BENIGN SPONTANEOUS PNEUMOTHORAX: A STUDY OF 70 CASES.

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Since the form of pneumothorax which is known variously as simple, benign, idiopathic, pneumothorax in the apparently healthy, and pneumothorax simplex was first described by McDowell in 1856, the condition has been the subject of continued study and investigation. By definition it is considered to be pneumothorax occurring in an apparently healthy person in whom there is no clinical or radiological evidence of underlying respiratory disease. It is mainly in the case of precise etiology that agreement has not yet been reached. For a number of reasons reliable information about certain aspects of the disease have become available only within comparatively recent years. First and most important of these is lack of material. Benign spontaneous pneumothorax is a comparatively rare disease. Thus, in one major Glasgow hospital over a ten-year period, it constituted 0.032 per cent of patients admitted and treated to a termination. It was calculated that, on an average, not more than one case per annum would be admitted for treatment to each medical unit. There is no means of assessing the number of cases too mild for medical advice to be sought, and which therefore escape diagnosis, or those cases which are treated out of hospital; but there is no reason to suppose that it is substantial. A second factor is that benign spontaneous pneumothorax, as its name suggests, is a rare cause of death: only a few post-mortem reports have been published. Necropsy has yielded valuable information; but it has often been inconclusive, or negative (Priest, 1937). Finally, the physician is naturally reluctant to subject to specialized investigation a patient for whom the immediate and distant prognosis is excellent, who is usually free from all symptoms within a few days of admission, and whose lung will generally be found to have completely re-expanded within a month. The thoracoscope is an instrument likely to provide essential information in the individual case (Brock, 1948), but it is available for use only in specialized Thoracic Units to which the majority of pneumothorax patients never have occasion to be admitted.

This paper presents a study of 70 cases of benign spontaneous pneumothorax by the follow-up method, with particular reference to etiology, and to the subsequent incidence of recurrence and of pulmonary tuberculosis.

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MATERIAL.

Seventy patients who were admitted for treatment of benign spontaneous pneumothorax to the medical wards of two major Glasgow hospitals in the ten-year period 1939-48, were asked to report for clinical examination, interrogation and chest X-ray, and 58 did so. The remaining 12, who were unable or unwilling to come to hospital, provided the desired information by letter, telephone, or to an almoner. Special attention was paid to the incidence of previous chest diseases, including asthma, and inquiry was made as to the occurrence of pneumothorax in other members of the family. The patients were asked for detailed information of the type of physical exertion, if any, in which they were indulging at the time of their first, or any subsequent, pneumothorax. The results are summarized in Table 1.

TABLE 1.

Benign Spontaneous Pneumothorax: Summary of Results.

Number of cases	70
Average age (years)	29.7
Sex: Males	65
Females	5
Location: Left	34
Right	31
Bilateral, consecutive	5
Deaths	0
Recurrences: One	9
Two or more	7
Total	16
Developed pulmonary tuberculosis	4

DISCUSSION.

Etiology. It is no longer considered that active pulmonary tuberculosis is necessarily responsible for the production of benign spontaneous pneumothorax. Although it was once thought that ulceration of the visceral pleura by a subpleural tubercle was the cause in 90 per cent of these cases (Tice, 1932), in the benign form evidence of active pulmonary tuberculosis must be exceedingly rare. No post-mortem report has been published showing the condition to be due to a ruptured subpleural tubercle. Even in three fatal cases associated with active pulmonary tuberculosis, the pneumothorax was shown to have been due to rupture of bullae, and not to tuberculous pleural ulceration (Perry, 1939). The presence of hydrothorax or of pleural adhesions is not considered as evidence in favour of a tuberculous etiology (Rottenberg & Golden, 1949). There is now much evidence to support the view, and considerable agreement, that the essential lesion in the majority of cases is rupture of emphysematous bullae. Ordinarily, these cannot be diagnosed at the time of the pneumothorax, though in some cases they may be visualised by radiography in later years. Gordon (1936) was of the opinion that all

cases were due to this cause. The healing of primary tuberculous lesions with scar-tissue formation resulting in an area of localized emphysema, with or without bronchiolar constriction, is thought to be an important factor in the etiology (Gordon, 1936 ; Taschmann, 1944). The formation of a valvular mechanism, as described by Hayashi (1915), has been noted, and pleural adhesions may be associated (Weber, 1921).

A congenital anomaly of the pleura appears to be a feature of a certain number of cases. Schminke (1928) describes a case of bilateral spontaneous pneumothorax in which multiple emphysematous bullae were found after death. He believed that a developmental anomaly in the lung tissue was responsible, with persistence of a peripheral zone of embryonal tissue not differentiating into alveoli : cyst formation resulted. In 9 cases reported by Norge (1940), the author supports the view of Kirschner (1938) in maintaining that these cases were due to a congenital pleural defect, with secondary formation of pleural blebs. In an important contribution Brock (1948) reports the results of inspection of the visceral pleura by thoracoscopy in 6 cases, in which air could be seen exuding from the pleura in the form of particles of fine froth (' cuckoo-spit '). He suggested that there was either a congenital or acquired defect of quality in the pleura (' porous pleura '), and that possibly imperfect development of elastic tissue was responsible. The latter is known to be notably defective in generalized emphysema.

In the opinion of Miller (1947) the first step in the formation of bullae is overdistension of the upper lobes in active, robust men under the influence of severe exertion. This observation is in accordance with Austin Flint's teaching that emphysema is always greater in the upper lobes than elsewhere. In this connection it is interesting to recall that in men, as opposed to women, respiration is predominently abdominal, and to consider whether this factor may be responsible for the striking sex incidence (see Table 1), for which no explanation has ever been offered.

Macklin (1939), by forcible overinflation of the lungs of cats, was able to produce localized interstitial emphysema. He noted that occasionally air would pass peripherally, with formation of subpleural bullae, and the production of a spontaneous pneumothorax. He maintained that the latter condition was most commonly produced in this way. Hamman (1939) was in agreement. The view that spontaneous pneumothorax is secondary to mediastinal emphysema, however, is contrary to the accepted teaching. It receives no support from the observation of Rottenberg and Golden (1949) that it did not occur in any of their ninety-seven cases. In the present series it was noted in only one case.

Some doubt still exists as to the importance of exertion in the production of pneumothorax. The condition has been found, by Kjaergaard (1932), not to be more common in heavy and manual workers.

Indeed, it is recorded by Leach (1945) that in 85 out of 100 cases the onset occurred while the patient was at rest or mildly active. An important observation was that of West (1884), who showed that the normal pleura cannot be ruptured by coughing or exertion. In post-mortem specimens he found that an inflation pressure of over 200 mm. of mercury was required to rupture the pleura, and remarked that it was never subjected to such a pressure during life. These findings were endorsed by Diez (1929). Approaching the problem from the opposite viewpoint, Heath (1946) noted that the disease was caused by aerial ascent only in excessively rare instances. During the course of 86,916 man flights, spontaneous pneumothorax occurred in only one subject. Friesdorf (1927) studied the rôle of effort in 177 cases and gave as his opinion that 40 per cent of cases occur after considerable exertion; 40 per cent of cases occur after slight exertion, and 20 per cent of cases occur after trivial movement. In this series precise details were obtained of the mode of onset in 78 separate pneumothorax incidents, with the following results :—

21% of cases occurred after severe exertion.
31% ,, ,, moderate exertion.
25% ,, ,, slight exertion.
23% ,, ,, at rest.

Perry (1939) has suggested that respiratory movements of greater amplitude following a period of shallow respiration may play a part in precipitating the onset. Thirty-five per cent of cases had this mode of onset in his series. It is not uncommon for pneumothorax to occur when the patient wakes from sleep, or on rising and dressing. In this series it has certainly been the impression that change in amplitude of respiration is a more important factor in the production of pneumothorax than the general level of physical activity. Thus, one patient developed a pneumothorax during a bout of hearty laughter while sitting in a cinema. Another was seized with severe chest pain while using a chest expander; he was wakened from sleep by the onset of his second pneumothorax seven years later. Finally, the observation of Rottenberg and Golden (1949), following a study of 97 consecutive cases, may be noted. They recorded that 'the association of exertion with the precipitation of pneumothorax was notable for its infrequency.' Possibly the importance of exertion has been over-emphasized, because it appears to play a contributory but non-essential part in the etiology.

Recurrent spontaneous pneumothorax. At one time it was considered that the incidence of recurrence in benign spontaneous pneumothorax was very low. In 1929, on reviewing the 358 cases of pneumothorax recorded in the literature to that date, Perry noted that only 33 had been recurrent. More recently it has been stressed, particularly by Brock (1948), that recurrent cases are more common than has been supposed. Indeed, Ornstein and Lercher (1941) have observed that repeated attacks may

be experienced by as many as 30 per cent of patients. Recurrence is known to be most likely to occur within a year of the initial illness, and fifteen or more repeated attacks have been recorded in a single subject. In the present series the incidence of recurrent pneumothorax was 23 per cent, and the maximum number of pneumothoraces occurring in one patient was 8. Although in general the prognosis is excellent, it is necessary for the physician to note that recurrences will follow in as many as 1 in 4 of his cases of benign spontaneous pneumothorax, and that in a number artificial pleurodesis, which was performed with satisfactory results in 4 of the present series, will be necessary.

Relation of benign spontaneous pneumothorax to subsequent pulmonary tuberculosis. In discussing the etiology of benign spontaneous pneumothorax, attention has been drawn to its non-tuberculous nature. Emphasis was laid on this point by Kjaergaard (1932) who, in a classical contribution to the subject, found that pulmonary tuberculosis was a very uncommon sequel. Only 1 of his 51 patients developed tuberculosis later, and this patient had been heavily exposed to the risk of infection. In a study of 250 cases recorded in the literature, Perry (1939) noted that tuberculosis had followed in 6, while in 67 of his own cases which he was able to trace, none had developed the disease. He concluded that the post-pneumothorax incidence of tuberculosis is not greater than 2 per cent, and that if pulmonary tuberculosis is not present at the time of the pneumothorax, it is no more likely to occur than in the average individual in the community.

In this series four cases developed pulmonary tuberculosis subsequently. The disease ensued 1 month, 3, 7 and 11 years respectively after the original pneumothorax. None died ; two recovered and two are still under observation. All four were adult males, the average age being 33 years. Of the 70 cases studied, 56 were admitted to hospital from the City of Glasgow, while 14 had their homes in the surrounding counties. In respect of the 4 patients who developed tuberculosis, 2 were domiciled in Glasgow and, of the remaining 2, 1 each was resident in the adjacent counties of Lanarkshire and Dunbartonshire. It is not uncommon to find reports of solitary cases of tuberculosis following pneumothorax (Hamman, 1916 ; Kleeman, 1918 ; Lindhagen, 1932), but as many as 4 in any one series has not yet been recorded. It was considered of interest to relate the latter observation to the incidence of pulmonary tuberculosis in the general community in the area, and to draw a similar comparison in the case of series reported by previous authors (Table 2). It will be noted that, if the unduly high incidence of pulmonary tuberculosis in this area is taken into consideration, there is no conflict with the view that tuberculosis is no more likely to develop in the subject of benign spontaneous pneumothorax than in the average individual in the general community.

TABLE 2.

Incidence of pulmonary tuberculosis following pneumothorax in published series of cases, with corresponding figures for incidence of pulmonary tuberculosis in the community.

Series.	No. in Series.	Number developing tuberculosis.	Area.	Tuberculosis morbidity per 100,000 of population.	Year. *
Kjaergaard (1932)	51	1	Copenhagen ..	(74)†	1932
Perry (1939)	67 (traced)	0	London ..	124	1939
Ornstein & Lercher (1941)	58	3	New York ..	123	1941
Rottenberg & Golden (1949)	97	0	New York ..	97	1947
Present Series (1950) ..	70	4	2 City of Glasgow	250	1948
			1 County of Lanarkshire ..	188	1948
			1 County of Dunbartonshire	74	1948

* Where the year for which the tuberculosis incidence is stated does not correspond with the year of publication of the author's communication, the most recent available figures are quoted.

† The tuberculosis morbidity rate not being obtainable, the mortality rate is stated.

Physique in pneumothorax. Taschmann (1944) considered that benign spontaneous pneumothorax occurred most frequently in tall, thin, adult males, and Norris (1940) found that two-thirds of his 26 cases were underweight. No similar observations have been recorded by other authors. On physical examination of 58 cases in the present series, no clinical evidence could be obtained to support the view that the physique of the subjects was related to the occurrence of the disease in any way. The heights and weights of an additional 3 patients were taken while in hospital, and it was found that they were respectively 75 per cent, 79 per cent and 96 per cent of the expected weight for age, height and sex. It is doubtful whether physique plays a significant part in the production of benign spontaneous pneumothorax, or predisposes patients to the disease.

Familial spontaneous pneumothorax. It has been suggested that pneumothorax may have a familial incidence (Bachmann, 1940). Hereditary congenital lung cysts are said to be responsible. Spontaneous pneumothorax occurring in two brothers is recorded by Larsen (1933). Two cases are reported in a family by Willis (1937), while as many as five members are said to have been affected (Gotzsche, 1933). There were no familial cases in the present series, and in the 640 cases of benign spontaneous pneumothorax hitherto recorded none, other than those mentioned, has been found. It is open to question whether familial spontaneous pneumothorax merits consideration as a separate entity.

SUMMARY.

The results of a follow-up of seventy cases of benign spontaneous pneumothorax admitted to the general medical wards of two major Glasgow hospitals in the ten-year period 1939-48 are recorded. The comparative rarity of the condition is noted. The etiology is discussed. Physical exertion is not considered to be an important factor in the causation. Twenty-three per cent of the cases were recurrent. Four cases developed pulmonary tuberculosis. The subsequent incidence of pulmonary tuberculosis has been related to the tuberculosis morbidity in the general community. No special type of physique was found to be associated with the disease. There were no familial cases.

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