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OK-432 treatment of early fetal chylothorax. Pregnancy outcome and long-term follow-up of 14 cases

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Objective

Fetal chylothorax is a rare condition associated with lunghypoplasia and hydrops. Treatment options include thoracocentesis, thoraco-amniotic shunting and pleurodesis using OK-432. Knowledge on long-term outcomes after treatment with OK-432 is limited. The aim of this study was assess pregnancy and long-term outcomes of children treated in utero with OK-432 due to chylothorax in early second trimester.

Methods

Follow-up on pregnancies and children treated in utero with OK-432 during the years 2003-2009 at Copenhagen University Hospital, Rigshospitalet for chylothorax in gestational age (GA)16+0 to 21+6 weeks. Anamnestic information from parents, physical examination, pulmonary function test, neuro-paediatric examination and intelligence testing by Wechsler Intelligence Scale were used for evaluation.

Results

Fourteen cases were treated with OK-432 during the study period. None had spontaneous abortion or premature rupture of membranes (PPROM) and median GA at delivery was 38+5 (24+4- 41+5) weeks. Twelve children were eligible for follow up. The median age at follow-up was 11. 4 (7. 8-13. 8) years. Pulmonary function was normal in all children and Mean Full Scale IQ was 98. 2±13. 8, suggesting that as a group these children did not differ from the Full Scale IQ mean and standard deviation reported in normal children (100±15). Four children had a diagnosed medical condition, attention deficit disorder or genetic syndrome at the time of follow up and the testing confirmed these diagnoses. The remaining 8/12 children had normal follow-up examinations.

Conclusion

This study adds to the existing knowledge on long-term outcome after in utero treatment with OK-432 for fetal chylothorax. OK-432 treated children have comparable survival rates and long-term outcomes to those treated with thoraco-amniotic shunts but with a lower risk of procedure related PPROM and higher GA at birth. This follow-up study furthermore confirms that fetuses with seemingly isolated chylothorax have an increased risk of underlying disorders.