A Case of Non-isooimmune Hydrops Fetalis with Infantile Cortical Hyperostosis

Sir,

We wish to report an unusual case of non-isooimmune hydrops fetalis which was associated with infantile cortical hyperostosis—an association which has not been reported before to the best of our knowledge.

Our patient was delivered to a 38-year-old, gravida 4, para 4, Saudi female after 28 weeks gestation. Until the time of delivery pregnancy was uneventful and routine screening for TORCH, rubella and hepatitis were negative, she was Rhesus O positive.

The baby was delivered spontaneously by vertex vaginal delivery, weighing 1.3 kg and was visibly grossly oedematous with ascites. Liver and spleen were both enlarged and 4 cm below the costal margins respectively.

The baby was immediately intubated and ventilated. Chest X-ray revealed hydrothorax; the femur on both sides and the radius and ulna on the right side showed generalized cortical hypertrophy more marked along the diaphysis. A detailed search was made for infection, metabolic and haematologic disease but all the investigations were negative.

With symptomatic treatment the baby fully recovered over an 8-week period. During these 8 weeks the bony changes on X-ray resolved completely. At follow-up, 14 weeks later, the baby was doing well and was both clinically and radiologically normal.

Hydrops fetalis until recently was mainly due to Rhesus iso-immunization. This has been reduced considerably with effective immunoprophylaxis with a concomitant relative increase in the incidence of non-isooimmune hydrops fetalis. In a brief review of the literature we could not find a case in which infantile cortical hyperostosis has been associated with hydrops fetalis.

This case illustrates that infantile cortical hyperostosis should be considered in the differential diagnosis of non-isooimmune hydrops fetalis, although the causative pathology remains a mystery.

References

Hydatid Cyst. An Unusual Presentation

Sir,

A 50-year-old Kuwaiti woman presented with a gradually increasing painless swelling of 30 years duration in the right supraclavicular area. She had no associated symptoms.

Examination revealed a well defined cystic, rounded swelling of 10 cm diameter in the right supraclavicular area. It was mobile, non-pulsatile, non-compressible and did not show any impulse on coughing. Regional lymph nodes were not enlarged. Systemic examination did not reveal any abnormality.

An X-ray of the chest revealed a soft tissue shadow in the right supraclavicular area. An X-ray of the chest was normal. A provisional diagnosis of lipoma was made and the swelling was excised. Histopathology of the specimen revealed it to be a hydatid cyst.

This is an unusual presentation for a hydatid cyst. It usually occurs at rates of 63% in the liver, 25% in the lung, 5% in muscles, 3% in the bones, 2% in the kidneys, 1% in the spleen and bones and 1% in other organs. The presence of a cyst peripherally is an indication to look for a cyst in the liver and lungs. Ultrasound of the abdomen and a chest X-ray in our case failed to reveal the presence of a cyst in these organs. Hydatid cyst can occur anywhere in the body from the big toe to the crown of the head. In areas where the occurrence of hydatid cyst is common e.g. Kuwait, the possibility of hydatid disease should be kept in mind when one is faced with any curious swelling.

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Utilization of Anti-epileptic Drug Levels in a Teaching Hospital in Saudi Arabia

Sir,

We read with interest the paper by Al-Ghamdi on Utilization of anti-epileptic drug levels in Saudi Arabia in a recent issue of this journal, and would like to make the following observations and comments.

In an earlier study in 1989 assessing the usefulness of monitoring anti-epileptic drug (AED) blood levels in epileptics under the usual out-patient care in the same institution, we showed a relatively poor correlation between AED blood levels and seizure control, and advanced the view that the absolute AED blood levels were poor indices for dosage adjustment although the values were useful for monitoring drug compliance in most instances.

Consequently we suggested that following the usual out-patient practice and under similar circumstances where factors such as timing of blood sampling and drug administration which can significantly affect AED blood levels cannot be adequately corrected for, clinical evaluation and seizure control were better indices for dosage adjustment than AED blood levels. We further advocated the need to identify specific

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