

clinical improvement was obtained after this was combined with trimethoprim-sulphamethoxazole (Hughes *et al.*, 1975). This is the first report from Japan of the treatment of the disease by trimethoprim-sulphamethoxazole. We favour the use of tracheal washings to demonstrate the agent, since the procedure is safe and the chance of finding the agent seems to be better than in sputum or gastric juice.

References

- Chan, H., Pifer, L., Hughes, W. T., Feldman, S., Pearson, T. A., and Woods D. (1977). Comparison of gastric contents to pulmonary aspirates for the cytologic diagnosis of pneumocystis carinii pneumonia. *Journal of Pediatrics*, **90**, 243-244.
- Cohen, M. L., and Weiss, E. B. (1971). Pneumocystis carinii pneumonia: percutaneous lung biopsy and review of literature. *Chest*, **60**, 195-199.
- Hughes, W. T., Feldman, S., and Sanyal, S. K. (1975). Treatment of Pneumocystis carinii pneumonitis with trimethoprim-sulfomethoxazole. *Canadian Medical Association Journal*, **112**, Special issue, 47S-50S.

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Identifying children 'at risk' from unexpected death in infancy

Sir,

The lay press has recently acclaimed the system which is in use in Sheffield for identifying children 'at risk' from unexpected death in infancy. Carpenter *et al.* (1977) point out, however, that this system may not apply in other communities. One of the purposes of the DHSS Multicentre Postneonatal Study is to discover whether a system based on Sheffield data might be equally applicable in other communities.

The obstetric and perinatal records of 234 cases of unexpected death in infancy have been studied in retrospect in 11 centres: Manchester, Liverpool, Leeds, Edinburgh, Oxford, Gateshead, Newcastle upon Tyne, Barnsley, Rotherham, Doncaster, and Birmingham. For each case a living control was chosen at random from those born on the same day and in the same centre as the index. Each case and control was 'scored' numerically according to the 'at birth' data analysis used in Sheffield (Carpenter *et al.*, 1977).

In all, only 49% of index cases scored 'at risk', together with as many as 27% of controls. This represents a low sensitivity, despite having over one-quarter of the population labelled 'at risk'. The poor discrimination between cases and controls would confer a low degree of efficiency on a prospective prevention programme. Although numbers in some centres were small, in only one centre was there case/control discrimination of the degree which is still shown in Sheffield.

I present these preliminary findings in advance of a more complete communication for several reasons; (1) Primary care teams could be led into a false sense of security concerning infants who did *not* score 'at risk', if prospective programmes are initiated prematurely using a system of low sensitivity. (2) The 'scoring' system used in Sheffield should be validated in other areas before use in those areas. It appears that the system may need revising before it can be of use in other communities. (3) Objective environmental data are not included in the present system. The subjective observation of the home environment which is used would be difficult to apply accurately elsewhere using multiple observers. (4) If an 'at risk' system is necessary to reduce the numbers of unexpected infant deaths in this country, a system should be derived with a high sensitivity, and which could be applied to many communities.

Finally, I would like to emphasise that the 'at risk' factors used in Sheffield do not have an aetiological basis, and that the aetiology of 'cot death' remains unexplained.

Reference

- Carpenter, R. G., Gardner, A., McWeeny, P. M., and Emery, J. L. (1977). Multistage scoring system for identifying infants at risk of unexpected death. *Archives of Disease in Childhood*, **52**, 606-612.

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Rickets in preterm infants

Sir,

We were interested to read Glasgow and Thomas's (1977) description of respiratory distress owing to rickets in 4 very small preterm infants. Over the past year we have diagnosed rickets (rachitic changes at the wrist metaphyses associated with raised levels of serum alkaline phosphatase) in 3 preterm infants but in none was rickets associated with respiratory problems. The Table gives salient clinical and biochemical details and an example of the severity of osteodystrophy is shown in the Fig. At the time of diagnosis the infants were receiving 400 IU supplementary vitamin D, which had been started 2 weeks after birth. Rickets was diagnosed in Cases 1 and 2 on finding craniotables at a routine follow-up examination and these positive findings prompted investigation of Case 3 who showed no clinical abnormality. None of the infants was in any way unwell when rickets was diagnosed. They were treated by increasing daily vitamin D intake. Cases 1 and 3 were given 1000 IU and Case 2 was given 2000 IU for one month with gradual reduction of the dose to 400 IU over the next 2 months. Within 3 months of diagnosis there were well defined radiological signs of healing in the 3 infants.

To explain their syndrome of subacute respiratory distress and severe metabolic bone disease, Glasgow and

Table Clinical and laboratory details of the 3 preterm infants who developed rickets

| Case no. | Sex | Gestation (w) | Birthweight (g) | Age at diagnosis (w) | Feeding | Laboratory investigations | | | |
|----------|-----|---------------|-----------------|----------------------|---|---------------------------|--------------------------|------------------------------|---|
| | | | | | | Ca (mmol/l) | PO ₄ (mmol/l) | Alkaline phosphatase (IU/l)* | 25 (OH) ₂ D ₃ (ng/ml) |
| 1 | M | 31 | 1300 | 10 | EBM for one month then Cow & Gate Premium | 2.44 | 1.88 | 1265 | — |
| 2 | F | 29 | 950 | 19 | Own mother's milk from birth | 2.52 | 0.95 | 3620 | 23 |
| 3 | M | 28 | 1225 | 22 | Own mother's milk from birth | 2.49 | 1.40 | 1765 | 12.2 |

*Normal paediatric level 56–190 IU/l (Forfar and Arneil, 1973).

Conversion: SI to traditional units—Calcium: 1 mmol/l \approx 4 mg/100 ml. PO₄: 1 mmol/l \approx 3 mg/100ml.

Thomas suggest that vitamin D deficiency causes softening and fracturing of the ribs and weakening of respiratory muscles which leads through impaired respiratory

movements to respiratory distress. If this is really the sequence of events perhaps it is surprising that the severe osteodystrophy in our infants was not associated with any respiratory problem. Admittedly the thoracic cage was not x-rayed so that we do not know for certain that the ribs were affected but since rickets is a generalised disturbance of bone growth it would be surprising if the ribs were spared. We wonder therefore whether some other factor is responsible for 'rachitic respiratory distress'.

Glasgow and Thomas draw attention to the association of copper deficiency and bone disease. Copper deficiency in preterm infants has been reported to cause enlargement of the costochondral cartilages, cupping and flaring of the long bone metaphyses, and spontaneous fractures of ribs, apnoeic episodes, and muscle hypotonia (Hambidge, 1976). The liver plays an important role in copper metabolism through the production of caeruloplasmin which is the main means of copper transport and in liver diseases plasma levels of copper are low (Alexander, 1974). In view of the disordered liver function in the Belfast infants we wonder therefore whether the syndrome of subacute respiratory distress with severe metabolic bone disease might be due at least in part to an abnormality of copper metabolism.

Finally Glasgow and Thomas postulate that their infants developed rickets because of malabsorption of vitamin D or impaired 25-hydroxylation of cholecalciferol in the liver. Plasma 25-hydroxycholecalciferol was measured in 2 of our infants and in both instances the levels were well within the normal range. It is unlikely therefore that malabsorption or impaired hepatic conversion was responsible for rickets. Instead it is tempting to speculate that transient interference of renal production of 1,25-dihydroxycholecalciferol or target organ unresponsiveness to this active hormone was responsible.

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Dr J. F. T. Glasgow comments:

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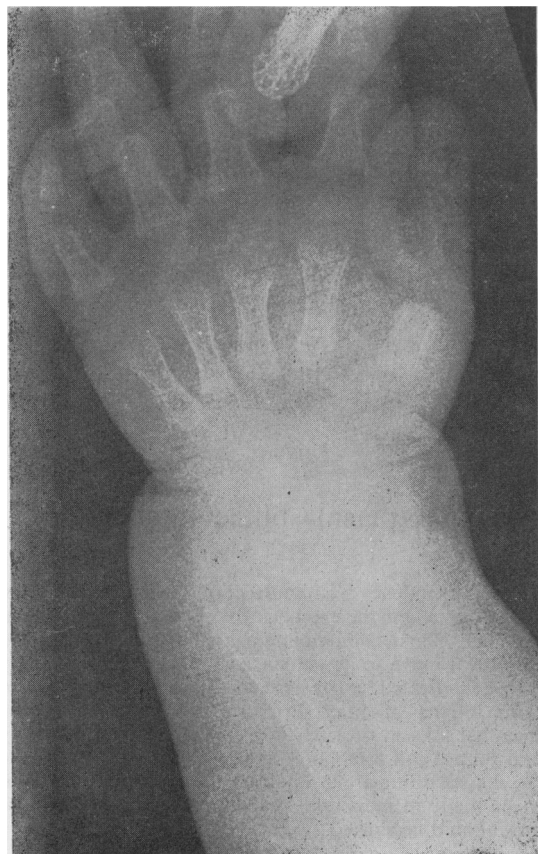


Fig. Case 2. X-ray of right wrist showing severe bone rarefaction with marked cupping of distal radial and ulnar metaphyses.