Hydrops fetalis due to abnormal lymphatics

Sir,  

Windebank et al reported a case of hydrops fetalis due to abnormal lymphatics.1 The baby, a girl, died at the age of 66 days. We are not aware of any other reports of this condition. Her parents have now had a second female child, with features similar to the first, born 16 months later. This baby died age 1 hour. We write to report her clinical history.

The parents are members of a travelling family. The mother's previous obstetric history was one pregnancy lasting 42 weeks and delivering a well male infant, weighing 3500 g, a miscarriage at 12 weeks, and the girl reported by Windebank et al. In the current pregnancy she first attended the antenatal clinic at 18 weeks, and serial ultrasound scans from 20 weeks showed fetal ascites and massive and increasing oedema. She went into spontaneous labour at 32 weeks. The membrane rupture delivery interval was 2 hours and 45 minutes, and she was delivered vaginally of a hydropic female infant whose weight was 3010 g, length 41 cm. As with the previous pregnancy the placenta was retained, requiring manual removal under general anaesthetic. It weighed 725 g and was pale and oedematous.

The baby's heart rate was 60/min, but respirations were absent. She was intubated and ventilated. Intensive treatment, including external cardiac massage, failed to improve her condition, and she died at age 1 hour. Blood was obtained from an umbilical venous line. The baby's blood group was A positive, as was her mother's, haemoglobin concentration was 114 g/l and packed cell volume 42-0%; concentration of serum albumin was 18 g/l, serum sodium 130 mmol/l, serum urea 2-3 mmol/l, and serum calcium 2-31 mmol/l.

Postmortem examination showed massive ascites and large serous effusions in both pleural cavities with anatomically normal, but hypoplastic, lungs. Apart from oedema the urinary system appeared normal, as did the heart, the great vessels, the liver, and the spleen. Probably this baby had the same condition as her sister. It is of note that with each baby there was a retained placenta. This baby appears to have been the more severely affected.

Windebank et al, thought the parents not related.1 Questioning of the family at the time of the birth of this child, however, established that the baby's paternal grandmother and maternal grandfather are first cousins. This was probably overlooked by Windebank et al, and this cause of hydrops fetalis may represent a previously unreported autosomal recessive condition. Perhaps it should be looked for in travelling families.

References

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Hydrops fetalis due to abnormal lymphatics.

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