**Background**

- Congenital cystic adenomatoid malformations (CCAM) is relatively rare congenital anomaly where a portion of the lung is replaced by a piece of non-functioning cystic lung tissue.
- The presentation of CCAM ranges from respiratory distress at birth to entirely asymptomatic lesions. 1,2
- Surgical intervention has long been recommended for management of symptomatic lesions due to the associated morbidity and mortality. 4
- When cases of known cystic lung disease are asymptomatic the ideal management is less defined.
- An increasing proportion of neonates are presenting asymptotically due to identification on antenatal ultrasound, presenting a significant challenge to pediatric surgeons.

**Rationale and Objective**

- Asymptomatic CCAM is an increasingly prevalent challenge in pediatric general surgery
- Debate persists over the optimal management of asymptomatic lesions
- Prior reviews of CCAM management exist but are largely non-systematic in nature
- To the best of our knowledge, no high quality systematic reviews comparing management approaches for CCAM currently exist.

Our objective was to perform a systematic review and meta-analysis to evaluate and compare the risks of elective surgery versus expectant management for cases of asymptomatic CCAM in children.

**Methods**

- **Search**
  - Electronic search of the Cochrane Central Register of Controlled Trials (CENTRAL), MEDLINE, EMBASE, CINHAL and PUBMED to identify relevant primary studies and reviews on the treatment of CCAM in neonates and children.
- **Hand Search**
  - The search was expanded by performing a hand search of the references of all included studies.
- **Review**
  - Both abstracts and full texts of retrieved articles were reviewed for inclusion in a two-tier review process by two independent reviewers. Where consensus on an article could not be reached a third reviewer was consulted.

**Results**

- 262 patients were symptomatic at birth requiring immediate surgery
- 641 patients were asymptomatic at birth and were managed surgically before or after symptom development or were managed conservatively (1 patient was lost to follow up before treatment)
- 389 of the asymptomatic patients underwent early elective surgery with 43 experiencing post-operative complications
- 151 of the asymptomatic patients did not undergo early surgery and went on to develop symptoms at a mean age of 14.6 months. These patients underwent surgery after developing symptoms with 54 experiencing post-operative complications
- 99 patients were managed conservatively and all remained symptom free at a mean follow up of 37 months.

**Figure 2. Percentage of study population who received each management course**

**Figure 3. Meta Analysis Results**

**Conclusions**

- Incidence of symptom development in asymptomatic patients treated with a conservative approach was 23.6 %.
- Post-operative morbidity is higher when surgery is performed after symptoms develop compared to those resected while the patient is asymptomatic.
- Overall study quality was low suggesting there is a need for more high quality studies to be done in the area of CCAM management.

**Quality Assessment**

- Two researchers independently assessed each study using the Methodological Index for Non-Randomized Studies (MINORS)
- All studies but one were retrospective in nature
- No studies employed a blinding technique for data collection
- No studies did a prospective calculation of study size
- Methodological biases are reflected in total MINORS scores
  - Range: 7 to 16
  - Median: 8.5
- Maximum score for non-comparative studies is 16, maximum score for comparative studies is 24

**References**