results are shown in the Table. In the Autolet group 3 infants did not wake at all during the procedure and a further 2, although awake, remained quiet with no increase in palmar water loss. There were no such infants in the heel prick group; in all infants palmar water loss showed a greater than 100% increase. Three infants in the manual group and 2 in the Autolet group required a second prick to obtain enough blood to fill in all 4 circles on the card.

Discussion

This study has shown that the Autolet is superior to the manual heel prick. Successful sampling was just as likely with the Autolet even though it was being used for the first time by many of the midwives. It was found to be popular, especially with the less experienced midwives who did not enjoy pricking heels.

The infants, too, preferred the Autolet method as judged by emotional sweating from the palm of the hand. Subjective assessment suggested that the infant cried less during the procedure and settled sooner. Indeed 3 of the 18 infants did not wake up. The Autolet heel prick is virtually painless; the discomfort of the procedure is largely due to holding the pricked heel while collecting the blood. After sampling with the Autolet the prick mark is virtually invisible. Repeat sampling—for example for frequent blood glucose estimations—is likely to cause less soreness of the heel.

We suggest that the Autolet method is an excellent way of obtaining heel prick blood samples in the newborn and could be used widely on postnatal wards, special care baby units, and neonatal intensive care units, in babies of any gestation.

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Correspondence to Dr N Rutter, Department of Neonatal Medicine and Surgery, Nottingham City Hospital, Hucknall Road, Nottingham NG5 1PD.

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Progressive inflammatory subglottic narrowing responsive to steroids

PETER PHELAN AND EDMUND HEY

Department of Thoracic Medicine, Royal Children’s Hospital, Melbourne, Australia

SUMMARY Four children aged between 2½ and 13½ years developed insidious subglottic stenosis of unknown cause over 3–12 months. In all, the initial diagnosis was asthma which resulted in inappropriate treatment. Endoscopically there was circumferential subglottic narrowing, and biopsy in 3 showed non-specific inflammatory changes. Corticosteroid therapy led to rapid and complete resolution.

Most cases of subglottic stenosis are the result of congenital malformations or of trauma from either an endotracheal tube or direct injury to the larynx. Insidious onset subglottic stenosis due to peri-chondritis, sarcoidosis, and Wegener’s granulomatosis has been reported in adults but these do not seem to have been recognised in children. There is one brief report of insidious onset subglottic narrowing of obscure origin in 2 children.

During the last 10 years, we have been concerned in the management of 4 children with progressive subglottic stenosis that did not seem due to any recognised disorder. In particular, the age at presentation and endoscopic and histological appearance were inconsistent with subglottic haemangioma which has been claimed to respond to corticosteroids but which usually presents with progressive...
subglottic narrowing in the first year of life. These 4 children are reported in order to draw attention to this entity and its striking response to corticosteroid therapy.

**Case reports**

The clinical features in these 4 children were remarkably similar (Table). In each child the onset was insidious over several months. The initial diagnosis was asthma which resulted in treatment for varying periods with bronchodilators. By the time of referral, severe dyspnoea was present owing to almost total laryngeal obstruction.

Cases 1 and 2 were otherwise normal. Case 3 had Hashimoto's thyroiditis and a tuberculin test with 13 mm induration after 5 units of PPD, but she probably had had a previous vaccination with BCG in Turkey. Case 4 later developed an area of histologically confirmed granulomatous panniculitis of the Weber-Christian type in the left upper eye-lid.

Inspiratory and expiratory flow volume curves demonstrated marked extra-thoracic tracheal obstruction in Cases 2 and 3 (Figure). Lateral radiographs of the neck in all patients showed marked subglottic narrowing and this was confirmed in Case 3 by a computerised tomography scan which demonstrated narrowing that was 18 mm long and 4 mm wide at its narrowest point. The obstruction did not extend more than 2 cm down the trachea in any patient. Other investigations were non-contributory.

Cases 1 and 4 had tracheostomies performed soon after presentation because of the degree of laryngeal obstruction. A biopsy was not performed in Case 2 because it was thought that reactive swelling might produce complete obstruction and the need for emergency tracheostomy.

All 4 patients were initially treated with oral corticosteroids in doses ranging from 30 to 60 mg prednisolone a day. In addition, Case 3 was given isoniazid. Response was rapid in all. After 48 hours there was symptomatic improvement which was confirmed by flow volume curves in Cases 2 and 3 and direct laryngoscopy in all.

It was possible to stop corticosteroids in Cases 1 and 2 after 4 weeks but there was a recurrence of the subglottic narrowing in Case 2 after 3 months. He again responded quickly to corticosteroids which were given for 4 weeks. These 2 patients were well when last seen 3 and 6 years after initial presentation.

Case 3 had a recurrence of symptoms and flow limitation on her flow volume curve when the prednisolone was reduced to below 15 mg daily after 2 months. After a further 6 months the dose was reduced to 5 mg on alternate days and the patient had a normal flow volume curve and was endoscopically normal. It is planned to stop steroids in the near future.

Case 4 has received prednisolone for 2 years, currently 5 mg on alternate days. He is asymptomatic and it is planned to stop the steroids in the near future.

**Discussion**

The nature of the subglottic disease in these children is unknown. The biopsy findings were essentially those of non-specific inflammation although in Case 4 there were granulomata present. There was no evidence of acute or chronic bacterial or viral infection. The inflammation was very sensitive to steroids.

Sharma and Lahori reported briefly two boys aged 2 and 3 years who developed sublottic obstruction over 3 months. Laryngoscopic findings and response to steroids were similar to those reported here. No biopsies were performed. These boys may have had the same condition as our patients.

While subglottic haemangiommas and lymphangiomas are of congenital origin, they may not present until the child is some months old. However, it would be extremely unusual for them to present after age 5 years and their endoscopic appearance is different from that observed in our patients. Neurofibroma and other fibromatous tumours can result in progressive glottic obstruction but the endoscopic and histological features are characteristic.

None of the children had clinical features sug-
gestive of the 3 conditions reported as being a cause of progressive subglottic narrowing in adults. Most reported cases of perichondritis with subglottic involvement have had associated inflammation of the nasal cartilages. Sarcoidosis is a rare disorder in children and usually there is generalised lymphadenopathy or changes on the chest radiograph. A diagnosis of Wegener's granulomatosis was entertained in Case 4 but there were no other features of the disorder although the boy did have granulomatous panniculitis. The relationship of this and of Hashimoto's disease in Case 3 to the subglottic lesion is unclear. There is a single case report of an adult with progressive non-traumatic subglottic narrowing but this did not respond to steroids.

It is important that paediatricians be aware of this disorder. These children all had findings typical of laryngeal obstruction although for months they were thought to have asthma, and were dangerously obstructed by the time they were finally diagnosed. The disorder appears to be very sensitive to corticosteroids and these should be used in patients suspected of having a similar condition before resorting to major surgery.

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Spontaneous hypothermia in a young boy

B K SANDHU, M BOMMEN, AND C G D BROOK

Middlesex Hospital, London

SUMMARY A 13-year-old boy who maintains his body temperature between 33 and 35°C was investigated. Tests of peripheral and central thermoregulatory function did not show a specific abnormality, except for some generalised hypothalamic dysfunction of unknown aetiology. He responded appropriately when his temperature was altered from its usual low level. His temperature regulatory mechanisms appeared to be set at a temperature lower than is normal.

Hypothermia is defined as a rectal or body-core temperature below 35°C. Accidental hypothermia is common—especially in the elderly—but spontaneous hypothermia is rare—particularly in young adults—if hypothyroidism is excluded. Several cases have been reported during the last 50 years, but studies of underlying central and peripheral thermoregulatory function are few, especially in children. We report results of investigations on a hypothermic 13-year-old boy.

Case history

The boy was born in India, a term normal delivery, birthweight 3·2 kg. He developed jaundice in the first 3 days of life which persisted for about 4 weeks. No blood tests were done or treatment given. He breast fed well and there were no other illnesses. He sat at age 8 months and walked at 19 months. He can communicate and read in his native language.

References


Correspondence to Prof P D Phelan, Department of Thoracic Medicine, Royal Children's Hospital, Melbourne 3052, Australia.

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