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Neurodevelopment in congenital diaphragmatic hernia survivors

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Objective

To our knowledge there is no data on the neurologic outcome comparing children with prenatally diagnosed with congenital diaphragmatic hernia (CDH), treated either antenatally or postnatally. In this study we evaluated the neurologic outcome of isolated left CDH survivors and correlated this to prenatal ultrasound findings, disease severity and fetal intervention.

Methods

This is a multicenter prospective cohort study performed in 4 experienced fetal treatment centers (Barcelona, Spain; São Paulo, Brazil; Leuven, Belgium; London, UK). Parents of children, diagnosed in utero with isolated left-sided CDH, and aged between 1 and 6 years at the moment of the study were invited to complete an age-adapted ages and stages questionnaire (ASQ). Prenatal variables (FETO, disease severity (observed/expected Lung-Head-Ratio; O/E LHR), gestational age at delivery, gender) were retrieved from the medical record, and demographics via the parents (education, profession).

Results

Of the 87 parents invited, 40 agreed to participate (46%), of whom 20 had FETO. Average age of children was 3.4 ±1.4 years. Overall, 38% children were reported to score below the cut-off in at least one domain, most frequently involving gross motor development (30%). There was no correlation between disease severity (O/E LHR) and abnormal neurologic ASQ-scores. Children who had FETO were born earlier (36.5±2.4 vs 38.0±1.2w; p=0.04) and had more severe pulmonary hypoplasia (o/eLHR: 30.2±5.1 vs 45.8±22.2; p=0.001). There was no difference in total ASQ-scores between children treated with FETO and those managed expectantly during pregnancy (39 vs 40%; p=0.9), neither in its subcategories (communication, gross and fine motor-function, problem solving, social interaction). There was no correlation between ASQ scores and severity of hypoplasia (o/e LHR) or liver herniation (up/down). There however was a correlation between ASQ result and gestational age at birth. Prenatal predicators for neurodevelopmental impairment were smaller AC and smaller FL.

Conclusion

In this cohort, children with prenatally diagnosed CDH were at increased for neurologic developmental delay. We did not find a correlation with disease severity. Children who underwent FETO prenatally, hence who had more severe hypoplasia and were born earlier, had comparable outcomes to those who did not. Children that were smaller on prenatal ultrasound had a higher risk of scoring below the cut-off on the ASQ. Further research needs to investigate if cerebellar size and MCA Doppler is able to predict better which fetuses are at risk.