In-utero pulmonary drainage in the management of primary hydrothorax and congenital cystic lung lesion: a systematic review
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CRD summary
The authors concluded that in-utero pulmonary drainage in foetuses with congenital pulmonary cystic malformations was associated with improved foetal survival among foetuses with hydrops foetalis. This was generally a well-conducted review, but the authors’ conclusion does not adequately reflect the limitations of the small number of poor-quality, biased observational studies.

Authors' objectives
To evaluate the effects of in-utero pulmonary drainage on perinatal survival in foetuses with primary hydrothoraces and/or congenital cystic lung lesions.

Searching
MEDLINE, EMBASE and the Cochrane Library were searched from inception to 2004, without any language restrictions; the search terms were reported. In addition, the references lists of known reviews and primary studies were screened. No attempts were made to locate unpublished studies.

Study selection
Comparative studies (randomised or observational) were eligible for inclusion; case series were excluded. All of the included studies were comparative observational studies.

Specific interventions included in the review
Studies that evaluated methods of in-utero drainage were eligible for inclusion. The included studies used different methods of in-utero pulmonary drainage: thoracocentesis, thoracoamniotic shunt, surgery and other. The timing of the intervention varied among studies but was not always reported.

Participants included in the review
Studies of pregnancies with foetuses with pulmonary pathology were eligible for inclusion. Most of the foetuses in the included studies had cystic lung lesions, mainly congenital cystic adenomatoid malformation or pulmonary sequestration.

Outcomes assessed in the review
Studies that evaluated gross perinatal survival were eligible for inclusion in the review.

How were decisions on the relevance of primary studies made?
Two reviewers independently selected studies for inclusion in the review.

Assessment of study quality
Two reviewers independently assessed validity using the following criteria: study design, control for confounding, method of data collection, blinding of the assessors, adequacy of description of intervention, and ascertainment of outcome (>90% follow-up or not).

Data extraction
One reviewer extracted the data and a second reviewer checked the extraction. Outcome data for overall perinatal survival were entered into 2x2 tables of treatment versus control, with participants stratified according to the presence or absence of hydrops foetalis.

Methods of synthesis
How were the studies combined?
Pooled odds ratios (ORs) and corresponding 95% confidence intervals (CIs) were calculated for treated versus untreated foetuses using a fixed-effect meta-analysis.

How were differences between studies investigated?
Clinical and methodological differences between the studies were discussed. Statistical heterogeneity was evaluated using the chi-squared test. Summary estimates for survival were also calculated separately for foetuses with and without hydrops foetalis; for studies that predominantly treated foetuses with hydrops foetalis and those that did not treat foetuses without hydrops foetalis; studies with no clear description of foetal hydrops; and for three different types of treatment (thoracocentesis, shunting and surgery).

**Results of the review**
Sixteen cohort studies with control groups were included in the review (n=608).

The methodological quality of the included studies was generally poor. None of the studies controlled for confounding or blinded the outcome assessors. Three studies collected data prospectively, nine reported retrospective data collection, and four did not report data collection methods. Four studies (three prospective and one retrospective) used consecutive patients; details were not reported in the other studies. The interventions were inadequately described in 12 studies. Nine studies reported the duration of follow-up (range: 2 weeks to 7 years). All of the studies reported follow-up rates of more than 90%.

Pulmonary drainage significantly reduced survival rates compared with no drainage (OR 0.56, 95% CI: 0.32, 0.97, p=0.04).

In foetuses with hydrops foetalis, survival was significantly increased among treated compared with untreated foetuses (OR 19.28, 95% CI: 3.67, 101.27, p=0.0005; 3 studies, n=57). In foetuses without hydrops foetalis, survival was significantly reduced among treated compared with untreated foetuses (OR 0.04, 95% CI: 0.01, 0.32, p=0.002; 2 studies, n=142 only 9 of whom were treated). There was no evidence of statistical heterogeneity. In most studies treatment was only given to foetuses with hydrops foetalis; foetuses without hydrops foetalis were treated conservatively.

**Authors' conclusions**
In-utero pulmonary drainage in foetuses with congenital pulmonary cystic malformations was associated with improved foetal survival among foetuses with hydrops foetalis.

**CRD commentary**
The review addressed a clear question that was defined in terms of the participants, intervention and outcomes; inclusion criteria for the study design were broad, but this was appropriate given the limited evidence identified. Several relevant sources were searched and attempts were made to minimise language bias. No attempts were made to minimise publication bias. Methods were used to minimise reviewer error and bias in the study selection, validity assessment and data extraction processes. Validity was assessed and the results discussed.

The data were combined in a meta-analysis, statistical heterogeneity was assessed, and various subgroup analyses were conducted. In their discussion the authors acknowledged the limitations of the evidence, but this was not reflected in their conclusion. This was generally a well-conducted review, but the authors’ conclusion does not adequately reflect the limitations of the small number of poor-quality observational studies.

**Implications of the review for practice and research**
Practice: The authors did not state any implications for practice.

Research: The authors stated that the review findings should help in the design of a randomised controlled trial to evaluate in-utero pulmonary drainage.
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This is a critical abstract of a systematic review that meets the criteria for inclusion on DARE. Each critical abstract contains a brief summary of the review methods, results and conclusions followed by a detailed critical assessment on the reliability of the review and the conclusions drawn.