"An act of William III made fireworks illegal; the penalty for manufacture or selling was five pounds, and for letting them off, whether on one's own premises or in a public place, was 20 shillings or one month's hard labour": but this legislation failed to control the industry. There is already a restriction on those allowed to buy fireworks—they must not be under 13 years of age; but in practice this regulation is ineffective because it is impossible to enforce in the face of connivance by parents and older boys. The effectiveness of controlling fireworks at the source—that is, at the factory—is clearly shown by the immediate and lasting reduction in blast injuries of the hands since production of the thunderflash was stopped.

The suggestion of legal enforcement does not imply callousness on the part of the trade: the contrary, in fact, is true. From time to time leading manufacturers have agreed among themselves to ban popular types of fireworks which have proved hazardous. For instance, air-bomb types which were produced for some years between the wars have been discontinued; rockets on sale to the general public are limited to the five-shilling size to prevent danger from fall-out of unduly large sticks; and twelve months ago it was decided to discontinue production of aerial torpedo types which fly about in an unpredictable manner (Bennett, 1961).

Substitution

Opposition to change in the field of preventing accidents comes commonly from vested interests, but in this case there is no need to change general custom or spoil sport or restrict production.

Fireworks have been displayed at spas and pleasure gardens under private enterprise since the middle of the eighteenth century: those of C. T. Brock at the Crystal Palace in London from 1865 to 1935 were perhaps the best known. Only a few years ago I attended a crowded privately organized display in a meadow at Tewkesbury, with the Abbey as the scenic backcloth—a perfect, natural setting. This seemed quite to steal the thunder from anything a few boys could put up. It was attended by the public, and, with money raised by the small individual admission fee, sufficient high-quality fireworks were displayed to make a most successful evening—and one which was both safe and, I feel sure, without loss to the manufacturers. With encouragement, local displays of this type could replace the precocious handling of explosives by children. How to develop this substitution profitably is surely a challenge to the trade as a whole.

Conclusion

On the evidence of recent data about firework injuries treated at the Birmingham Accident Hospital, a serious proposition is put forward to reduce the disgraceful and unnecessary suffering of the Guy Fawkes celebration year after year.

This proposition combines legal restriction with an opportunity for the industry to take the initiative in reorganizing the distribution of their products.

Legislation is called for to prohibit the retail distribution of fireworks except for organized displays—nothing less will prevent these injuries.

As a substitute, safe organized displays might well be encouraged for public enjoyment and the welfare of the industry.

I am grateful to my colleague, Mr. J. S. Cason, and to Mr. P. Jameson Evans of the Midland Eye Hospital, for allowing me to use their cases, and to Mr. William Gissane and Dr. John Bull for their interest in this problem which has been a stimulus through the years.

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PULMONARY AGENESIS

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The congenital absence of a lung or part of a lung is always a surprising finding and is something of a rarity. Burger (1947) collected 54 cases and Wexels (1951) recorded 87. Oyamada *et al.* (1953), in an exhaustive review of the literature, were able to find 73 cases of absent lung and 38 cases of hypoplasia. In 1955 Hochberg and Naclerio reviewed 100 cases and stated that they could find "less than 150 cases reported in the medical journals." Single cases have been reported from time to time in the American literature, notably by Warner *et al.* (1955) and Steiner (1956).

A further case came to necropsy here recently and was accompanied by a number of other defects. The fact that three other examples were treated in one general hospital in the past four years suggests that the condition may not in fact be so rare as has been thought.

Case 1

This patient, the first child of healthy parents, was born normally at home. Regular respirations were established 10 minutes after birth, but breathing was wheezy and a good deal of mucus was produced. A cyanotic attack occurred seven hours later and was terminated by aspiration of the pharynx. He was brought to hospital when 11 hours old. On admission the temperature was 90° F. (32.2° C.) and respiration rate 68 a minute. His colour was normal except for the extremities, which were cold and cyanosed. The right thumb was small and was attached to the hand by a tag of skin. It was noticed that the anus was imperforate and no dimple was seen. An unusually large median raphe extended from the anal region forward along the midline of the scrotum and penis.

Examination of the chest revealed that the left hemithorax was a little fuller than the right. The trachea was deviated to the right and the heart apex beat was behind the sternum. The heart sounds were most clearly heard on the right side of the chest. The percussion note was impaired and the breath sounds were diminished on the right side, while rales were heard, chiefly on the left side. The rest of the physical examination was normal and a catheter was passed into the stomach without difficulty.

X-ray examination of the chest showed the heart to be in the right hemithorax and there was no evidence of a right lung (Fig. 1). X-ray examination of the abdomen with the child inverted demonstrated the presence of air within a short distance of the perineum. Within a few hours a blackening appeared at the perineal end of the raphe. This spread steadily forwards across the scrotum and to the end of the prepuce. It was presumed to be meconium.

Thirty-six hours after admission operation was performed. The rectum was demonstrated $\frac{1}{2}$ in. (1.3 cm.) below the perineal skin. The rectum was identified and the rectal mucosa was sewn to the perianal skin.

On the third day small amounts of meconium were passed and urine was passed once, but the abdomen soon became distended and coils of intestine were visible. Partial relief followed passage of a rectal tube. From this time onwards great difficulty was experienced in evacuating the bowels.

FIG. 1.—Case 1. Radiograph of chest in life. The heart is in the right hemithorax.

Frequent use of the rectal tube with small saline washouts and the instillation of detergent kept the distension within bounds until shortly before death.

On the fifth day the bladder was palpably distended and no further urine had been passed. A urethral catheter could not be passed even under an anaesthetic, there being an apparent obstruction at the internal sphincter. Suprapubic catheterization was therefore performed, and was maintained until the 23rd day, when urine was passed normally through the urethra.

On the 12th day the left kidney was thought to be palpably enlarged and the urine was found to be infected with *Escherichia coli*. This infection was treated with nitrofurantoin ("furadantin"), to which the organism was sensitive. The infection was never completely controlled despite changes in treatment. The blood urea rose from 86 mg./100 ml. on the 12th day to 380 mg./100 ml. shortly before death.

FIG. 2.—Case 1. Post-mortem photograph showing single bronchus. There is no trace of the right bronchus or right lung.

gested intestinal obstruction. X-ray examination of the abdomen at this time showed gross distension of the colon with faecal masses. Colostomy was considered, but prior to operation a micturating cystogram was taken on the 52nd day. This revealed a large left-sided hydronephrosis and

megaureter without evidence of any kidney or ureter on the right side. On the 53rd day the infant suddenly collapsed and died.

Post-mortem Findings.—The body was that of an undersized wizened infant. The right thumb was attached by a thin fold of skin. On opening the thoracic cavity the heart was seen to be in the right side of the chest. Further investigation revealed that the right lung was completely absent and the apparent dextrocardia was due to mediastinal shift. The heart itself was normal. Further dissection revealed that the trachea led directly into the left main bronchus (Fig. 2). There was no attempt at formation of a right bronchus. The right kidney was absent and the left greatly hypertrophied. The whole of the pelvic colon was ballooned. Histological evidence of Hirschsprung's disease was not found.

Case 2

This patient was the second child of healthy unrelated parents, the other child, 2½ years older, being normal. Pregnancy was uneventful and he was delivered normally at term; birth weight 7½ lb. (3,400 g.). He was a little slow to breathe and was noticed to have a short, thick neck. He soon settled and thrived on the breast without respiratory symptoms until he was nearly 3 months old, when he was sent to hospital. This was necessitated by the occurrence of cyanosis and grunting respirations 10 minutes after the evening feed.

Examination revealed a greyish cyanosed infant, weighing 10 lb. (4,535 g.). His temperature was 99° F. (37.2° C.), pulse 156, and respirations 66. The neck was short, with a bony projection in the midline posteriorly and a small dimple at the summit. The right side of the chest was prominent and the left flattened, with very little movement on respiration. The percussion note was dull and breath sounds were diminished on the left side. The heart apex beat was not felt, but there was a systolic murmur at the base.

X-ray examination of the chest showed displacement of the heart and mediastinum to the left, with the heart in the left hemithorax. The right lung was normal. X-ray examination of the spine revealed multiple vertebral abnormalities in the cervico-dorsal region. Bronchoscopy showed a trachea of normal dimensions with inflamed mucosa at the lower end. Only one main bronchus (the right) was seen. A bronchogram revealed a normal right bronchial tree, but there was no filling on the left.

The child was nursed in oxygen and given penicillin and streptomycin. The cyanosis and respiratory distress slowly improved and oxygen was discontinued after a week. At this time the pulmonary signs were unchanged, but the murmur had become continuous and was best heard posteriorly over the upper left chest.

Since discharge from hospital he has become progressively more cyanosed and dyspnoeic on exertion. Clubbing of the fingers developed. At the age of 2 years the continuous murmur was still present and the second heart sound was loud and single. Venous angiography at this age showed the presence of a single ventricle and common truncus.

Case 3

This patient, the ninth child of healthy unrelated parents, was born normally at term after an uneventful pregnancy and labour. Weight at birth was 7 lb. 2 oz. (3,230 g.). All his siblings are healthy. He was breast-fed from birth until he was first seen as an emergency admission to hospitals at the age of 3 months. He had always had some cough and wheezing, but on the day of admission, shortly after a feed, he became very dyspnoeic and cyanosed. He then experienced generalized convulsive movements and was brought to hospital.

By the time he had reached the ward the convulsion had ceased and he was no longer cyanosed. His temperature

was 99° F. (37.2° C.), pulse 166, and respirations 48. The head—circumference 16½ in. (41 cm.)—seemed large in proportion to the trunk, the chest circumference being 14½ in. (36.7 cm.). His weight was 9 lb. 14 oz. (4.480 g.). Accessory auricles were present on both sides. The neck was short. An inspiratory wheeze was audible and the chest was asymmetrical, with prominence of the left side and flattening of the right. The trachea was displaced to the right and there was dullness and reduced air entry on this side. No area of cardiac dullness was noted on the left side, but the heart sounds were normal. X-ray examination showed the heart to be displaced into the right side of the chest. The right hemithorax was smaller than the left and no lung markings were seen on that side except for an apparent herniation of the left lung across the midline.

Bronchoscopy revealed a small slit-like trachea, and the left main bronchus was the only main bronchus visible. Bronchography showed the presence of opaque material in the left main bronchus, but no evidence of a right main bronchus was seen. The branches of the left main bronchus were poorly defined.

From this time onwards he was rarely out of hospital and was treated repeatedly for respiratory infections. He gained weight slowly, and at the age of 11 months weighed 15½ lb. (6.915 g.). His mental development approximated that of a 3-months-old baby. At this age a further bronchogram was made with the object of delineating more clearly the subdivisions of the left main bronchus. He died suddenly just after the procedure was finished.

Post-mortem Examination.—Both the right lung and the upper lobe of the left lung were absent. There was no trace of the right main bronchus. In addition there were horseshoe kidneys, vertebral defects in the cervical and thoracic spine, and accessory auricles.

Case 4

This patient, the only child of healthy unrelated parents, was born normally after an uneventful pregnancy, with a birth weight of 7¼ lb. (3.290 g.). She was breast-fed for five months, and weaned without difficulty. Since the age of 4 months she had suffered from attacks of cough and wheezing, and a rattle in the throat caused some concern to the parents. She was admitted to hospital at the age of 9 months. Her weight was 15 lb. 14 oz. (7.200 g.). Her temperature was 99° F. (37.2° C.), pulse 130, and respirations 50. Abnormal findings were confined to the chest, and consisted of slight diminution of the size of the right hemithorax with partial absence of the right pectoralis major muscle. The heart and trachea were deviated to the right. The percussion note was impaired on the right side and breath sounds were feeble on that side.

X-ray examination of the chest suggested collapse of the right lung with displacement of the mediastinum to the right and herniation of part of the left lung across the mediastinum. The tuberculin test (1:1,000) was negative. Bronchoscopy revealed a very small right bronchial tree and no upper- or middle-lobe orifices were seen. A bronchogram confirmed the absence of the right upper- and middle-lobe bronchi. The remaining bronchial components filled normally.

Since that time she has developed normally and the signs have remained unchanged except that a grade 2 systolic murmur has appeared. The cough and wheezing have diminished and are only noticeable with winter colds. At the age of 4½ she was 44 in. (112 cm.) tall and weighed 38 lb. (17.2 kg.), and her intelligence seemed normal. She has undergone recent Mantoux conversion without any changes in the chest x-ray picture.

Discussion

The condition described is one of unknown aetiology. Whatever the precise cause may be, presumably there

must have been interference early in the development of the embryo. Even at the 3-mm. stage lung buds can be detected as outgrowths from the trachea (Baxter, 1953), and it may be of significance with reference to Case 1 that the mesonephros also begins to form at this time. The nature of the interference remains a mystery. Familial incidence is noticeably absent, while in none of the cases described was there any history of virus infection, x-ray exposure, or trauma.

Although lobar agenesis is comparatively uncommon and in most cases the lung is completely absent, the amount of surviving lung tissue varies. Schneider (1912) drew attention to this variation and suggested a classification. He divided the cases into three groups: (1) those with no trace of bronchus, vascular supply, or lung on the affected side; (2) those in which there was a demonstrable bronchial remnant; and (3) those in which the bronchus was slightly more developed, lay within the mediastinum, and ended in a fleshy structure without lobes.

When such a large amount of pulmonary tissue is absent it seems somewhat surprising at first sight that difficulties in diagnosis occur. In infants, however, although there is often fullness of the opposite hemithorax, the mediastinal displacement may pass unnoticed, since even in health the apex beat is often impalpable. A cardiac bruit is not uncommon and may be due either to increased blood-flow through a single pulmonary artery or to concomitant congenital heart disease. The latter is a frequent accompaniment, and was found in 23 of the 73 cases collected by Oyamada *et al.* (1953). Other congenital abnormalities, too, commonly coexist, and it is often these rather than any pulmonary maldevelopment that lead to the death of the patient (Case 1). Indeed, death from respiratory insufficiency is by no means invariable. Some patients have reached an advanced age without difficulty, and it seems likely that respiratory distress is precipitated by intercurrent infection.

In the differential diagnosis in infancy, diaphragmatic hernia, pneumothorax (especially associated with staphylococcal pneumonia), lobar emphysema, and dextrocardia need consideration. The clinical characteristics of these conditions are well known and the increasingly grave symptoms caused by the first three will usually lead to their detection.

Summary

Four further cases of pulmonary agenesis are described and the literature is briefly reviewed. The coexistence of other congenital abnormalities is mentioned and their role in causing death emphasized.

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