was felt to impede adequate ventilation and spontaneous limb movement was not noted. It is possible that the combination of gentamicin and phenobarbitone may have potentiated limb mobility and that it was the lack of normal movements and tone which predisposed to joint contractures. We suggest that careful and regular passive movements of all major joints be made during the course of paralysis and that infants treated with aminoglycosides or phenobarbitone, or both, be observed with particular care for the early detection of joint contractures.

We thank Mrs Robinson and the Physiotherapy Department for advice and help.

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Received 27 September 1983.

Intrathoracic extramediastinal cystic hygroma

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SUMMARY A case of intrathoracic, extramediastinal cystic hygroma presenting with pneumonia in a 6 year old boy is described.

Cystic hygroma usually presents in the neck or axilla. Intrathoracic cystic hygromas are rare, usually situated in the mediastinum, and generally present in adult life.

Case report

A 6 year old boy with a cough, fever, and left sided pleuritic chest pain was admitted to hospital and treated with antibiotics and physiotherapy for left sided pneumonia and pleurisy. After rapid improvement he was discharged on day 6. Chest radiographs at follow up two weeks after discharge showed a persistent capacity confluent with the left hemidiaphragm, although the boy was asymptomatic (Figure). No comparison could be made with earlier radiographs as the boy had not undergone x ray examination before pneumonia. Apart from some dullness to percussion and reduced breath sounds at the left base, there were no abnormal findings on examination.

Barium meal and follow through excluded a diaphragmatic hernia. Bronchoscopy showed some superior displacement and slight compression of the

left main bronchus. At thoracotomy a lobulated cystic tumour measuring 9 cm × 4 cm was found lying between the diaphragm and the undersurface of the left lower lobe. Roughly triangular in shape with its apex arising anteriorly between the diaphragm and the pericardium, it passed posteriorly over the diaphragm and was attached to the chest wall by an adhesion resembling a cord. There were inflammatory adhesions to the lung and diaphragm, both of which were otherwise normal.

The mass comprised one large and many smaller cysts containing either discoloured blood or gelatinous material. The cyst walls consisted of thick fibrous tissue with a few nondescript flattened lining cells. They contained many focal aggregates of lymphocytes and plasma cells and showed prominent foreign body giant cells around collections of cholesterol crystals. There was a thin layer of smooth muscle in the walls of some of the cysts. The lesion was identified as a cystic hygroma.

Discussion

Intrathoracic cystic hygromas outside the mediastinum are particularly rare, and diagnosis requires the exclusion of a number of other conditions. Hydatid cysts, teratomatous dermoid cysts, and embryonal cysts of respiratory and alimentary origin all have a distinctive lining and are easily differentiated. A more difficult differential diagnosis in the mediasti-

Figure Radiographs in an asymptomatic, 6 year old boy showing persistent opacity confluent with left hemidiaphragm.

nododiaphragmatic angle is that of a pericardial coelomic cyst. This is usually a simple unilocular structure with a thin fibrous wall lined by mesothelial cells, which are often so flattened that they are hard to identify. Cystic hygromas are lined by endothelial cells, but these too may be so attenuated that they are barely recognisable. In contrast to a coelomic cyst however, hygromas are generally complex multilocular structures with smooth muscle remnants of a vascular media and lymphoid aggregates in their wall. The lesion in our patient showed all these features and was therefore regarded as a cystic hygroma.

Cystic hygroma is thought to represent a hamartomatous malformation of the lymphatic system. Clinical presentation depends largely on anatomical site. The usual presentation in children is as a painless, fluctuant, transilluminable swelling in the posterior triangle of the neck on the first day of life, although some hygromas have now been detected by ultrasound before birth.

Occasional mediastinal hygromas in small children may present with large airway obstruction, or even pulmonary hypoplasia, but in older children and adults this is unusual and they are usually detected as a mass seen on routine radiographic examination. Other presenting features include chest discomfort, chylothorax, and partial obstruction of the superior vena cava. Inguinal hygromas may be confused with inguinal hernias, and retroperitoneal or mesenteric lesions may cause severe abdominal pain or simply a painless increase in

girth. They should be treated with surgical excision. Aspiration and radiotherapy have been used where total excision was not possible owing to size or site.

The incidence in different sites varies a little from one report to another, but cystic hygroma develops most commonly in the neck (78–90%) and axilla (20%).^{1,2} Occasionally it may develop in the thorax, retroperitoneum, inguinal region, or scrotum. Intrathoracic lesions form less than 10% of the total number and are generally cervicomedial. Less than 1% are totally within the thorax and most of these develop in adults (75%) with less than 8% in children under 1 year old.^{1,2} This is in appreciable contrast to the cervical cystic hygroma that generally presents at birth (50–60% of cases) or by 2 years of age (90%).¹

Intrathoracic cystic hygromas are particularly rare outside the mediastinum. Skinner and Hobbs described such a lesion in 1936,³ but it has subsequently been decided that what they described was a pericardial coelomic cyst.⁴ The only definite cases are those described by Santy *et al* at the base of the left hemithorax in a 47 year old man⁵ and by Yacoub and Lise posteriorly in the lower chest in a 65 year old man.⁶ Lindskog, in a discussion at the end of Drash and Hyer's paper, refers to two previously reported cases in which smooth muscle fibres in the cyst walls suggested a lymphatic origin, but he does not mention the ages of the patients. Ours would therefore seem to be the first description of a cystic hygroma within the thorax but outside the mediastinum in a child.

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Received 2 September 1983

Precocious puberty: a follow up study

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SUMMARY Follow up of women previously seen between 1951 and 1971 with precocious puberty showed that most had normal menstrual cycles and that fertility was generally normal in those with 'constitutional' precocious puberty. Three patients, however, with the McCune-Albright syndrome had not yet proved to be fertile.

There is very little information on subsequent menstrual and fertility patterns in patients presenting with precocious puberty. We undertook the present study to provide further information on this subject.

Subjects and methods

We reviewed the medical records of all female patients with precocious puberty who were seen at the Hospital for Sick Children, Great Ormond Street, between 1 January 1951 and 1 January 1972. For the purposes of this study precocious puberty was defined as the progressive development of secondary sexual features and accelerated growth before the age of 8 years or menstruation before the age of 10. Seventy one patients who fulfilled these criteria were approached through their family doctors and requested to answer a short questionnaire concerning their height, menstrual pattern, and, if applicable, obstetric history.

Sixty four patients responded to our enquiry. Six patients did not reply to our letters and one patient had died of a brain tumour. Twelve patients had presented with precocious menarche alone—that is, regular menstruation in the absence of secondary sexual development—and as these cases have been

described in detail elsewhere they are not referred to in this paper.¹

Forty two patients aged between 15 and 37 years (mean age 23.6) at the time of this study had been considered to have constitutional precocious puberty when they initially presented with signs of puberty starting between the ages of 1 and 8 years (mean age 5.0). As most patients were seen before the introduction of computed tomography it is quite possible that some who had a small intracranial lesion, for example a small hypothalamic hamartoma, were included in this idiopathic group. Three patients had been treated with medroxyprogesterone, given as a depot injection in two cases.

Ten patients aged between 15 and 34 years at the time of this study had some pathological condition that had caused the accelerated pubertal changes (Table).

Results

Constitutional precocious puberty.

Menstrual pattern. Of the 42 patients with constitutional precocious puberty, 40 reported regular menstruation at roughly monthly intervals. The two remaining patients reported having infrequent and irregular periods.

Table Aetiology of precocious puberty in 52 subjects

Cause	No of patients
Idiopathic	42
Lesion of central nervous system	6
The McCune-Albright syndrome	3
Granulosa cell tumour of ovary	1