



## Decision-Making Criteria for Observational Management of Congenital Pulmonary Airway Malformations (CPAMs)

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### ARTICLE INFO

#### Article history:

Received 12 January 2018

Accepted 1 February 2018

#### Key words:

Congenital pulmonary airway malformations

CPAM

Observational management

### ABSTRACT

**Purpose:** The purpose of this study was to determine practice patterns of Canadian surgeons managing congenital pulmonary airway malformations (CPAMs) and factors influencing practice.

**Methods:** Pediatric surgeons in Canada were surveyed regarding their experience, evaluation, and management CPAMs, and what factors they feel qualify patients for observation vs resection. Data were summarized, and Fisher's-Exact and Kruskal-Wallis Tests applied where appropriate.

**Results:** Sixty eight percent (n = 46) of surgeons responded. However, three surveys were incomplete and excluded. The median age of initial assessment by a pediatric surgeon was one month. 98% (42/43) use CXR for initial imaging, and 83% (36/43) recommend CT scan for further evaluation. Observation is offered always, almost always, or sometimes by 2%, 35% and 37%, respectively. Only 16% almost never, and 9% never offer it. Years in practice was not associated with this decision (p = 0.41). Of surgeons who offer observation, 78% (28/37) use morphology to guide their decision, and 63% (21/37) use lesion size (<1 cm to <5cms). 68%(23/37) consider the number of lesions, and 61%(14/23) of those only offer observation to solitary lesions.

**Conclusion:** Most pediatric surgeons in Canada offer observational management to patients with asymptomatic CPAMs. While practice variations exist, detailed imaging with a CT scan early in life to determine the morphology, size, and number of lesions guides practice.

**Level of Evidence:** V

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## 1. Background

Congenital pulmonary airway malformations (CPAMs) are estimated to occur at rates ranging from 1 in 10,000 to 1 in 35,000 pregnancies [1]. Their natural history is poorly defined, partly due to the tendency for CPAMs to be surgically resected early in life. The pathogenesis and prognosis remain controversial, leading to debate regarding management [2].

Ninety-five percent of CPAMs are identified on antenatal ultrasound [3]. In most cases, standard delivery with postnatal evaluation is performed, as few CPAMs cause fetal or neonatal complications [4]. Postnatal assessment often includes a thorough physical examination, a chest X-ray and a thoracic CT scan or MRI [1,5,6]. Most children with CPAMs are asymptomatic at birth [7,8], however approximately 20–40% of infants with CPAMs experience varying degrees of respiratory distress in the immediate postnatal period [9]. If surgical resection is recommended, it typically involves a lobectomy or segmental resection, performed via thoracotomy or thoracoscopy [6].

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No optimal approach has been established for the management of asymptomatic CPAMs. Rather than excising an asymptomatic CPAMs prophylactically, some surgeons prefer conservative or expectant management where lesions are observed with follow-up and resected if symptoms develop [10]. The debate of how to best manage asymptomatic CPAMs is largely focused on weighing the risks of living with a CPAM against the risks of surgery [8]. In 2006, a survey polling Canadian surgeons identified interinstitutional and intrainstitutional variability in CPAM management practices [10]. Overall, most Canadian surgeons (67%) pursued resection of asymptomatic CPAMs, and performed the resection when the patient was 2 to 18 months old [10]. For surgeons who preferred an observational approach (33%), follow-ups ranged from every 3 months to yearly [10]. The study herein aims to build upon the 2006 study, by identifying Canadian pediatric surgeons' current approach to the management of CPAM lesions. Despite surgeons previously choosing surgical or observational management, it is suspected that many surgeons employ a selective approach, offering surgery and observational management on a case-by-case basis. The primary objective of this work, therefore, is to identify what key features Canadian pediatric surgeons use to direct management of patient's with CPAM lesions. The results of this study help guide future decision-making in the treatment of patients with CPAM lesions.

## 2. Materials and Methods

Ethics approval for this study was obtained through the IWK Health Centre Research Ethics Review Board (REB#1021682). Pediatric surgeons practicing in Canada, registered through the Canadian Association of Pediatric Surgeons (n = 68) were surveyed using a 32-item questionnaire (Table 1). The questionnaire was distributed to participants via email on three occasions between November 2016 and January 2017. The questionnaire was designed to establish: (1) what methods and modalities surgeons use for the initial evaluation of infants with CPAMs and at what age these are obtained; (2) whether surgeons recommend observation, or surgical resection, for some or all lesions; (3) what methods and modalities are used for follow up, and at what frequency when observational management is recommended; (4) whether a minimally invasive or open approach is offered for surgical management; (5) what factors surgeons consider to qualify patients for either observational management with follow up or surgical resection; and (6) whether the experience of the surgeon or their location of training influenced their approach to management. In this survey, observational management is defined as patient follow-up without planned resection of the asymptomatic CPAM. It was assumed that surgeons offer resection for symptomatic CPAMs.

A draft survey was tested on four surgeons practicing at the IWK Health Centre in Halifax, Canada and minor adjustments were made

after their feedback. Data collected from the finalized survey were analyzed and presented with summary statistics. Fisher's Exact and Kruskal-Wallis Tests applied when comparing groups for differences. A p value of 0.05 was considered statistically significant.

## 3. Results

A final response rate of 68% (46/68) was obtained. Three surveys were more than 50% incomplete and were therefore discarded from the analysis. The majority of respondents were Canadian-trained (88%) and 100% are currently practicing in Canada. The experience of the surgeons varied with 16% having been in practice less than 5 years and 34% more than 20 years. 100% report managing infants with CPAMs as part of their practice with 56% estimating they see between 3 and 5 consults for CPAM/year.

The median age at which pediatric surgeons report recommending initial assessment is one month (IQR 1,2) with 21% recommending evaluation at birth and 44% within 1 month of birth. Ninety-eight percent of surgeons surveyed recommend a chest x-ray as the initial imaging modality with 57% obtaining this at birth and the remainder within the first month. Eighty-three percent of respondents recommend a chest CT scan to evaluate the lesion and the median recommended age for this was 4 months (IQR 3,6). Only 7% and 20% of surgeons utilize MRI or US to evaluate CPAMs, respectively.

**Table 1**  
Survey questions distributed to members of the Canadian Association of Pediatric Surgeons via SelectSurvey.NET.

Theme	Question
Demographics	For how many years have you been practicing Paediatric Surgery? Where did you complete the majority of your Paediatric Surgery training? Do you currently practice Paediatric Surgery in Canada?
Experience	Do you manage congenital pulmonary airway malformations (CPAMs) in your practice? Approximately how many CPAMs do you see in consultation per year?
Initial Assessment	In patients diagnosed with a CPAM on prenatal imaging, who are asymptomatic at birth: • At what age do you recommend initial assessment occur by a Pediatric Surgeon? • Do you obtain a CXR to evaluate the lesion? • Do you obtain an US to evaluate the lesion? • Do you obtain a CT to evaluate the lesion? • Do you obtain a MRI to evaluate the lesion?
Surgical Management	If recommending resection for asymptomatic CPAMs, do you: (a) Perform it yourself, (b) Refer to a colleague in your centre, (c) Refer to a colleague at another centre (d) I do not recommend resection for any asymptomatic CPAM If performing resection, what is your first choice of approach? (a) Open/ Thoracotomy, (b) Minimally invasive/ Thoracoscopic, (c) I do not perform resections for CPAMs If performing resection of symptomatic or asymptomatic CPAMs, do you: (a) Perform a lobectomy of the affected lobe whenever possible, (b) Perform a wedge resection of the affected lobe whenever possible (c) I do not perform resections for CPAMs.
Observational Management	Do you ever offer observational management for asymptomatic CPAMs? If you offer observational management for asymptomatic CPAMs, do you offer: (a) Regular evaluation by a Paediatric Surgeon (History and Physical Examination to monitor for symptoms +/- Imaging), (b) Regular evaluation by the Primary Care Physician (History and Physical Examination to monitor for symptoms, +/- Imaging), (c) I do not offer an observational approach If you offer observational management for asymptomatic CPAMs, when would you prefer to see the infant/ child for your initial assessment? (a) Prior to discharge, (b) < 1 month old, (c) 1–2 months, (d) 3–4 months, (e) > 4 months, (f) I do not offer observational management of asymptomatic CPAMs If you offer observational management for asymptomatic CPAMs, do you obtain regular-repeat imaging of the lesion? (a) Yes, (b) No, (c) I do not offer an observational approach
Criteria for CPAM management	The size of an asymptomatic CPAM influences my decision to offer observational management rather than resection. (a) Yes, (b) I do not use size to determine eligibility for observational management, (c) I do not offer observation to any asymptomatic CPAMs If yes, I feel observational management is indicated for asymptomatic CPAMs: (a) < 1 cm, (b) < 2 cm (c) < 3 cm, (d) < 4 cm, (e) < 5 cm The timing of diagnosis of an asymptomatic CPAM influences my decision to offer observational management rather than resection: (a) Yes, (b) I do not use the timing of diagnosis to determine eligibility for observational management, (c) I do not offer observation to any asymptomatic CPAM • If yes, I feel observational management is indicated for asymptomatic CPAMs: (a) Identified prenatally, (b) Identified after delivery The morphology (cyst size, the presence of solid components and/or single or multiple lobes involved), as observed on initial CT scan/ MRI or ultrasound, influences my decision to offer observational management rather than resection. (a) Yes, (b) I do not use the morphology to determine eligibility for observational management, (c) I do not offer observation to any asymptomatic CPAMs If yes, I consider the following important considerations of morphology: Select all that apply: (a) Cyst size, (b) The presence of a solid component, (c) Whether the lesion involves single or multiple lobes The number of lesions, as observed on initial CT scan/ MRI or ultrasound, influences my decision to offer observational management rather than resection for asymptomatic CPAMs: (a) I only offer an observational strategy to solitary lesions, (b) I will offer and observational strategy when multiple lesions are present, (c) I do not use the number of lesions to determine eligibility for observational management, (d) I do not offer observation to any asymptomatic CPAMs If multiple asymptomatic CPAMs are identified on initial imaging and I am offering resection, I: (a) Resect all lesions if possible, (b) Resect the dominant lesion and any small lesions that are immediately accessible during that surgery, (c) Only resect the dominant lesion I use a completely different strategy when deciding which CPAMs can be observed: (a) Yes, (b) No

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