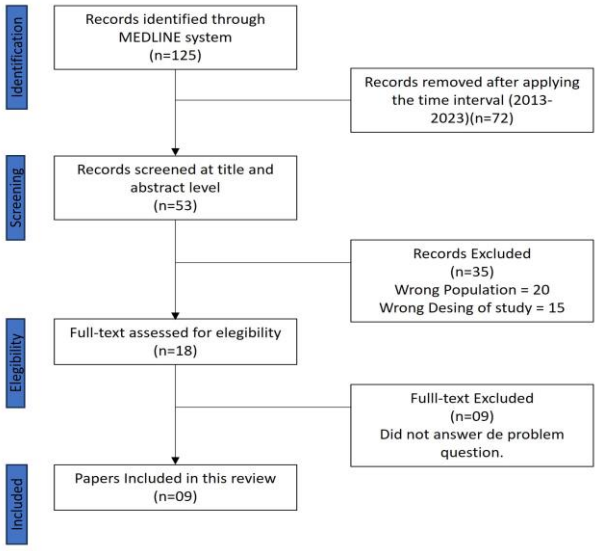

Figure 1. Diagram illustrating the flow of manuscript selection included in this study. **Source:** Authors**.**

3. DEVELOPMENT

In this search, 125 articles were identified from the search in the selected database. After a thorough analysis by two researchers regarding the application of inclusion and exclusion criteria, nine manuscripts remained, which constituted the final framework of this review (Table 1).

It is worth mentioning that all manuscripts included in the final analysis of this study were of retrospective type. If the samples analysed in each of the listed studies are summed, 990 children were diagnosed with pulmonary malformations, with most of them diagnosed with CPAM (among symptomatic and asymptomatic cases).

Table 1. Overview of the manuscripts included in the present study.

Authors/ Year of Publication	Country of Study	Objective/ Design of Study	Main Findings
Verhalleman *et al.*, 2022 [15]	Belgium	Assess the diagnosis, clinical signs, and strategy for congenital cystic adenomatoid malformations (CCAM). Retrospective study involving 55 patients diagnosed with CPAM at the University Hospital of Leuven between 1993 and 2016.	In patients who were asymptomatic (40%), elective surgery was performed, characterized by a mini-thoracotomy and only a lobectomy was performed. The researchers suggest that the elective procedure can be performed safely in asymptomatic patients, and it can be scheduled during youth in those cases where there is a predisposition to future complications.
Style *et al.*, 2019 [16]	USA	To examine the postsurgical outcomes of a consecutive series of children treated with elective operations for congenital lung malformations. Retrospective study carried out between July 2001 and June 2016 at the Texas Children's Fetal Center.	Of 220 children investigated, 110 had asymptomatic lesions and had elective resection. Of these 110, the median maximum fetal CVR was 0.8 [range 0.1–2.2], and the median age at operation was 4 (1.5–60) months (58% had resection at ≤4 months). Overall complication rate, including pneumothorax and pleural effusion, was 15%. When comparing those with resection under 4 months to those over 4 months, there were no significant differences in complication rates or length of stay. Operative time was shorter for patients with early resection (154 ± 59 vs 181 ± 89, p = 0.05). Conclusion: early elective resection of congenital lung malformations before 4 months of age is feasible and not associated with greater operative risk.
Esposito *et a.l*, 2021 [17]	...	To report a European multi-institutional experience on the thoracoscopic management of children with congenital lung malformations. We retrospectively collected 102 patients (49 girls and 53 boys) with a median age at surgery of 1 year (range 6 months to 1.5 years), who underwent thoracoscopic lobectomy in five European pediatric surgery units. Among the clinical conditions, CPAM was the most prevalent (n = 47). Overall, the condition was asymptomatic in 77/102 (75.5%), while symptoms such as recurrent pneumonia and/or respiratory distress were present in 25/102 (24.5%).	The researchers suggest that thoracoscopic lobectomy is a safe and effective procedure with excellent cosmetic results if performed by experienced professionals. In conclusion, they strongly recommend surgery in patients with congenital lung malformations up to the first year of life, to reduce the risk of infection and technically facilitate the procedure, despite the small size of the patients. The surgeon's experience and use of miniaturized instruments and occlusion devices remain key factors in a successful outcome.
Ng at al., 2014 [9]	UK	To review the outcome of all congenital lung malformations (CLMs) diagnosed prenatally and treated conservatively in the studied center. Retrospective study that grouped all children treated between 2001 and 2011, totaling 74 children with CLM's.	Of the 74 children analyzed, emergency lobectomy was performed in one symptomatic newborn. Elective lobectomies were performed at the parents' request in three asymptomatic children. Two patients developed pneumonia in the affected lobe during early childhood and underwent lobectomy at 3 years of age. One patient with bronchopulmonary sequestration required embolization due to cyanotic episodes. The remaining 65 patients were treated conservatively and, at the time of publication of the manuscript, none required hospital admission. Less than a quarter report mild respiratory symptom, such as coughing or wheezing. The average follow-up is 5

			years. The study also suggests that the majority of injuries detected in children are asymptomatic and conservative treatment seems to be prudent in these cases, although they recognize the need to evaluate each case specifically and only then consider surgery.
Furukawa *et al.*, 2015 [18]	Japan	To evaluate surgical intervention strategies for congenital cystic lesions of the lungs (CCL), with a focus on the safety of lung resection. The clinical characteristics of 27 children (CCAM, n=16; bronchial atresia, n=4; bronchogenic cyst, n=3; pulmonary sequestration, n=3; lobar emphysema, n=1) treated at the unit studied between the years 1995 and 2014.	Of the 27 patients, 14 were asymptomatic and 13 were symptomatic. The mean age at surgery was 4 months in the asymptomatic group and 4.1 years in the symptomatic group. The average operating time was 167 minutes in the asymptomatic group and 275 minutes in the symptomatic group (P<0.001). The average amount of intraoperative bleeding was 15g in the asymptomatic group and 83.4g in the symptomatic group (P<0.05). Conclusion: The researchers suggest that the surgical procedure in asymptomatic patients should be encouraged within a period of up to six months after birth.
Thakkar *et al.*, 2017 [19]	UK	To report the Oxford experience through a retrospective study over a 10-year period, during which 91 patients had prenatal suspicion of cystic lung lesion.	Of the 88 live births investigated in the study, a total of 64 (73%) patients underwent surgery, with the most common injuries being congenital pulmonary airway malformations (CPAMs) (24), hybrid injuries (19) and pulmonary sequestrations (12). The median age at surgery was 5 months (1 day to 17 months). Using a minimal access approach, 41 (64%) cases were completed with 17 performed open from the start. Conclusions: Excision of asymptomatic lesions is safe with minimal complications. There is a risk of infection and a definite, although low, risk of malignancy, which may outweigh the benefits of conservative treatment.
Khan *et a.l*, 2021 [20]	UK	Retrospective study that seeks to report the ten-year experience (2008-2017) of an institution in the development of video-assisted thoracoscopic resection (VATS) of CPAM in children.	72 children, with a mean age of 10 months, underwent VATS; of these, 7 (10%) required conversion to open thoracotomy. Twenty (27.7%) were 'P-symptomatic' and the duration of surgery when compared to 'asymptomatic' children was longer 269 (range 129-689) versus 178 (range 69-575) minutes (P = 0.01). Postoperatively, 8 children (11%) had grade III/IV (Clavien-Dindo) complications; persistent air leak/pneumothorax (n = 5), chylothorax (n = 1), pleural effusion (n = 1) and middle cerebral artery seizure/thrombosis (n = 1). There was no mortality. Twenty-four children (33.3%) were reported as 'symptomatic-P' after surgery after a mean follow-up of 2.18 years. Surgical intervention had no impact on 'symptomatic-P' status (P = 0.46). Conclusion: The researchers suggest that these results indicate that the risks arising may outweigh the benefits of surgery in asymptomatic children.
Jiahang Zeng *et al.*, 2021 [21]	China	Evaluate the ideal surgery time in children with CPAM based on the risk of infection. Retrospective study that included 237 children diagnosed between January 2012 and January 2020 at Guangzhou Women an Children's Medical Center.	Infection rates in patients under 2 years of age were significantly lower than patients over 2 years of age (11.4% vs. 45.7%, p < 0.001). Considering the high risk of ifnection, less chance of minimally invasive surgery, higher rate of pulmonary lobectomy and greater blood loss in patients older than 2 years, this study suggests early surgical treatment (<2 years) in asymptomatic patients with CPAM I and CPAM II.
Bo Xia *et al.*, 2017 [22]	China	To analyze patients with CCAM undergoing surgical resection	Among the 115 patients, most were asymptomatic (96). The mean size of the lesions was much

	and evaluate the clinical role of lesion volume, X-ray and CT in these patients, as well as the need for surgery and clinical factors. Retrospective study that included 115 children with CCAM, diagnosed between September 2012 and June 2014, from The Maternal and Children Hospital of Guangdong Province.	larger in symptomatic children (5.28cm; range 3.4-7.5) than asymptomatic ones (mean 3.34; range 0.5-7.4), however the size of the lesions did not was related to the prognosis. There were 11 patients with a prenatal diagnosis of resolution of the lesions by USG, whose postnatal investigation with CT showed persistence of the lesions. The X-ray showed a high rate of false negatives (60%), but with good resolution in urgent cases (congenital lobar emphysema, mediastinal shift or small pneumothorax). Therefore, early chest X-ray and CT are indicated. The study also recommends early surgical intervention for CCAM.

Source: Authors.

4. DISCUSSION

The surgical treatment of symptomatic patients with CPAM is well-described in the literature and is not controversial. Commonly described symptoms include dyspnea, hemorrhage, cyst infection, pneumothorax, and nutritional deficits, for which surgical intervention is necessary to prevent complications[23]. However, when it comes to early surgical treatment for asymptomatic patients, studies are controversial and inconclusive[11], highlighting the importance of this manuscript.

The natural history of the disease is still not fully understood, with lesions ranging from regression in the first year of life to progression to malignancy in adulthood; thus, there is no reliable evidence on the optimal management of affected children[24]. In this context, studies have been developed that support both extremes of this issue: operating or not operating asymptomatic patients. In a recently developed meta-analysis, of 168 patients followed for a long period, only 36% of patients treated non-surgically remained asymptomatic at the last follow-up[25]. From another perspective, in a meta-analysis conducted years earlier, 1,070 patients were analysed, and symptoms developed in only 3.2% of asymptomatic cases[26].

An undefined proportion of cases may develop malignancy[24]. Studies advocating conservative treatment argue that the incidence of pleuropulmonary blastoma is low (<1/100,000), as well as cases of bronchioloalveolar carcinoma in adults, even after complete lobectomy in childhood, implying that surgery may not be protective. Therefore, surgical treatment would be reserved for cases of patients with risk factors for pleuropulmonary blastoma, which include patients with bilateral or multifocal lung lesions or those with a positive family history of cancer[9].

Seventy percent of centers worldwide adhere to a policy of prophylactic resection, regardless of symptoms or lesion size[9]. Today, there are several arguments advocating for early surgical resection, for reasons that include: prevention of recurrent infections and, to a lesser extent, malignant transformation; lower risk of emergency surgery; longer time for compensatory alveolar growth. Many studies indicate that emergency surgery is associated with higher morbidity, with increased postoperative ventilation requirements and hospital stays longer than 7 days[24,27,28].

Although there is no consensus on the ideal age for surgery, some advocate for operating in the neonatal period and others after 4 weeks of age to reduce the risk of anesthesia before this age[24,29]. A recent retrospective study aimed to determine the optimal time for surgical resection in the first year of life and found no significant differences in complication rates, hospital readmissions, or conversion from VATS to open surgery, suggesting that surgery is equally safe when performed from the first month of life to the first year of life[24]. Other authors have concluded that the morbidity associated with surgery is significantly higher in infants under 3 months and the duration of the intervention increases significantly in children over 9 months, which may be related to inflammation associated with recurrent infections, which often can be asymptomatic, thus suggesting that the optimal time is between 3-9 months of age[28].

In the present study, of the 09 manuscripts selected based on the inclusion and exclusion criteria, two of them (both conducted in the United Kingdom) indicate conservative management (clinical monitoring) of asymptomatic cases as a safe and appropriate strategy[9,19]. The other 07 studies indicate that the potential benefits outweigh the inherent risks of surgical procedures, justifying the importance of early surgical approach in asymptomatic cases[15–18,20].

5. CONCLUSION

Although there is currently a divergence of opinions regarding early surgical intervention in asymptomatic children diagnosed with CPAM, most current studies point towards performing the procedure due to considerations of patient safety, decreased potential risk when symptoms develop, and reduced likelihood of future complications. Additional longitudinal/ follow-up studies appear to be interesting possibilities for future conditions.

6. FINANCING

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