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Correspondence

Benign paroxysmal torticollis in infancy

Sir,

I read with interest the paper by Deonna and Martin.[1] The syndrome may be a separate and new clinical syndrome, but we can only be certain if such patients are carefully screened for possible medication. It is essential to consider the differential diagnosis of a drug-induced dystonia. We have drawn attention to this problem previously.[2 3]

The statement made by Deonna and Martin that benign paroxysmal torticollis is 'unlike any other known form of intermittent torticollis' is not sufficiently substantiated. Their criteria do not exclude drug-induced torticollis. Case 3, reported by us in 1970,[4] fulfilled all these criteria (except for the patient's age), but was caused by metoclopramide. Our patient was aged 7½ years, but even an infant may be given dystonia-inducing drugs and generally the parents do not tell us unless we ask. The symptoms and signs of 'paroxysmal torticollis' are strikingly similar to those of drug-induced dystonia. They include abnormal rolling of the eyes (oculogyric crisis), retrocollis, curved trunk (tortipelvis), and neck pain. The clinical picture is too complex to be described with the restrictive label of paroxysmal torticollis. All patients with a paroxysmal torticollis need a detailed history, searching for dystonia-inducers. Experience taught us that enumerating these drugs for the parents (phenothiazines, butyrophenones, metoclopramide, etc.) is essential and that denial by them does not exclude this aetiology. In addition, careful observation and an accurate description of *all* accompanying symptoms are mandatory. Only by so doing will we help our patients, and contribute to clarifying the cause of these symptoms.

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MARIA CASTEELS-VAN DAELE
Department of Paediatrics,
Academisch Ziekenhuis Gasthuisberg,
University of Leuven,
B-300 Leuven, Belgium

Dr Deonna comments:

We agree that drug-induced acute dystonia, like many other conditions, should be included in the differential diagnosis of any paroxysmal postural abnormality in infants and children. We also agree that failure to obtain a positive history of drug ingestion (as in our cases) does not exclude this possibility. New drugs which are potential 'dystonia-inducers' are increasingly used in babies with gastrointestinal reflux, at an age when benign paroxysmal torticollis often starts. This was recently reported from our department.[1]

However, the postural abnormality seen in benign paroxysmal torticollis remains throughout the course of the attack and, unlike dystonic spasms due to a drug reaction,[1 2] does not seem to fluctuate. If one has not had the opportunity to observe the child carefully during the attack the recurrence of identical symptoms over many months in the absence of drug ingestion is the other strong argument in favour of benign paroxysmal torticollis. We have also paid attention to the other symptoms reported in association with paroxysmal torticollis and have been struck more by the presence of systemic disturbances (pallor, vomiting, pain) than by extrapyramidal signs.

Although we agree that the numerous causes of symptomatic torticollis should be ruled out before making the diagnosis of benign paroxysmal torticollis, enough clinical evidence has now accumulated to recognise it as a separate and new clinical syndrome of unknown, possibly diverse, aetiologies including migraine.

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Lipoatrophy in a patient on highly purified beef insulin

Sir,

Lipoatrophy at injection sites was reported in 10% of diabetics treated with conventional insulin preparations, but it was more common in girls and in young patients.[1] Treatment by the injection of monocomponent porcine insulin is the treatment of choice[2] either into the periphery or centre of the atrophic sites, although the latter is painful. I think that lipoatrophy in a patient treated exclusively with highly purified beef insulin has not previously been reported.

A 4-year-old girl developed diabetes mellitus and was stabilised on highly purified beef insulin zinc suspension (Neulente (Wellcome) 14 units daily). Her diabetic

control was unsatisfactory; she had occasional hypoglycaemic episodes and results of tests on the urine were poor. She generally resisted attempts to be restrained when being injected and developed a predilection for having injections in the lateral aspects of both thighs. Her dose of insulin was increased progressively to 20 units daily and after 8 months of treatment she was noted to have moderately severe lipoatrophy of injection sites. Because of this and her unsatisfactory control, she was changed to monocomponent porcine insulin with a mixture of Monotard MC and Actrapid MC (Novo Laboratories) once daily, and her mother was instructed to inject her in the periphery of the atrophic sites. During the next few weeks her dose of insulin stabilised to Actrapid MC 4 units and Monotard MC 8 units daily. Four months later her injection sites no longer showed lipoatrophy and her diabetic control was much improved, but she continued to co-operate poorly when being injected.

Reeves *et al.*[3] studied 14 patients with lipoatrophy at conventional insulin injection sites with a mean duration of treatment of 10 years (range 4 months to 10 years) and produced convincing evidence that lipoatrophy is the result of local immunological reaction to impurities in the insulin. The excellent response to monocomponent porcine insulin in these patients, together with the rarity of local reactions in patients treated exclusively with these insulins, lends strong support to this hypothesis. However, Jones *et al.*[4] recently reported the development of lipoatrophy in a patient treated exclusively with monocomponent porcine insulin (Monotard MC) which resolved on injecting a mixture of Actrapid MC and Monotard MC into atrophic sites. In their case there was no evidence of a local immunological reaction so that the cause of lipoatrophy in every patient is not known.

This report describes lipoatrophy developing in a diabetic girl treated with one of the new generation of highly purified beef insulins over a period of 8 months. I have subsequently treated another young girl with an almost identical presentation and outcome. Because of their purity, these insulins are thought to be unlikely to produce immunologically mediated reactions, but this case suggests that doctors will need to remain vigilant for the development of local injection site reactions in patients treated with highly purified beef insulin.

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L G Evans-Jones
Chester City Hospital,
Hoole Lane,
Chester CH2 3EH

Resuscitation of preterm babies at birth reduces the risk of death from hyaline membrane disease

Sir,

We were disturbed by the title of the paper by Robson and Hey.[1] Details of any method that reduces the risk of death from hyaline membrane disease should be published, but we do not think that the authors demonstrated the effect claimed.

Testing a hypothesis using two populations from different time periods is fraught with difficulties[2] and interpretation of significant results has often been challenged. In their study errors were made worse by interpreting a non-significant result as showing a cause-effect relationship. They reported that of infants of 1000–2000 g born between 1960 and 1967 there were 50 (10·7%) deaths out of 465 such births, whereas between 1971 and 1976 there were 23 (8·7%) deaths out of 264 low birthweight births. The authors stated that this represented a significant reduction, and we would like to know on what statistical test this conclusion is based. In fact, chi-squared with Yates's correction $= 0{\cdot}6$, df $= 1$, $P > 0{\cdot}5$.

We think that the following points should also be made: (a) Death from hyaline membrane disease bears a very strong relationship with gestation, but given gestation, birthweight has little part to play.[3 4] Preterm babies should *not* be defined by birthweight. (b) Comparison of risks pertaining in two groups should have ensured that in the two groups there were similar distributions of a number of background factors incontravertibly shown to be associated with death from hyaline membrane disease, particularly gestational maturity and whether delivered by caesarean section.[5] (c) The proposition that asphyxia is related to deaths from hyaline membrane disease rests largely on twin studies; the second twin is more likely than the first twin to suffer this fate, [6–8] especially if the interval between delivery of the two is longer than 30 minutes.[6] It seems possible that if this is important then obstetric management of labour, with increased use of oxytocics and shorter first and second stages, may have a more beneficial effect than the resuscitative methods employed after delivery.

Robson and Hey's claim may be correct but their data do not support it.

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