Treatment of pneumato
cosis cystoides coli by oxygen
inhalation

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SUMMARY Endotracheal intubation and intermittent positive pressure ventilation were used to overcome the difficulties of oxygen therapy for pneumato
cosis cystoides coli in a 9-year-old child with Down's syndrome and a congenital heart defect. As compared to previously reported cases, a shorter
duration of treatment and a lower arterial oxygen tension proved successful.

It has been known for a long time that cystic collections of gas may occur in the bowel wall of both
animals and man. The first description of the condition is attributed to du Vernoi in 1730, and John
Hunter described the first case in England (Koss, 1952; Kenney, 1963). In adults the gas is present in
cysts measuring up to a few centimetres in diameter lying beneath either the mucosa or serosa. The condi-
tion is known as pneumato
cosis cystoides intestinalis
or coli depending on the extent of disease process. It may be idiopathic (Koss, 1952) or associated with
other diseases including pulmonary disease (Botsford and Krakower, 1938; Doub and Shea, 1960), pyloric
stenosis (Dhall et al., 1968), malignant conditions
(Finney, 1908; Dale and Pearse, 1950; Williams et al., 1963; Thorpe, 1965), collagen diseases
(Meihoff et al., 1968, White et al., 1970; Mueller et al., 1972), adhesions from previous surgery
(Koss, 1952), and occurs after long-term administration of practolol
(Thein and Asquith, 1977).

Most reports of gas within the bowel wall of children are different as the gas is present as small
bubbles or linear collections (Stone et al., 1968). These are now recognized to be examples of necro-
tizing enterocolitis that occur mainly in the newborn
(Bell et al., 1971), though a similar appearance may be found in other conditions
(Robinson et al., 1974).
It is suggested that the term enteropneumatosis be
used to describe these collections of gas.

Four cases of pneumato
cosis cystoides coli have been
previously recorded in children. 2 cases were in girls
aged 13 and 16 years after abdominal surgery
(Koss, 1952) and 2 were associated with dermatomyositis
(Mueller et al., 1972; Oliveros et al., 1973).

Case report

A girl was born in 1966 at term, weighing 2.8 kg, and

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set to maintain normocarbia. Control of ventilation was facilitated with intermittent doses of phenoperidine and diazepam as required. Full monitoring was carried out including blood gas analysis, initially 3-hourly. The Pao₂ attained with an F₁O₂ 1.0 was between 208 and 237 mmHg (27.7–31.6 kPa). 24 hours later colonoscopy showed that the cysts had deflated. The child was extubated and the post-intubation period was uneventful. Her symptoms settled immediately and she has remained well for over 6 months with normal bowel actions and no soiling. Plain x-rays of the abdomen 24 hours and 10 weeks after treatment were normal (Fig. 4). She has returned to school and leads an active life.

Discussion

The aetiology of the cysts in pneumatosis cystoides is not known. The association with other gastrointestinal diseases, particularly obstructive lesions, has led to the suggestion that gas from the lumen of the bowel is forced through the damaged mucosa (Koss, 1952). Coincidental pulmonary disease indicates that the gas may pass through the mediastinum to the bowel mesentery, and support for this theory has come from animal experiments (Keyting et al., 1961). However, this does not explain the formation of submucosal gas cysts whose contents are related
to intestinal gas (Forgacs et al., 1973). Studies of the composition of the gas have shown it to be variable and have not shed any light on the aetiology (Kenney, 1963; Forgacs et al., 1973). It is unlikely that anaerobic organisms are involved in the production of the cysts as was suggested by Down and Castleden (1975) since perforation has not caused peritonitis in this condition (Koss, 1952).

Until recently the management of the condition was unsatisfactory. Some cases have undergone spontaneous resolution (Doub and Shea, 1960; Navani, 1966; Moore, 1968) and in others resection has been followed by recurrence (Sames, 1964). Forgacs et al. (1973) predicted that by breathing a high concentration of oxygen the total tension of gases at the venous end of the capillaries would be lowered, thereby increasing the rate of diffusion of gas from the cysts (Nunn, 1969). As the cysts deflate they reach a critical diameter at which they will collapse (La Place relationship).

Since Forgacs's original report several other cases have been successfully treated (Down and Castleden, 1975; Watson, 1976), though there have been some recurrences (Forgacs et al., 1973; Van der Linden, 1974). Administration of oxygen by a face mask or oxygen tent in an otherwise healthy child is difficult, particularly if the child is mentally retarded. When the severity of this patient's symptoms demanded definitive treatment, and conventional methods of oxygen administration failed, it was felt that endotracheal intubation and IPPV with 100% oxygen was justified. In man pulmonary damage due to a high inspired oxygen concentration does not develop in less than 24 hours even with an FIO2 of 1:0 (Leigh, 1975). In this patient the right-to-left shunt made it impossible to achieve a Pao2 of 300 mmHg (400 kPa) as recommended by Forgacs et al. (1973).

It is interesting to note that the cysts deflated with a maximum Pao2 of 237 mmHg (31·6 kPa) and that the duration of treatment was 24 hours as compared to the 6 days used by Forgacs et al. (1973) and by Down and Castleden (1975). This method of oxygen therapy should be considered when conventional methods of oxygen administration are impossible.

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References


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