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Review Article

Surgical versus conservative management of congenital pulmonary airway malformation in children: A systematic review and meta-analysis

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ABSTRACT

Background: The ideal management of infants born with asymptomatic congenital pulmonary airway malformation (CPAM) is controversial. We performed a systematic review and meta-analysis comparing elective resection versus expectant management.
Methods: We searched CENTRAL, MEDLINE, EMBASE, CINAHL, and PubMed for studies describing the management of asymptomatic CPAM and reporting on postoperative morbidity, mortality, and length of hospital stay (LOS). We performed meta-analyses when possible and provide a narrative summary of results.
Results: One nonrandomized prospective and eight retrospective studies met our inclusion criteria. Out of 168 patients, 70 underwent surgery before symptoms developed with seven experiencing postoperative complications (10.0%); 63 developed symptoms while being managed expectantly and subsequently underwent surgery with 20 complications (31.8%). Thirty-five patients continued to be followed nonsurgically (three months to nine years of follow-up). Morbidity was higher with surgery after symptom development (6 studies; odds ratio 4.59, 95% confidence interval (CI) 1.40 to 15.11, $P < 0.01$); there was no difference in LOS (3 studies; mean difference 4.96, 95% CI −1.75 to 11.67, $P = 0.15$). There were no related deaths.
Conclusions: Elective resection of asymptomatic CPAM lesions is safe and prevents the risk of symptom development, which may result in a more complicated surgery and recovery.

Congenital pulmonary airway malformation (CPAM) is a relatively rare congenital disorder characterized by replacement of a portion of lung by a nonfunctioning cystic section of abnormal tissue [1,2]. CPAM represent 25% to 30% of all congenital lung malformations, with a slight male predominance [3]. While the prevalence is estimated to be between 1 in 10,000 and 1 in 35,000 pregnancies [1,3,4], the true prevalence is difficult to discern given the portion that go undiagnosed owing to a lack of clinical symptoms.

There is much variation in the presentation and evolution of CPAM, ranging from prenatal death to respiratory distress in early life to entirely asymptomatic lesions [2,5]. An increasing proportion of neonates are now presenting asymptomatically, as many are identified on antenatal ultrasound. Clinical presentation typically dictates treatment, however when cases of known CPAM are asymptomatic, optimal management is less defined [6,7].

Arguments favoring either elective surgery or expectant management of asymptomatic lesions are many. They include estimates that delaying surgery until symptom onset will result in a more complicated procedure with worse surgical outcomes [8–13], as well as counterarguments stating that, as rates of symptom development are largely unknown [14,15], intervening before symptoms appear may result in exposure of healthy patients to unnecessary surgical risks [15]. These differing viewpoints illustrate the lack of consensus as to whether surgical management or an expectant approach for asymptomatic CPAM is associated with a smaller risk of morbidity in the short and long term.

Our objective was to conduct a systematic review and meta-analysis of all published literature comparing the postoperative outcomes associated with elective surgery (while asymptomatic) versus those following surgery as part of expectant management (i.e., performing surgery only upon the development of symptoms) in children born with asymptomatic CPAM lesions. This research was carried out for the Canadian Association of Pediatric Surgeons (CAPS) Evidence-Based Resource [16], the aim of which is to encourage evidence-based practice in pediatric surgery by making up-to-date evidence easily accessible.

1. Methods

1.1. Selection of study topic

We used a Dephi-like method [17] to determine the issues of greatest concern to pediatric surgeons regarding the surgical management of CPAM.

1.1.1. First round

We sent an online survey to 18 experts with proficiency in this area, identified primarily on the basis of a literature search. This survey was

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hosted on the CAPS website and consisted of the following open-ended question: "In your practice with the surgical management of congenital pulmonary airway malformation, what issues do you find to be controversial and in need of further research and/or consensus?"

1.1.2. Second round

We developed a questionnaire listing all issues raised by the experts in the first round. We sent this questionnaire to the respondents from the first round, asking them to select the three issues which they felt were the most important. The issue that received the highest number of votes is the focus of this review.

1.2. Literature search

To identify all studies on elective surgery and expectant management of asymptomatic CPAM lesions, we conducted an electronic literature search of the Cochrane Central Register of Controlled Trials (CENTRAL), MEDLINE (1966 onwards), EMBASE (1980 onwards), CINAHL (1982 onwards), and PubMed (1966 onwards) in January 2015. We used the search terms ("asymptomatic" AND "congenital" AND "cystic" AND "adenomatoid malformation") OR ("lung" AND "lesion") AND ("management" OR "treatment" OR "therapy."). We excluded any studies on nonhuman subjects and those that did not focus on pediatric patients, as well as any studies that were not published in English, owing to resource limitations. We also excluded editorials, case reviews, any studies that did not evaluate postoperative outcomes, and any studies where it was not possible to extract data only on patients who were asymptomatic at birth. Finally, we hand-searched the reference sections of all relevant articles to identify additional studies. Two independent reviewers conducted all article screening; where consensus on an article could not be reached, a third reviewer was consulted.

1.3. Outcomes

Our primary outcome of interest was postoperative morbidity including respiratory distress, respiratory infection, pneumonia, pneumothorax, and death. Our secondary outcome of interest was length of stay in hospital following surgery (LOS).

1.4. Quality assessment

We used the Methodological Index for Non-Randomized Studies (MINORS) to assess the quality of the included studies, as all were nonrandomized [18]. The MINORS criteria comprise 12 items, each of which is assigned a score of 0, 1 or 2, for a maximum total score of 24 (comparison studies) or 16 (noncomparison studies). Higher scores are indicative of greater methodological quality. Two researchers independently assessed each study using the MINORS criteria and discussed their score for each item to reach a consensus. When consensus could not be reached, a third researcher was consulted. Had we identified any, we would have used the Cochrane "Risk of bias" tool to assess randomized controlled trials [19].

1.5. Data extraction

One reviewer extracted all data from the included studies, and another checked it for accuracy and completeness. We extracted the following data exclusively on patients who were asymptomatic at birth: number of patients undergoing elective surgery, number undergoing surgery as part of expectant management (after symptom development), age at symptom development if applicable, postoperative complications, mortalities, and total length of postoperative hospital stay, as well as number of patients successfully managed conservatively at the end of follow-up, and length of follow-up for these patients.

1.6. Statistical analysis

We conducted our meta-analyses using Review Manager 5.3 [20], using random-effects models [21], to produce an odds ratio (OR) for morbidity, as this is a categorical variable, and a mean difference (MD) for length of stay, as this is a continuous variable, along with 95% confidence intervals (CIs). We produced Chi^2 and I^2 values to assess homogeneity. Had we included a sufficient number of studies, we would have created funnel plots to help assess the risk of reporting and other biases [19,22].

2. Results

2.1. Studies included

Our electronic search yielded 342 studies. After applying our exclusion criteria to titles and abstracts, 63 studies remained. Full-text review resulted in eight of these studies being included, while an additional one was identified from hand-searching the reference sections of included articles; nine studies were ultimately included [10–12,14,23–27]. Seven of these were amenable to inclusion in meta-analyses [10–12,14,23–25] (Fig. 1).

2.2. Study characteristics

We did not identify any RCTs; all but one of the included nine studies were retrospective [26]. Studies were published between 1996 and 2011 and were conducted in Australia [14,27], Belgium [25], Canada [23], China [24], Israel [26], Italy [10], Japan [12], and the United States of America [11]. The MINORS scores for our included studies ranged from 7 to 14, with a median score of 9 (Table 1).

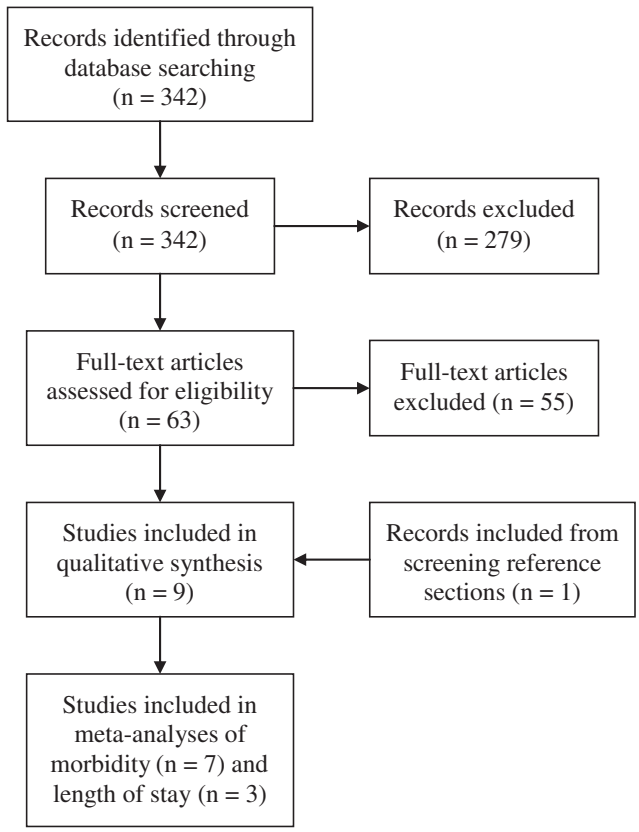

Fig. 1. Flow diagram of search and screening process.

Table 1
Characteristics of included studies.

Study	Managed surgically									Managed nonsurgically		MINORS
	Asymptomatic				Symptomatic							
	n	c	Mean LOS	Mean age at surgery (range)	n	c	Mean LOS	Mean age at symptoms (range)	Mean age at surgery (range)	n	Mean length of follow-up (range)	
Aziz 2004 [23]	15	2	4	9 m (2 m–2 y)	15	4[a]	6[b]	NR	5 y (7 m–15 y)	17	36m[c] (5 m–9y)	14
Chow 2007 [24]	6	3	NR	5.4 m (22 d–20 m)	1	1	NR	5 m (−)	6 m (−)	5	22 m (3 m–2.8 y)	8
dell'Agnola 1996 [10]	7	0	NR	4.3 m (7 d–7 m)	1	1	NR	NR	6 m (−)	0	–	7
Evrard 1999 [25]	6	0	12.7	10.5 m (9 d–3 y)	6	3	19.2	17 m (7 w–3.5 y)	81 m (7 w–27 y)	0	–	7
Keidar 2001 [26]	6	2	4.6	9.8 m (NR)	0	–	–	–	–	0	–	9
Marshall 2000 [11]	7	0	4.2	8.3 m (2 d–16 m)	9	3[a]	12.9[b]	21 m (1 m–7y)	35.9 m (1 m–15 y)	0	–	12
Raychaudhuri2011 [27]	14	0	7.2	11.1 m (3.3–34 m)	0	–	–	–	–	5	NR	9
Sueyoshi 2008 [12]	8	0	15	0.52 m (±6 d[d])	13	0	13	26.8 m (±26 m[d])	41.8 m (±44 m[d])	6	21.6 m (10–40 m)	13
Wong 2009 [14]	1	0	NR	1 m (−)	18	8	NR	NR	48 m[a] (1 m–13 y)	2	6.5 m (6–7 m)	8

n: number of patients, c: number with complications, LOS: length of stay in hospital, m: months, y: years, d: days, NR: not reported, w: weeks, −: not applicable.
[a] No significant difference between groups, as reported by the original study.
[b] Asymptomatic surgery patients had significantly shorter LOS than symptomatic patients, as reported by the original study.
[c] Study reported median rather than mean.
[d] Study reported standard deviation rather than range.

2.3. Elective surgery versus expectant management

One hundred sixty-eight patients were asymptomatic at birth with sufficient data available for inclusion in our analyses. Just less than half of these patients (n = 70, 41.7%) underwent elective surgery with 7 (10.0 %) cases of postoperative complications. The remainder were managed expectantly (n = 98, 58.3%), with 63 eventually developing symptoms between one month and seven years of age and consequently requiring surgery (64.3%); 20 experienced postoperative complications (31.8%). Details of the complications experienced by patients in each study are provided in Table 2. The remaining 35 patients (35.7%) continued to be managed nonsurgically at final reported follow-up. Regarding these expectantly-managed patients, we were able to combine data from two studies to calculate a median and mean follow-up of 17 months and 18 months respectively for seven patients [14,24]. One additional study reported a median follow-up length of three years (range: 5 months to 9 years) for 17 patients [23], while another reported a mean follow-up length of 21.6 months (range: 10 to 40 months) for six patients [12], but neither provided individual patient data. A fifth study reported that 11 patients were being followed nonsurgically but did not report length of follow-up [27]. There were no deaths related to the condition or surgical complications of lung resection in any of our included patients.

We were able to pool the results from six of the included studies that reported on postoperative morbidity [10,11,14,23–25]. The results of this meta-analysis showed that total morbidity (number of patients who experienced postoperative complications) was significantly higher when surgery was performed after symptom development compared to resection when patients were asymptomatic (OR 4.59, 95% CI 1.40 to 15.11, P = 0.01). Heterogeneity was low ($Chi^2 = 2.29$, df = 5 (P = 0.81); $I^2 = 0\%$; Fig. 2).

We were also able to pool the results from three studies that reported on length of stay [11,12,25]. There was no significant difference between patients who had surgery while asymptomatic and those who had surgery after the development of symptoms (MD 4.96, 95% CI −1.75 to 11.67, P = 0.15), although the trend favored asymptomatic patients. It should be noted that this analysis only included a total of 48 patients, and heterogeneity between studies was fairly high ($Chi^2 = 5.37$, df = 2 (P = 0.07); $I^2 = 63\%$; Fig. 3).

Table 2
Intra- and postoperative complications in patients managed surgically.[a]

Study	Asymptomatic	Symptomatic
Aziz 2004 [23]	1 pneumothorax 1 pleural effusion	2 pneumothorax 1 wound infection 1 bronchopleural fistula
Chow 2007 [24]	2 pneumothorax 1 chylothorax 1 right phrenic nerve palsy	1 pneumothorax 1 pleural effusion
dell'Agnola 1996 [10]	0	1 transfusion
Evrard 1999 [25]	0	2 prolonged air leak 1 pneumothorax 1 pleural effusion 1 residual CPAM
Keidar 2001 [26]	2 residual CPAM	–
Marshall 2000 [11]	0	2 bronchopleural fistulas 1 hemorrhage (required re-exploration)
Raychaudhuri 2011 [27]	0	–
Sueyoshi 2008 [12]	0	0
Wong 2009 [14]	0	3 transfusion 2 pneumothorax 2 sepsis 1 bronchopleural fistula 1 pneumonia 1 residual CPAM 1 bleeding

0: no complications; CPAM: congenital pulmonary airway malformation; −: not applicable.
[a] Multiple complications may have been present in a single patient.

3. Discussion

3.1. Summary and comparison with the literature

With the wide use of antenatal ultrasound, CPAM is often diagnosed before birth, and may remain asymptomatic for years. Whether asymptomatic children should undergo elective lung resection is disputed [6,7]. We have attempted to compare patients who underwent resection of CPAM lesions while asymptomatic to those who were managed expectantly and had surgery only upon the development of symptoms, in terms of morbidity and length of stay in hospital.

We identified nine studies reporting on a total of 168 eligible patients with CPAM who were asymptomatic at birth. Just less than half of these patients underwent elective surgery, with approximately 10% experiencing postoperative complications. The rest were managed expectantly, with 64 eventually requiring surgery owing to the development of respiratory symptoms and more than 30% of these patients experiencing postoperative complications. Therefore, more patients who were symptomatic at the time of surgery had complicated recoveries compared to those who were asymptomatic when surgery was performed. Approximately 36% of patients who were managed nonsurgically remained asymptomatic at follow-up, however the length of follow up was relatively short (less than three years for more than half of the patients for whom length of follow-up was reported). Results of a meta-analysis of six

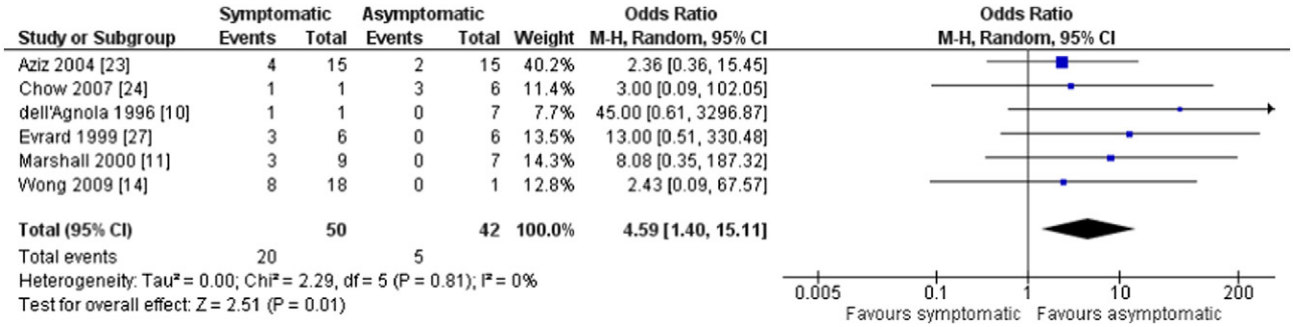

Fig. 2. Meta-analysis of postoperative morbidity for patients who were symptomatic versus asymptomatic at time of surgery.

studies reporting on morbidity confirmed a significant benefit of resection while the patient was asymptomatic compared to symptomatic, revealing that symptomatic patients were more than four times as likely to experience postoperative complications. There was no significant difference between groups in terms of length of stay based on a meta-analysis of three studies, although results were quite heterogeneous.

Our results ring true, as it is common knowledge among surgeons that operating on a healthy patient generally leads to a better outcome. If a CPAM patient has experienced lung infections, surgery will be more difficult owing to inflammation and scarring of the lung tissue, and subsequently a more complicated recovery may follow. In the majority of pediatric surgical cases, of course, the need for surgery cannot be anticipated, and therefore there is no choice but to operate after the onset of symptoms. However in cases of antenatally diagnosed CPAM, surgeons have the opportunity to perform surgery on healthy patients before symptoms appear.

In fact, our results are similar to those of a systematic review and meta-analysis comparing elective and emergency surgery for all types of congenital cystic lung lesions [15], finding that surgery after symptom development is associated with more complications than elective surgery. In our series, close to two-thirds of the expectantly managed patients eventually developed symptoms and therefore required surgery, suggesting that nonsurgical management is not a viable option in many cases. Stanton reported that only 3.2% of expectantly-managed patients became symptomatic, however it appears that patients were only followed for less than one year [15]. In our series, many children who became symptomatic were well more than one year of age. When it came to length of stay, patients in the Stanton review who were asymptomatic at surgery were favored, although not significantly so [15]. If the number of included patients was larger, this difference may have become significant, as it would be expected that a complicated recovery would extend the length of hospitalization.

In addition to the risk of postoperative complication, a consideration for choice of management of asymptomatic CPAM lesions concerns whether elective surgery or expectant management may impact on lung function as the child grows. It has been long believed that early resection may allow for compensatory lung growth, although it has been demonstrated that children who had lung resection at a later age had pulmonary function comparable to that of children who underwent early resection [28]. Still, early resection is safe [29], and the presence of lesion-associated symptoms or lung infection at the time of surgery is associated with decreased future lung function [30,31], indicating that early resection, before the development of symptoms, is probably the better choice. In addition, although it is unclear whether the risk of malignancy in CPAM patients is greater than that in the general population [1,32], or whether the excision of a CPAM lesion prevents the risk of future malignancy [33], there is a small risk of a CPAM lesions disguising a malignancy, thereby delaying its identification and treatment. Pleuropulmonary blastomas have been diagnosed postsurgically in large CPAM lesions and may be indistinguishable from CPAM on imaging [4,34], and bronchoalveolar carcinomas have been found alongside CPAM lesions [1,35–37], further lending support to the argument that early elective resection is preferable. In addition, the increased radiation exposure associated with serial imaging to follow expectantly managed patients may increase malignancy risk [1,32].

3.2. Limitations and future directions

Our results should be interpreted with caution owing to the nature of the included studies (in that all but one were retrospective). We did not identify any RCTs evaluating the benefit of elective surgery compared to expectant management of asymptomatic CPAM lesions. As no RCTs exist on this topic thus far, it is unlikely that one of necessary size and scope will be forthcoming. Instead, large-scale, prospective databases of CPAM patients with long-term follow-up may be a more feasible alternative, and it has been suggested that national patient registries be instituted [7]. Another limitation is the heterogeneity in baseline characteristics between the studies owing to the different inclusion criteria of each study, although the studies included in the meta-analysis of morbidity were fairly heterogeneous in terms of results and study design. Finally, it is likely that some degree of reporting bias exists, however this is difficult to assess, especially with a small number of included studies [19,22].

4. Conclusions

Based on our results, we recommend elective resection of asymptomatic CPAM lesions, rather than expectant management. Lung resection in an asymptomatic patient is quite safe, prevents the risk of symptom development later in life, and may help prevent the development of malignancy. The ideal timing of elective resection will be the topic of a future review.

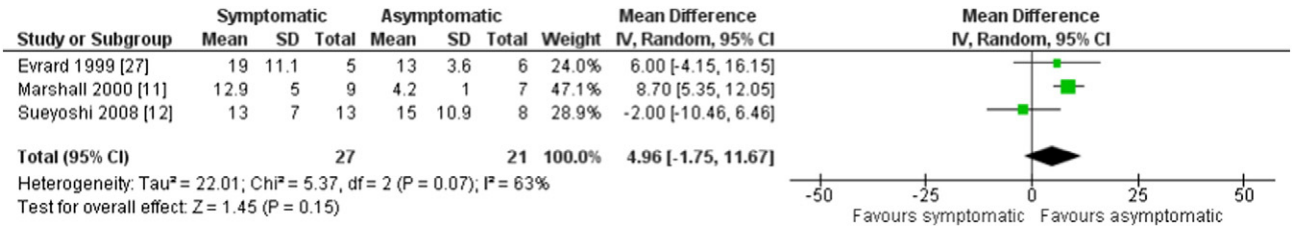

Fig. 3. Meta-analysis of postoperative length of stay for patients who were symptomatic versus asymptomatic at time of surgery.

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