Mixed type CCAM presenting with ascites at 21 weeks: successful treatment with steroids and single thoracocentesis

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Objective

To present management options and outcome of a large Mixed type CCAM presenting with ascites at 21 weeks.

Methods

Case report.

Results

A large, mixed type (micro and macrocystic) CCAM was diagnosed at 21 weeks in pregnancy, occupying the whole right hemithorax. The lesion was 26 cm3, according to tridimensional volumetry (VOCAL) and CVR was 1. 67, with a markedly left mediastinal shift, hepatomegaly and mild fetal ascites (3. 3mm thick). The patient received a first course of steroids followed by mild increasement of the ascites (5. 9mm thick) and therefore a thoracocentesis was performed, with drainage of 11mL of fluid and immediate reduction of the lesion volume to about 14cm3. This was followed by a second course of steroids. The lesion size returned to its original volume in a one-week follow-up but CVR remained below 1. 6 up to 28weeks. From 30 weeks onwards, CVR increased progressively from 1. 7 at 30 weeks to 2. 8 at 37weeks. But there was no recurrence of fetal ascites nor other signs of hydrops and amniotic fluid volume remained normal, in spite of the lesion remaining large (103cm3 at 37 weeks) with considerable mediastinal shift. Delivery occurred at 38 weeks and the newborn presented mild dyspnea, prompting a right lower lobectomy one day after Birth, with confirmation of prenatal findings.

Conclusion

There is some evidence that steroids could be beneficial in large microcystic CCAM, while thoraco-amniotic shunting could be useful in selected macrocystic cases, when they present life-threatening ascites / hydrops. It is unclear whether prenatal interventions are beneficial in mixed (micro and macrocystic) CCAM. Shunting in early second trimester has a significant complication rate, in particular PPROM and catheter displacement. This case illustrates that a less agressive approach could be attempted in some mixed (micro and macrocystic) cases. It remains unclear if the good outcome could be credited to the steroids administration, to the thoracentesis or to both.