ATELECTATIC BRONCHIECTASIS IN CHILDHOOD

BY

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The occasional occurrence in radiograms of the chest of triangular shadows, situated behind the heart-shadow on the left or across the cardio-phrenic angle on the right, has attracted the attention of radiologists for many years. It is, however, only since the introduction of lipiodol for diagnostic purposes that the frequent association of these shadows with bronchiectasis has been recognized. Many of the earlier writers on the subject regarded the triangular shadow as being diagnostic of a mediastinal effusion, as described by Dieulafoy¹, but almost consistently negative results on needling and the absence of fluid when open operation was adopted (Sergent and Bordet²) led to the realization that effusion could only rarely be held responsible. Rist, Jacob, Trocmé, and Soulas^{3, 4} recognized the presence of bronchiectasis in a number of classical cases which they described, and considered that the shadow was due to the formation of a secondary 'adhesive mediastinal pleurisy' over the bronchiectatic area, associated with pneumonic infiltration within it. A similar interpretation has been accepted by the majority of French clinicians who have described cases. In England and America the triangular shadow is generally regarded as being due to collapse of the lower lobe, or of an accessory lobe, of the lung, though this view does not of course exclude the possibility of pneumonic consolidation or fibrosis within it, nor of thickening of the pleura with which it is covered.

The term 'atelectatic bronchiectasis' has therefore been applied to the cases in which there is dilatation of the bronchi within the collapsed area; the evidence for this interpretation will be discussed subsequently. Cases have been reported in this country by Sparks⁵, Morlock and Pinchin⁶, Ellis⁷, and Rodgers⁸, and the condition has been considered from the clinical and radiological points of view by Moll⁹ and Kerley¹⁰ respectively. There is little doubt, however, that the condition is more frequent than the extent of the literature might imply.

Present investigations.

It is with the object of illustrating certain clinical features of the condition as it occurs in childhood that the following eight cases are reported. Five show the classical picture, while three others are included for the light they throw on the ætiology and pathogenesis. All have been under observation in the Children's Department of the London Hospital during the past two years.

LIPIODOL INVESTIGATION.—All the cases except one were investigated by the injection of lipiodol. This was carried out by the crico-thyroid route through a small needle, using 10 c.cm. lipiodol. This amount is rather more than is generally recommended for children, but no ill effects are likely to follow if the injection is carried out sufficiently slowly. The children were given $1\frac{1}{2}$ grn. of nembutal by mouth and $\frac{1}{2}$ grn. of morphia subcutaneously three-quarters of an hour before the injection. In some cases the nembutal produced a satisfactory state of drowsiness in which it was possible to carry out the injection under local anæsthesia of the neck and cocaineization of the trachea. In most instances, however, it was found more satisfactory to give a general anæsthetic, for which nembutal served as an excellent, though somewhat uncertain, preparation, in order to abolish the cough reflex, the child being kept lightly anæsthetized until it was seen from the first film whether adequate filling of the collapsed area had occurred. The injection was carried out with the child lying supine with the shoulders raised on a single pillow and the neck extended. As soon as the injection was completed, the shoulders were raised on a second pillow and the child turned on to the affected side. In four cases the filling of the affected area was rapid and complete; while in three the lipiodol entered the collapsed lobe less readily than the surrounding lung.

True atelectatic bronchiectasis : Cases 1-5.

Cases 1 and 2.—Bronchiectasis limited to a collapsed area behind the heart-shadow.

Case 1.—(Fig. 1) No. 40867.—B. H., a girl, aged $8\frac{1}{2}$ years. Infancy and development were normal until 1927 when at the age of 3 years she had bronchopneumonia and is said to have been ill for nearly 5 months. She attended the out-patient department in 1929 (aged $4\frac{3}{4}$) on account of persistent cough. Her weight was then $32\frac{1}{2}$ lb. Crepitations were heard over both lower lobes posteriorly. The Pirquet test was negative. Since this time the chest signs have persisted, and there has been intermittent cough, more frequent in the winter. Sputum has only occasionally been present with exacerbations of the cough, and has never been foetid. In January, 1930, when she was 5 years old, slight clubbing of the fingers was first noted. The general health has been moderately good except for occasional attacks of 'bronchitis' during the winter, although she has been consistently somewhat under-weight for her age. In June, 1932, she was admitted to hospital for investigation.

On examination: A moderately well-developed child with good colour, weighing 44 lb.; no cough whilst in hospital. Postural drainage failed to produce sputum. The fingers showed a slight degree of clubbing. The chest was not deformed and moved evenly. The apex beat was in the 5th intercostal space in the mid-clavicular line. There was no impairment of the percussion note, but slight diminution of air-entry at the left base. Coarse râles were heard at both bases posteriorly. No other physical signs of disease were present except caries of 3 molars.

Radiological examination: An area of increased density behind the heart-shadow, extending from the hilum of the left lung to the diaphragm, is seen in repeated films taken since the patient's first attendance in 1929.

Lipiodol injection (June, 1932) showed bunching together and cylindrical dilatation of the bronchi within the collapsed area, and spreading-out of the bronchi and emphysema of the upper lobe (Fig. 1).

Since this time, the physical signs have not altered appreciably; she now weighs $47\frac{1}{2}$ lb. (normal $52\frac{1}{2}$ lb.), and with the exception of infrequent cough has at present no symptoms.

Case 2.—(Fig. 2 and 3) No. 40489.—R. B., a girl, aged $9\frac{1}{2}$ years. The child was well until the age of $3\frac{1}{2}$ years (1926), when she had pneumonia, followed by persistent cough, which was most troublesome at night. She was brought to hospital in 1928, at the age of $4\frac{1}{2}$, on account of this symptom. At this time the percussion note was normally resonant over both lungs, and air-entry equal, but dry rhonchi were heard at both bases posteriorly. The heart was not displaced; there was no clubbing. Cough produced a moderate amount of yellow sputum. The tonsils, which were

FIG. 1.—Case 1. B. H. Dilatation and bunching together of bronchi within collapsed left lower lobe, and spreading-out of bronchi of left upper lobe.

large and unhealthy, were removed when she was 5. At this time she weighed 33 lb. The cough becoming much less troublesome, she failed to re-attend the out-patient department until September, 1931, when, at the age of 8, she weighed 37 lb. Severe but unproductive cough had recurred for the past 6 weeks. A radiogram taken at this time shows evidence of a collapsed area behind the heart. The Mantoux test was negative (1 in 1,000 dilution). The child was sent to a convalescent home for 2 months. On her return, examination of the chest showed many crepitations at both bases posteriorly, with an area of tubular breathing below the angle of the

left scapula. Early clubbing of fingers was noted. She was admitted to hospital for lipiodol examination in March, 1932.

On examination: A thin but otherwise healthy-looking child, with marked foetor of the breath. Weight $37\frac{1}{2}$ lb. (normal 53 lb.). A slight degree of clubbing of the fingers was present. There was considerable muco-purulent post-nasal discharge and caries of several teeth. The chest moved evenly; there was no displacement of the heart. The percussion note was impaired at the left base, and there was diminished air-entry in this area. Coarse crepitations were heard at the right base, and an area of tubular breathing at the angle of the left scapular.

Lipiodol injection showed typical bronchiectasis and bunching together of the bronchi within the collapsed area behind the heart shadow (Fig. 3).

FIG. 2.—Case 2. R. B. Collapsed left lower lobe behind heart shadow.

Since her discharge from hospital, the child has been admitted to an open-air school and her general health has been good, though the gain in weight has been poor. Cough has not been severe or productive. The physical signs have remained unchanged, except that the breath sounds are now definitely amphoric rather than tubular in the small area at the angle of the left scapula. She now weighs 43 lb. at the age of $9\frac{1}{2}$ (normal $57\frac{1}{2}$ lb.).

Cases 3 and 4.—Left-sided atelectatic bronchiectasis with dilatation of adjacent bronchi.

Case 3.—(Fig. 4) No. 41876.—M. M., a girl, aged 9 years. The mother died 3 years ago from rheumatic carditis; the father attends hospital with chronic bronchitis; one sister is alive and well. The patient was well except for repeated attacks of bronchitis until September, 1928 (age 5), when she suffered from broncho-pneumonia following measles, and was in Bethnal Green Hospital for 6 weeks. Since this time she has had persistent cough, and was brought to the London Hospital in February, 1929, on account of this symptom. At this time the chest showed flattening at the left base, and impaired percussion note with absent breath sounds over the left lower lobe. The Pirquet test was negative. Radiological examination (Feb. 8, 1929) showed a triangular shadow behind the heart shadow and increased lung markings at the right base. She was admitted to hospital in March, 1929, when she had frequent cough and persistent signs at the left base. The apex beat was in

FIG. 3.—Case 2. R. B. Dilatation of bronchi within collapsed left lower lobe.

the 4th space $\frac{1}{2}$ in. external to the mid-clavicular line. There was no clubbing. She was discharged to long convalescence, but in August, 1929, the radiogram of the chest was unchanged. She was admitted to a residential school until November, 1932, and during this period had persistent cough with a little non-purulent sputum. She suffered from broncho-pneumonia for the second time in December, 1930, but otherwise her general health has been moderately good. She was admitted to hospital for lipiodol examination in December, 1932.

On examination: A moderately well-developed child weighing 52 lb. (normal 55 lb.). Her colour is good; there is no clubbing. There is marked pigeon-breast deformity of the chest, with flattening of the lower ribs anteriorly on both sides, and slight flattening and diminished movement of the left side of the chest posteriorly. The apex-beat is visible in the 4th and 5th spaces $\frac{1}{4}$ -in. outside the mid-clavicular line. The percussion note is slightly impaired in the left mid-axillary

line to the level of the 4th rib above, and there are scattered rhonchi heard over the left lower lobe posteriorly. Air entry is equal on the two sides. There was frequent unproductive cough while the child was in hospital.

Radiological examination: Generalized increase of lung markings with fibrosis of right zone 3 and collapse of the left lower lobe.

Lipiodol injection showed bunching together and dilatation of the bronchi within the collapsed area and an appearance suggestive of varicosity of the two adjacent bronchi. The remainder of the bronchial tree appeared normal.

Case 4.—(Fig. 5) No. 30748.—C. F., a boy, aged 13 years 3 months: an only child; both parents well. Infancy was normal. Pneumonia followed measles when he was

FIG. 4.—Case 3. M. M. Dilatation and bunching together of bronchi within collapsed left lower lobe and 'varicose' appearance of two adjacent bronchi.

4½ years old, since when he has had cough which occurs throughout the day and is usually unproductive. He has had a variable amount of sputum, which has never been noticeably offensive, and recently has been almost entirely absent. His general health has been good. He was first seen at the London Hospital in April, 1927, at the age of 7½, when slight clubbing of the fingers was noted and examination of the chest showed impaired percussion note with crepitations and occasional rhonchi at the left base; air-entry appeared normal. The apex-beat was 1 in. to the left of the mid-calvicular line in the 5th intercostal space; the heart sounds were normal,

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Radiological examination showed an increase of lung-markings at the right base, the left base being obscured by the heart shadow. Repeated X-rays since this time have shown the presence of a sharply-demarcated area of increased density behind the heart shadow. As in several of the long-standing cases, the opacity is not strictly-speaking triangular in shape, the hypotenuse of the triangle running almost vertically downward from the region of the hilum to the diaphragm.

Lipiodol injection (April 4, 1932) was unsuccessful owing to vomiting during the procedure, but a second injection carried out (June 6) showed bronchiectasis within the collapsed area, and marked dilatation of the two adjacent bronchi. The remainder of the bronchial tree appeared normal.

Orthodiagraphic examination (Dr. William Evans) showed the heart displaced to the left, but revealed no obvious abnormality of the heart chambers.

FIG. 5.—Case 4. C. F. Displacement of heart, bronchiectasis within collapsed area behind heart shadow, and dilatation of two adjacent bronchi.

Considerable constitutional disturbance followed the first injection of lipiodol, with dullness of the percussion note and diminished air-entry at the right apex, lasting 3 or 4 days. Sputum examination was negative for tubercle bacilli. In May, 1932, 2 weeks after the injection, the boy developed acute hæmorrhagic nephritis, but made an uneventful recovery. Since this time he has been in excellent general health, and has been all but free from both cough and sputum for 3 months.

On examination (Dec., 1932): A well-developed and nourished boy, weighing 91½ lb. (normal 85 lb.). Height 4 ft. 10½ in. The apex-beat is in the 6th intercostal space on the left, in the anterior axillary line. There is a soft localized systolic murmur in the mitral area. The thyroid is slightly enlarged. The chest moves evenly; there is no appreciable scoliosis, but slight flattening of the left lower ribs.

The percussion note is impaired at the left base posteriorly, and rhonchi are heard in this area on deep inspiration. There is a small area of bronchial breathing below the angle of the left scapula. The fingers show a moderate degree of clubbing.

The report and illustration of this case are included by courtesy of the Honorary Editors of the Proceedings of the Royal Society of Medicine.

Case 5.—Bronchiectasis at both bases with bilateral atelectasis.

Case 5.—(Fig. 6 and 7) No. 30174.—L. A., a boy, aged 9½ years. Parents and 4 siblings are alive and well; 1 child was still-born. Infancy was uneventful until the age of 8 months, when the patient had whooping-cough followed by bronchopneumonia. Since this time he has had persistent cough of variable severity. There

FIG. 6.—Case 5. L. A. Bilateral atelectasis. On the left the collapsed area is almost entirely obscured by the heart shadow.

has been little or no sputum except during occasional exacerbations of the cough in winter. He has played games normally, and the general health has been good. He was first seen at the London Hospital in 1929 at the age of 5½ years on account of the cough. He was then of normal weight for age, and appeared well. There was clubbing of the fingers, and crepitations and rhonchi were heard at both bases posteriorly. No other abnormal physical signs were detected.

Radiological examination of the chest showed heavy hilar shadows with no definite evidence of fibrosis, but a well-defined triangular shadow across the right cardiophrenic angle. A subsequent examination in the left semi-oblique position showed in addition an area of collapse behind the heart shadow on the left.

He was sent away for prolonged convalescence in 1930. Since this time the boy gained weight steadily, and apart from cough and frequent night-sweats has suffered but little disability. The physical signs and X-ray appearances have remained almost unchanged since the first observation, though râles are now more marked throughout the right lower lobe than on the left. He now weighs 64 lb. (normal 64 lb.). Sputum examination was negative for tubercle bacilli.

Lipiodol examination in October, 1932, showed cylindrical bronchiectasis at both bases. On the right side this appears to be principally limited to the collapsed area, with some dilatation of the adjacent bronchi, but on the left the bronchiectasis is more extensive. The area of collapse in this case does not appear to involve the whole of the left lower lobe.

FIG. 7.—Case 5. L. A. Bilateral bronchiectasis. On the right this is mainly limited to the collapsed lower lobe; on the left it extends beyond the collapsed area, which does not appear to involve the whole lower lobe.

Discussion of Cases 1-5.—All these cases show the radiological picture of atelectatic bronchiectasis, a triangular opacity across the cardiophrenic angle on the right, or lying behind the heart shadow on the left side, within which bronchial dilatation is demonstrable by lipiodol injection. In the first two cases, bronchiectasis is limited to the collapsed area; in Cases 3 and 4 the two immediately adjacent bronchi also show some degree of dilatation, and in the fifth case there is not only atelectasis at both bases, but bronchiectasis extends considerably beyond the collapsed area on the left

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side. The question whether the triangular opacities represent collapse of the whole of the lower lobe of the lung or of an accessory lobe will be considered subsequently.

Although the series is small, the similarity of the histories and clinical findings in these cases is sufficiently striking to justify certain generalizations.

ONSET AND SYMPTOMS.—In every case the information was volunteered that symptoms dated from an attack of pneumonia (probably broncho-pneumonia) in infancy or early childhood; in two cases this followed measles, and in one pertussis. In only one case was there a previous history of bronchitis. The presenting symptom has in every case been cough, which, while sometimes troublesome, has not been such as to cause any real disability or interference with normal activity. The complete absence of foetid sputum and the infrequency of sputum of any kind has been very striking throughout the time that these children have been under observation. Sputum is, of course, liable to be swallowed in early childhood, but these patients are now at an age when it can be raised easily if present in considerable amount. Postural drainage has also proved unproductive. There is, in fact, nothing to indicate an anærobic infection of the bronchi of the type which produces the characteristic foetid ‘bronchiectatic’ sputum, although one child has shown some foetor of the breath.

Similarly, constitutional disturbances have been very slight; the children have all had moderately good general health, and have gained weight steadily. Only in Case 2 is the patient considerably underweight for age. A second attack of pneumonia occurred in Case 3, but recovery was uneventful. Night sweats were complained of in Case 5, but were unaccompanied by pyrexia, and did not occur while the boy was under observation in hospital. Case 5 showed some constitutional disturbance after lipiodol injection, and subsequently developed acute hæmorrhagic nephritis. It is possible that the procedure lighted up some latent infection in the lung, but it did not result in any permanent increase in pulmonary symptoms.

The chronicity of the condition is as follows:—

		Present age.	Age at which pneumonia occurred.	Duration of symptoms.	Period of hospital observation.
Case 1	...	8 years	3 years	5 years	3½ years
Case 2	...	9½ „	3½ „	6 „	4 „
Case 3	...	9 „	5 „	4 „	4 „
Case 4	...	13½ „	4½ „	9 „	6 „
Case 5	...	9½ „	8 months	9 „	4 „

During the periods of observation, no significant changes have taken place in the severity of symptoms or in the physical signs, and it appears reasonable to suppose that bronchial dilatation is of relatively long standing in every case, although only recently demonstrated by lipiodol injection.

PHYSICAL SIGNS.—As already indicated, the children are moderately well-developed, and with one exception are not much under the average weight

for age. Examination of the chest in these long-standing cases usually shows little or no deformity or diminution of movement on the affected side. The percussion note is normal, or slightly impaired at the base posteriorly. Air-entry appears almost equal on the two sides. In two cases there was tubular (changing in one to amphoric) breathing heard over the affected area posteriorly. Indeed, the only physical sign occurring consistently was the presence of râles or crepitations at the base on the affected side. The heart was normal in position or only slightly displaced in four cases, and markedly displaced towards the affected side in one.

Clubbing of the fingers was present in four of the cases and absent in one; in three of the former it was noted for the first time over three years ago. The clubbing was in each instance relatively slight, a finding in keeping with the almost complete absence of toxæmic symptoms.

It is clear, therefore, that in cases of this type where the collapsed lung tissue occupies a small area in the paravertebral groove, diagnosis of the condition is not likely to be made on the physical signs alone.

These cases may be correctly described as examples of 'silent' or 'dry' bronchiectasis. As long as they remain in this stage, sputum is slight in amount or absent, there is little evidence of toxæmia, and the general health is good. It may be noted that hæmoptysis, a symptom occurring in many of the classical cases of 'dry' bronchiectasis, has not occurred in any one of the present series. The clinical importance of the condition, however, lies in the fact which is exemplified in many of the reports of atelectatic bronchiectasis in older children and adults, that secondary infection of the dilated bronchi is always liable to occur.

It will also be seen that while the bronchiectasis may remain strictly limited to the atelectatic area, there is a tendency for the adjacent bronchi to become affected. This may well be a passive process, due not so much to infection as to the 'spilling-over' of retained bronchial secretions which cannot readily be coughed out of the atelectatic area. These in turn will tend to clog the neighbouring bronchi, which, being situated next to an atelectatic area, themselves cannot be readily evacuated.

Reports of Cases 6-8, with comments.

As already indicated, investigation of chronic cases showing the characteristic triangular shadow in the radiogram almost invariably demonstrates an associated bronchiectasis within it, unless of course the shadow represents not atelectasis but the extremely rare condition of chronic mediastinal effusion. The following case (Case 6) illustrates the fact that where the collapse is an acute condition, and where the lung re-expands before the bronchial secretions have had opportunity to accumulate and stagnate within the atelectatic area, lipiodol injection may show no evidence of bronchial dilatation.

Case 6.—Collapse of the right lower lobe with re-expansion.

Case 6.—(Fig. 8 and 9) No. 31990.—J. F., a boy, aged 10½. Both parents and 5 other children are alive and well. The boy had diphtheria at the age of 3, and attended the children's out-patient department at the age of 8 with 6 months' history of cough with non-purulent sputum. No physical signs of disease were detected in the chest, and the general health was good; there was no clubbing. The patient was free from symptoms in two months, and was quite well until May, 1932 (aged 10). He was admitted to hospital under Professor Ellis on May 24th with 5 days' history of fever, cough, and pain in the left side of the neck. The patient had been kept in bed, and the cough had become more severe. On the day of admission the sputum was blood-stained.

FIG. 8.—Case 6. J. F. Collapse of right lower lobe.

On examination: A well-developed and nourished boy, temperature 101°, respiration 36, pulse 136. There was no clubbing or cyanosis, but definite dyspnoea. The heart was not displaced. The chest showed diminished movement over the right lower lobe; the percussion note was dull over this area, and a pleural rub was heard in the right mid-axillary line at the level of the fourth rib. The breath sounds were tubular in character all over the right lower lobe, with ægophony at the right base posteriorly.

The case was diagnosed as right lower lobe pneumonia. The patient ran an irregular swinging temperature (to 104°) with morning rise and evening fall, finally settling on the 16th day of the illness. The chest was needled on the 10th day, but no fluid obtained. The physical signs in the chest persisted after the temperature had fallen to normal.

Radiological examination on June 21st (33 days after the onset of symptoms) showed a triangular shadow on the right across the cardiophrenic angle, indicating collapse of the right lower lobe.

In order to re-expand the collapsed lung tissue, the patient was treated by breathing exercises, consisting in blowing water from one bottle to another. A second radiological examination on June 30th showed the opacity still present in the right cardiophrenic angle, though less dense and tending to become less obvious on

FIG. 9.—Case 6. J. F. The right lower lobe which has re-expanded fills less well than the upper lobe, but the bronchial tree shows no abnormality.

deep inspiration, suggesting better aeration. The opacity appeared smaller with the patient's back to the film, showing that the affected area was situated posteriorly.

An examination of the sputum for tubercle bacilli was negative.

The patient was discharged to convalescent home with some dullness and diminished air entry at the right base still present. He re-attended the out-patient department on his return 2 months later. He still had occasional cough with a small amount of sputum; there were râles scattered over the right lower lobe, and slight diminution of air entry, but the percussion note was unimpaired.

Radiological examination showed complete disappearance of the triangular shadow in the antero-posterior and lateral positions.

Lipiodol injection was carried out in December, 1932. The bronchi at the right base filled less well than the rest of the lung, but appeared normal. There was no evidence of bronchiectasis.

Two further cases (Cases 7 and 8) are included as illustrating primary causes of pulmonary collapse other than pneumonia in children, and because of their possible relationship to atelectatic bronchiectasis. It has not been

FIG. 10.—Case 7. B. R., May, 1931. Collapse of right lower lobe (triangular shadow in cardiophrenic angle), with raising of the right crus of the diaphragm, following inhalation of peanut.

possible to investigate the former by lipiodol injection, but from the persistence of cough and the X-ray appearance it is not improbable that bronchial dilatation either has occurred or is in process of developing.

Case 7.—Collapse of the right lower lobe, probably due to inhalation of a foreign body.

Case 7.—(Fig. 10 and 11) No. 30682.—B. R., a boy, aged $7\frac{1}{2}$ years. Both parents and 1 brother are alive and well. There has been no tuberculosis in the family. The boy was well until February, 1931, aged 5 years and 8 months, when he had a sudden attack of violent coughing and dyspnoea while eating. He was put to bed, and the

cough and dyspnoea lasted for a week. A second attack of dyspnoea occurred in April, 1931, the spasm being such as to stimulate asthma, and there were 4 more similar attacks in the succeeding 5 weeks. The boy was admitted to hospital on May 7, 1931.

On examination (May, 1931): A well-developed and nourished Jewish boy; the respiration and temperature were normal while he was in hospital. There was no clubbing. The throat was clear. The apex-beat was in the 5th space 1 in. inside the mid-clavicular line. The left lung appeared normal; the right chest showed

FIG. 11.—Case 7. B. R., June, 1932. The right lower lobe has re-expanded, but there is an area of dense fibrosis obscuring the right cardiophrenic angle. Several calcified glands are seen, and an area of infiltration in the left upper lobe.

diminished movement, and there was impairment of the percussion note in the right axilla and all over the right lung posteriorly, most marked over the lower lobe. The Mantoux test was negative (1 in 2,000 dilution). The right chest was needled posteriorly, but no fluid obtained.

Radiological examination showed the heart displaced slightly to the right, and a triangular shadow present in the cardiophrenic angle. The right diaphragm was raised.

The signs in the right chest persisted, and the boy was transferred to the Müller Convalescent Home, Broadstairs, where he has been until the present time under the care of Dr. H. M. Raven, to whom I am indebted for the subsequent history. The boy continued to have frequent cough and some respiratory spasm until October, 1931, when after a severe fit of coughing he produced what was thought to be a peanut. Since this time the cough has become much less frequent and severe (though

still present), and there has been no respiratory spasm. There is no sputum, and the breath is not offensive.

Radiological examination by Dr. C. J. Heaton, at the Margate General Hospital in June, 1932, showed that the dense triangular shadow across the right cardiophrenic angle had disappeared, but that there was heavy fibrosis along the ramifications of the right lower bronchus, and several small calcified glands at the cardiac margin on the right and around both hila. The right crus of the diaphragm was no longer abnormally raised. On the left side there was an appearance 'suggesting a resolving process (? tuberculous) in the upper lobe of the lung' (Fig. 11).

In this case (Case 7) acute collapse of the lung appears to have followed the inhalation of a foreign body, and re-expansion of the atelectatic lobe to have occurred when the foreign body was coughed up. It will be noted

FIG. 12.—Case 8. V. H. Anterior-posterior view, showing tuberculous focus in right middle zone; the collapsed left lower lobe is almost entirely obscured by the heart shadow.

that when the child was first seen, the heart was displaced towards the side of the collapse, and the diaphragm raised on the same side. The fact that the second X-ray, taken a year after the foreign body had been coughed up, shows heavy fibrosis, with an appearance of streaking, across the right cardiophrenic angle, suggests that the lower lobe may not have been completely re-expanded and is the seat of fibrosis and possibly of bronchial dilatation. The picture is, however, complicated by the presence of several calcified glands at each hilum, which are not seen in the film taken in May, 1931,

Although the Mantoux test was negative in a dilution of 1 in 2,000 in 1931, the possibility of the present picture being due to chronic tuberculosis affecting a *locus minoris resistentiae* cannot be excluded.

In the first five cases of typical atelectatic bronchiectasis, pulmonary tuberculosis has as far as possible been excluded by tuberculin tests or sputum examination. The fact that pulmonary tuberculosis may, however, be associated with a closely similar radiological picture is illustrated by Case 8.

FIG. 13.—Case 8. V. H. Collapse of left lower lobe visible in semi-oblique position.

Case 8.—Pulmonary tuberculosis with collapse of the left lower lobe.

Case 8.—(Fig. 12 and 13) No. 41466.—V. H., a girl, aged 6½ years. Both parents are alive and well; 4 other children are alive and well, though one is said to have had pleurisy at the age of 8. There has been no known tuberculous contact. Except for measles during infancy, uncomplicated by pneumonia, there have been no acute illnesses though frequent feverish attacks; cough has been persistent since infancy. Until recently there has been little sputum, which was not purulent. At the age of 5½ years she was brought to hospital with a history of pains in left ankle for 2 years, and swelling of the joint for the past month. On examination, the joint was found to be swollen but not red or tender, with some limitation of extension and wasting of the muscles of the left calf. The chest was normally resonant, with equal air entry, but there were diffuse rhonchi throughout both lungs. Radiological

examination of the left ankle showed swelling and thickening of the synovial membrane, but no rarefaction or erosion of the bone. The Mantoux test was strongly positive in a dilution of 1 in 1,000. The joint was immobilized in plaster for 9 weeks, after which time the child was sent to a convalescent home. The cough and chest signs persisted throughout this time, and in April, 1932, at the age of 6, the sputum became increased in amount and was said to be foul on several occasions after severe attacks of coughing on waking. The child had several attacks of follicular tonsillitis during the spring and early summer of 1932, and her weight remained stationary. She was admitted to hospital in September.

On examination: A pale, moderately well-developed but poorly nourished child, weighing $38\frac{1}{2}$ lb. (normal 41 lb.). The temperature, pulse and respiration were normal while she was in the ward, and cough was frequent. The left chest moved slightly less freely than the right, and the percussion note was impaired at the left base. There was no appreciable inequality of air entry on the two sides. The apex-beat was in the 5th space in the mid-clavicular line; the trachea was not displaced. The tonsils were small and cryptic and the nodes palpable. There was no clubbing of the fingers.

Radiological report: The diaphragmatic movements are not restricted. Lungs: calcified nodes in right middle zone and at left hilum. There is general relative loss of translucency over the right lung with pleural involvement and streaking in zone 1. Collapsed left lower lobe seen behind the heart shadow in left semi-oblique position.

Lipiodol injection showed poor filling of the collapsed area, but sufficient oil entered the left lower lobe to show several areas of dilatation or cavitation.

The patient was unable to raise any sputum while in hospital, but stomach-washings injected into a guinea-pig gave rise to typical tuberculous lesions in which tubercle bacilli were demonstrable.

The child was transferred for sanatorium treatment.

In this case it is significant that there is no previous history of broncho-pneumonia, and the collapse of the left lower lobe has presumably occurred in the course of an active tuberculous infection. The X-ray appearance is suggestive of a focus in the right middle zone, and it is possible that tuberculous sputum has been inhaled from this area and has caused blocking of the left lower lobe bronchus with resultant collapse. The pressure of an enlarged mediastinal gland on the left lower bronchus is another possible explanation. The collapsed area filled poorly with lipiodol, but there is evidence of either bronchial dilatation or cavitation within it. It will also be noticed that the collapsed lobe is almost entirely obscured by the heart shadow in the anterior-posterior view (the small portion visible being indicated with an arrow), but is well seen in the left semi-oblique position. It illustrates the fact that collapse of the left lower lobe may easily be overlooked unless the possibility of its occurrence is specially kept in mind. In order to demonstrate its presence in the anterior-posterior view, an X-ray of increased penetration is frequently necessary.

General discussion.

The conclusion that the triangular shadow already described represents an area of atelectasis, or rather collapse of a complete lobe, is based partly on direct inspection of the area involved post mortem or in the course of open operation, and partly on the configuration of the bronchi. Additional evidence is furnished by such cases as those described by Bezançon et al.¹¹

and Sparks⁵ where the shadow has disappeared, presumably due to re-inflation of the lung, although the bronchiectasis is of relatively long standing. Re-expansion and disappearance of a similar shadow is well seen in a case described by Findlay¹² and in Case 6 of the present series, where the collapse was of recent origin and unassociated with bronchiectasis. In the former, re-expansion was effected by inhalation of CO₂ and in the latter by breathing exercises. The manner in which disappearance occurred is in both cases much more in favour of the shadow representing collapsed lung than fluid effusion.

Open operation has been carried out on a number of the reported cases, and the appearance of the lung somewhat differently interpreted. As already mentioned, Rist et al.³ described a case where operation showed an 'adhesive pleurisy' instead of the mediastinal effusion for which search was being made; Sergent and Bordet² recorded induration of the parenchyma of the posterior border of the lung in a somewhat similar case. Singer and Graham¹³, however, state emphatically that operation having been carried out on a considerably larger number of such cases 'these triangular shadows have been found to represent atelectatic, bronchiectatic lower lobes, with the characteristic cyanotic appearance and indurated feeling on palpation.

. . . The unaffected lobe or lobes had hypertrophied so much as to fill out the chest cavity and surround the collapsed lobe.' Their findings do not suggest that the lobes affected were accessory lobes, though this view has recently been urged by Kerley¹⁰, who regards the collapse as due primarily to lack of mechanical support in a lobe supplied by a para-cardiac branch of the lower lobe bronchus, which contains insufficient cartilage to keep the tubes patent.

In the single post-mortem specimen from a case of this type which I have had the opportunity of seeing, the collapsed area behind the heart involved the whole of the left lower lobe. The left upper lobe had expanded to fill the remainder of the left chest, and it is significant that while the left upper lobe happened in this case to be the site of a long-standing unresolved pneumonia, the collapsed lower lobe did not appear to be involved in the same pneumonic consolidation. There was very considerable peribronchial fibrosis in the atelectatic area, bronchiectasis, and a small amount of pus present in the larger cavities. While there was disorganization of the cartilage in the walls of the dilated bronchi, it appeared probable that this was secondary, due to erosion, rather than a primary absence of cartilage causing collapse. Indeed, the undilated bronchi were normally patent. There was some thickening of the pleura over the lower lobe, but this was considerably less marked than at the apex.

The lipiodol plates in the present series of cases also support the view that the whole lower lobe is affected, as the collapsed area is supplied from the first division of the main bronchus, while the bronchial tree in the upper lobe or lobes is spread out fan-wise from compensatory over-expansion and emphysema. This is well illustrated in Fig. 1 and 3,

The question whether the bronchiectasis preceded the atelectasis or vice versa, is one which it is difficult to answer with any certainty. It has been widely held that collapse will tend to occur around a bronchiectatic area, and that chronic infection of the lung parenchyma and fibrosis will tend to maintain it. Many of the cases give a history of broncho-pneumonia in infancy, from which period the symptoms have dated. A case reported by Bezançon et al.¹¹ might perhaps be quoted in support of this view, as the triangular shadow was seen to appear suddenly during an infective exacerbation of a bronchiectasis dating back to infancy.

On the other hand, it has been pointed out by Kerley¹⁰ that there is considerable evidence against this view. Purulent sputum, wasting, and evidence of toxæmia are often absent until long after the diagnosis of bronchial dilatation is established, and cough is not necessarily persistent or severe. This suggests that the bronchial dilatation is at first a non-infective condition due to retention of bronchial secretions within a collapsed area, and as has been shown in many reported cases the typical clinical picture of bronchiectasis does not develop until secondary infection of the dilated bronchi takes place. It might reasonably be said that none of the five typical cases in the present series show clinical evidence of active infection of the bronchiectatic areas. It is not proposed to discuss the mechanism of collapse of the lung, but it is well established that collapse of a lobe, particularly a lower lobe, may follow the inhalation of a foreign body, as probably occurred in Case 7, or result from a pneumonia. In the absence of definite proof we are inclined to think that the usual sequence of events is as follows: Collapse of one or both lower lobes, and failure to re-expand, following broncho-pneumonia in infancy results in retention of bronchial secretions and bronchial dilatation. The presence of râles in every case makes such a retention of secretions appear probable, although the condition is relatively 'dry' in comparison with the type of bronchiectasis showing abundant sputum. Secondary infection may not occur for many years, and during this period constitutional symptoms will be little in evidence, cough will not be frequent or productive, and what sputum there may be will not be foetid. In rare cases, even when the bronchiectatic condition is of long-standing, re-expansion of the lung may take place with amelioration of symptoms (Bezançon et al.¹¹, Sparks⁵), but in general it may be said that the longer the condition has lasted the less likely is this to occur. This is comprehensible when it is considered that even in the absence of active bronchial infection there will almost inevitably be some degree of fibrosis of the atelectatic lung in the course of time, and it is even possible that the phrenic nerve may become nipped by fibrous tissue with resulting partial paralysis of the diaphragm. This latter eventuality would tend to maintain a condition of collapse.

Secondary infection when it occurs (and this may be relatively early or not until middle life) is likely to result in the rapid appearance of constitutional symptoms and render the prognosis extremely bad. The short

expectation of life which is usually allowed in bronchiectasis in childhood is due in part to the non-recognition of cases of this type. Not only is the characteristic shadow likely to be overlooked when it is obscured by the heart shadow, as is generally the case, many more examples occurring on the left side than on the right, but many of the children will not come under observation, owing to the absence of symptoms, until secondary infection has occurred.

From the point of view of diagnosis it may be said that whenever this X-ray appearance is seen and is of long-standing, bronchiectasis is almost invariably present, although there may be little to indicate it in the way of symptoms or physical signs; confirmation should be obtained by lipiodol injection.

Treatment.—Treatment is essentially preventive. No case of pneumonia with persistent cough or signs in the chest should be discharged before pulmonary collapse has been excluded by X-ray examination. If evidence of this is found, every effort should be made to re-expand the collapsed lobe by CO₂ inhalations or breathing exercises. When the condition is of long-standing, however, it is extremely improbable, though not impossible, that benefit will be obtained from these methods, and attention must be concentrated on preventing secondary infection and attention to the general health. Postural drainage should be tried, but is not likely to be effective. A long convalescence, and if possible a country life or open-air school are probably the most important lines of treatment during childhood. We have seen temporary benefit from repeated bronchoscopy and aspiration in a case where secondary infection had occurred, but it is doubtful if it should be advised in the uncomplicated case. In the same way, when the bronchiectasis is strictly limited to one lower lobe, it would seem logical to perform lobectomy. Unfortunately the operation is one with a high mortality in childhood, and cannot at present be advocated in cases where the symptoms are slight and the general health good. It would seem best to make every effort to prevent secondary infection until the child has reached an age when operation can be better supported.

Antiseptic inhalants are unlikely to be of any value, except in cases where the bronchiectasis has spread to the surrounding bronchi, as they will not enter the collapsed area to any appreciable extent. It is possible that lipiodol injection may be of real benefit where satisfactory filling of the whole area involved takes place, since the heavy oil will tend to displace the stagnant secretion from the dilated bronchi, allowing it to be coughed up. It is said that lipiodol may even remain in the cavities for many weeks; this has not, however, been confirmed in any of the cases of the present series that have been examined four or more weeks after its injection, although sometimes lipiodol has still been visible in the alveoli of the unaffected lobes.

Summary.

1. From a clinical consideration of five cases of atelectatic bronchiectasis in childhood, it is seen that the condition may exist, probably

for long periods, without the classical features of bronchiectasis such as foetid sputum and evidence of toxæmia.

2. The view is held that bronchial dilatation occurs in these cases secondarily to collapse of the lower lobe of the right or left lung, with retention of bronchial secretion. Extension of the process may occur from spilling-over of retained secretion into the adjacent bronchi.

3. Collapse of one lower lobe probably occurs most frequently as a result of, or coincident with, broncho-pneumonia in early childhood, but the possibility of either inhalation of a foreign body or of pulmonary tuberculosis being responsible in certain cases is also considered.

4. Treatment should be directed to the re-expansion of the collapsed lung tissue in the early stages, and to prevention of secondary infection when fibrosis has occurred. It is doubtful if lobectomy is as yet advisable for cases of this type in childhood.

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