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

Sir,

We wish to report an unusual case of non-isoinmune 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 4 and 3 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-isoinmune 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-isoinmune hydrops fetalis, although the causative pathology remains a mystery.

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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 neck 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
indications for measuring AED blood level. It was therefore rather disturbing to read of such categoric statements by the authors as 'the rate of inappropriateness of measurement of AED levels was 86%'. Although the variables used in their audits criteria are acceptable in principle, their application by the authors seem incomplete. The quality of the data evaluated by them as regards reliability, and completeness of the information supplied in the requests, level of competence of those making the requests and under what circumstances these were made, were not addressed or commented on. These factors definitely influence the accuracy of their data and thus question the validity of their results and conclusions. For example, it would be difficult to make meaningful inferences on data where the indications for measurement were 'not stated' in more than half (55.8%) of the cases (Table 2). In addition, the degree of patient drug-compliance which has been shown to be as low as 65% in Saudi epileptics\(^5\) was not commented upon.

Furthermore, the social issue of varying patterns of clinician behaviour and other social factors including patient-initiated requests were not addressed or recorded at the 14th Congress Limitations to their study, it was gratifying to note that they have come to similar suggestions as ourselves as regards the need for defining adequately indications for monitoring AED blood levels in epileptics. They also reiterate the need for both standardization of methodology with the establishment of Reference Laboratories and a quality control scheme for AED in the Kingdom of Saudi Arabia.

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References

Vitamin D Status of Saudis

Sir,

I read the two articles by Dr Sedrani \textit{et al.}, in \textit{Saud Med J} 1992; 13(3), on the vitamin D status of Saudis. Basically, both articles had a common purpose—that of determining levels of plasma 25-OHD concentration of Saudis as a measure of vitamin D status of the population. One article dealt with regional differences in values for both males and females, whilst the other investigated the exact same group's problem with vitamin D levels except that they had been subdivided by age categories. Both articles intended to find some urban/rural differences in vitamin D status. The introduction, subjects studied, methods, and statistical analysis were all the same. Some of the tables in one article could have been combined or elaborated to include those of the other article.

It could have made a difference if some other relationships had been studied, for example, regional age/sex distribution and frequency of insufficiency of vitamin D, or regional socioeconomic levels and vitamin D status. Statistical analysis could have been expanded to include these relationships.

One such factor also mentioned in one of the articles was that nutritional habits play a role in determining adequate vitamin D status. It was suggested that intake of phytate-containing foods such as fruits and vegetables could cause the deficiency. What is more fitting than to do a dietary survey on a small subsample to check this possibility? Once the subjects were interviewed anyway, a short dietary recall of recent or past intake could have confirmed the role of food in explaining the low vitamin D serum levels. Most methods of evaluating nutritional status are based on the premise of a sequence of stages representing increasing severity of nutritional adequacy: inadequate dietary intake leading to exhaustion of adaptive mechanism, leading in turn to depletion of tissues, evidence of deficiency in blood and body fluids, and finally, anatomic and clinical lesions.\(^4\) More often than not, signs of clinical disease become more apparent as a direct consequence of an impaired metabolic or physiologic function because of a lack of or increased intake of certain nutrients.\(^5\)

Some cases in point could be the diet-serum lipid athero-sclerotic relationship or the fat-cancer postulated relationship. These two well known associations have been studied repeatedly by epidemiologists and evidence points to some positive correlations. My recently completed doctoral thesis on the effect of diet on the incidence of disease studied these above-mentioned relationships.\(^6\)

Some corollary findings were made and it was noted that while inadequate intake of food could produce clinical deficiency symptoms, excessive intakes as a result of affluence could affect the Saudi people in the worst ways. For example, it was found that cardiovascular disorders were positively correlated with lifestyle factors such as attending parties and the intake of sweets and meats and other expensive foods. Many investigators support the concept that rising affluence is widely regarded as an important causative factor in the epidemic of coronary disease that presently afflicts many Western industrialized societies.\(^5,6\)

Dietary surveys that set out to establish a group's nutritional status are best conducted in the most accurate way that utilizes biochemical data. This is the reason why the two articles by Professor Sedrani should be appreciated; but they could have made a good contribution to the field of nutrition in the Kingdom had they included a survey of food intake in order to serve as a check on whether or not other factors could explain the vitamin D deficiency apart from the limited sunshine.

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