Hirschsprung's Disease—Fraser and Wilkinson

factory histological examination; it should be done only when the result of radiological examination is equivocal.

A third of the patients in this series died, a clear indication of the serious potentialities of Hirschsprung's disease. That such a high mortality should be found in a series of patients in whom the disease had been recognized within the first month of life adds weight to the former estimates (Bodian et al., 1949; Eek and Knutrud, 1962) that 50% died in the first year of life.

The higher mortality in neonates has also been emphasized by Hofmann and Rehein (1966): 15 of their 53 neonates died. They prefer to treat the condition by washouts and hot packs rather than by colostomy, to which they resorted only when conservative treatment failed. Only two of their patients died during conservative treatment and the remaining 22 survived subsequent resection of the aganglionic bowel. However, of the 31 in whom a colostomy had to be made, 8 died and 19 had a complicated course, and five more died subsequently after resection of the affected bowel.

It seems to us that when Hirschsprung's disease is diagnosed soon after birth the safest form of treatment is decompression of the obstructed bowel by a colostomy or ileostomy with at least delay as possible. Resection of the abnormally innervated segment of bowel should be delayed until the child is 1 year old or weighs 20 lb. (9.1 kg.).

It is not surprising that the mortality rate should be higher in the babies with the "long segment" type of disease, especially when the full extent of the lesion is not recognized at the first or subsequent operations to relieve the obstruction, and the colostomy or ileostomy is first made in aganglionic bowel.

In Hirschsprung's disease the term "abnormality of bowel habit" may imply either constipation or bouts of diarrhoea owing to the passage of faecal stained mucus round an impacted mass of faeces. In its most extreme form diarrhoea may be due to a sort of enterocolitis with mucosal ulceration and toxaemia that is often fatal. In another series of 47 patients with Hirschsprung's disease (Bill and Chapman, 1962) there were 24 with enterocolitis, of whom 21 were neonates and 8 died; though in this series enterocolitis did not occur after colostomy, it did in two of our patients (Cases 8 and 9). Bill and Chapman also found that patients who had once had enterocolitis were liable to its recurrence, especially after rectosigmoidectomy. Swenson and Davidson (1960) have reported a mortality rate of 33%, in patients who were treated medically, but in those for whom a colostomy was made as soon as the disease was diagnosed the mortality rate was only 4%. It seems that the best way to prevent enterocolitis may be to make a colostomy as soon as the diagnosis of Hirschsprung's disease is made, but it must be recognized that this does not always provide complete protection from enterocolitis or prevent death from it.

**Summary**

Of 54 babies suffering from Hirschsprung's disease who were admitted within a month of birth, 18 (33%) died—5 of enterocolitis and 11 of other complications of the treatment; 2 mongols were not treated surgically.

The clinical disturbance associated with this form of congenital intestinal obstruction usually begins within the first two or three days after birth and is characterized by vomiting, abdominal distension, and abnormality of the bowel action. The clinical diagnosis was confirmed by x-ray examination after a barium enema in 24 out of 27 patients in this series; in one patient the appearances were equivocal and in two others the bowel had been washed out before the examination.

Neonatal intestinal obstruction due to Hirschsprung's disease should be relieved by a colostomy or ileostomy as soon as the diagnosis is established.

We are indebted to the late Mr. G. H. Macnab, Mr. D. J. Waterston, and Mr. H. H. Nixon for allowing us to include patients under their care in this survey.

**REFERENCES**


Overinflation of the Lungs in Coal Miners

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*Brit. med. J.*, 1967, 3, 10–14

Coal miners whose chest radiographs are normal tend to be more breathless than non-miners of the same age group, but a pattern of dysfunction distinct from chronic non-industrial lung disease has not been observed.

The purpose of this report is to compare the results of lung-function tests in coal miners referred to a physiological laboratory because of unexplained dyspnoea with those obtained in a group of men suffering from chronic non-specific lung disease.

**Patients**

The first group consists of 17 miners (two from Yorkshire and 15 from West Lancashire coal fields) who were referred during 1963–6 to a lung-function laboratory providing a routine service for the region. In each case the miner had been referred by a consultant physician because physical and radiological examination had failed to reveal an adequate cause for his disability. These 17 cases represent the total number of miners referred to the laboratory in this way. Apart from clinical or radiological signs compatible with either chronic non-specific lung disease or pneumoconiosis, no cardiac or pulmonary cause for dyspnoea could be found in any of these patients. The only miners attending the laboratory during 1963–6 and excluded from this report were those referred for routine preoperative assessment or for the investigation of a disease not directly related to their occupation (mitral stenosis, bronchiectasis, bronchial carcinoma, or tuberculosis).

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† Senior Medical Registrar.
‡ Medical Registrar.

Regional Cardiothoracic Centre, Broadgreen Hospital, Liverpool 14.
The occupational history and the clinical and radiological findings for the 17 miners are recorded in Table I. The average age was 57 years at the time of the investigation. Only five of the miners were still working underground, the remainder having left the mines an average of five years previously. The average duration of underground work was 32 years, 12 of the 17 miners having worked underground for 30 or more years. Of the five with less than 30 years underground, two had worked in other dusty occupations (26 years in a lead mine and seven years in a granite quarry respectively). The 17 miners all complained of either moderate (grade 2) or severe (grade 3) dyspnoea on effort (see Methods). The average duration of effort dyspnoea was six years. Five miners denied having any cough, while 12 complained of cough with a variable amount of sputum. Most of them stated that cough and dyspnoea started together, but none was sure about the total duration of cough.

### Table I.—Occupational and Clinical History and Radiographic Findings in Group 1 (Coal Miners)

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Years Under-</th>
<th>Years or</th>
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<th>Grade of</th>
<th>Signs of</th>
<th>X-ray Evidence of</th>
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<td>Pneumoconiosis</td>
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* Plus 26 years in lead mines.
† Plus 7 years in granite quarry.

### Physical Signs of Bronchitis

Physical signs of bronchitis were present in seven: expiratory wheeze was evident in five and coarse basal crepitations without wheeze in two others. The chest radiographs showed evidence of simple pneumoconiosis (categories 1–3) in eight cases and were clear in the remaining nine.

The second group consists of 15 male non-miners aged 50 and over referred to the laboratory as cases of chronic bronchitis and/or emphysema. These patients fulfilled the criteria required for a diagnosis of chronic non-specific lung disease: they were all suffering from chronic cough with sputum and/or persistent breathlessness not associated with localized lung disease of any kind, generalized specific infective diseases of the lungs (pneumoconiosis, generalized pulmonary fibrosis or granulomata, primary cardiovascular or renal disease, diseases of the chest wall, anaemia, or psychoneurosis (Ciba Guest Symposium, 1959). The 15 patients represent the total number of males aged 50 and over with chronic non-specific lung disease for whom adequate physiological data were available in the laboratory records.

### Methods

The physiological techniques used are those described in previous publications (Ogilvie et al., 1963), and include: (1) the subdivisions of the lung volume, including vital capacity (V.C.), functional residual capacity (F.R.C.), and residual volume (R.V.); (2) the forced expiratory volume in one second (F.E.V.1.); (3) the forced inspiratory volume in one second (F.I.V.1.); (4) the forced expiratory volume in one second (F.E.V.1.); (5) arterial P02 tension by the rebreathing method (P02); and (6) transfer factor for carbon monoxide (diffusing capacity, D.L.) by the single-breath technique.

The degree of dyspnoea was recorded for each miner before the physiological tests were carried out, according to the following grading:

- Grade 1 (mild). Dyspnoea only on severe exertion such as running on the level or hurrying upstairs.
- Grade 2 (moderate). Dyspnoea on one flight of stairs or walking upstairs inclines. No dyspnoea when walking at a normal pace on level ground.
- Grade 3 (severe). Dyspnoea when walking at a normal pace on level ground.

The normal values for lung volumes were predicted from the formula of Baldwin et al. (1948). The normal values for transfer factor were predicted from the formula of Ogilvie et al. (1957). The results in the two groups were also compared with those obtained in five normal men aged 50 and over studied in the same laboratory. These five men had been referred to general surgical clinics of the hospital from the same area as the
TABLE III.—Physiological Findings in Group 2 (Chronic Non-specific Lung Disease)

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TABLE IV.—Physiological Findings in Five Normal Men

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Fig. 1.—Vital capacity. Fig. 2.—Residual volume. Fig. 3.—Total lung capacity. Fig. 4.—F.E.V., as percentage of vital capacity. Fig. 5.—M.I.F.R./M.E.F.R. ratio. Fig. 6.—Transfer factor.
miners and were all free of symptoms, signs, and radiological
evidence of respiratory disease.

**Results**

The vital statistics and physiological findings in the two
groups are shown in Table II (miners) and Table III (non-
specific lung disease) and the results of the physiological tests
are compared in Figs. 1 to 6. The results obtained in the five
normal subjects are recorded in Table IV and are also shown in
Figs. 1 to 6.

**Vital Statistics.**—The mean age was 57 years (range 41–66)
in the miners (group 1) and 58 years (range 50–70) in patients
with chronic non-specific lung disease (group 2). The mean
height was 67 in. (170 cm.) (range 63–72 in.; 160–183 cm.)
and mean weight 151 lb. (68.4 kg.) (range 106–182 lb.; 48–
82.5 kg.) in group 1, and 67 in. (170 cm.) (range 63–69 in.;
160–175 cm.) and 141 lb. (63.9 kg.) (range 109–187 lb.; 49.4–
84.8 kg.) respectively in group 2.

**Lung Volumes.**—In Table V the mean values for V.C, R.V.,
and T.L.C. are expressed as the percentage of predicted normal
according to the formula of Baldwin et al. (1948). Results are
shown for the two groups in the present study and for the five
normal subjects. According to the formula of Baldwin et al.
and the findings in our own normal subjects, the residual
volume is increased to about two and a half times the predicted
normal in both groups, while the total lung capacity is increased
by nearly one-third in the miners and by nearly one-quarter in
the patients with non-specific lung disease (see also Figs. 2
and 3).

**Ventilatory Capacity.**—The impairment of ventilatory
capacity was considerably greater in group 2 than in group 1,
the mean M.V.V. having nearly twice the value in the latter
group (see Table V). This difference was due to the greater
degree of expiratory airways obstruction in group 2: the mean
M.I.F.R. was approximately the same in the two groups, but the
mean M.E.F.R. in group 2 was less than one-quarter of the
value obtained in group 1. The values for the M.I.F.R./
M.E.F.R. ratio and for the F.E.V.1 as percentage of vital
capacity are compared for the two groups in Figs. 4 and 5. It
will be noted that in 12 of the miners but in none of the other
group, the M.I.F.R./M.E.F.R. ratio was within the normal
range (0.5–1.5). Eight miners, but none in the other group,
showed a normal F.E.V.1 as percentage of V.C. (over 60%).

**Transfer Factor (Diffusing Capacity).**—The mean transfer
factor as percentage of predicted normal was 99% in group 1
and 42% in group 2. Fourteen of the 16 miners had a normal
transfer factor (over 75%) as compared with 3 of the 15 patients
in group 2 (see Fig. 6).

PCO₂.—This was measured by the rebreathing method in
seven patients from each group. The mean value for PCO₂ in
these patients was 36 mm. Hg in group 1 and 43 mm. Hg in
group 2. The PCO₂ was elevated in one patient from group 1
and in two patients from group 2.

**Discussion**

In any study of disability in coal miners the mode of selection
is clearly important. In the present investigation the miners had
all been referred by a general practitioner to a consultant
physician and thence to a regional pulmonary function labora-
tory because physical and radiological examination had not
revealed an adequate cause for their disability. Inevitably
the question of eligibility for pension arose in a number of
these patients, and in several the possibility of a "compensation
neurosis" had been raised by the referring physician. It is not
surprising, therefore, that more than half of these miners had
no physical signs of bronchitis nor any radiological evidence
of dust in the lungs and that only two (Cases 4 and 5) were in
receipt of a pension for pneumoconiosis.

This group of miners had certain other features in common,
less obviously related to the method of selection. Fifteen of the
17 were over the age of 50 and 12 had left the mines several
years previous to the investigation. Twelve of the 17 had spent
30 years or more underground and, of the remaining five, two
had been engaged in other forms of dusty work. Finally, they
all showed an increase in residual volume, total lung capacity,
and R.V./T.L.C.% comparable to that observed in patients
with chronic non-specific lung disease but without a commensurate
degree of expiratory airways obstruction or impairment of the
transfer factor.

Several authors (Gilson and Hugh-Jones, 1955; Carpenter
et al., 1956; Higgins et al., 1956; Leathart, 1959) have found
that miners with normal chest radiographs have a poorer M.V.V.
than those whose films show simple pneumoconiosis. Further-
more, Gilson and Hugh-Jones (1955) reported a close correla-
tion between M.V.V. and dyspnoea in coal miners, but when a
group in hospital was studied the correlation between R.V./
T.L.C.% and dyspnoea was almost as good. In Fig. 7 M.V.V.
has been plotted against R.V. for the miners in the present study
and the grade of dyspnoea is indicated for each case. The eight
miners with simple pneumoconiosis tend to have a higher
M.V.V. and smaller residual volume (mean M.V.V. 80 l/min.;
mean R.V. 3.3 l.) than the miners with clear films (mean M.V.V.
50 l/min.; mean R.V. 4.8 l.). Moreover, the grade of dyspnoea
in these eight miners (grade 2) was less than in seven of the
nine with clear films (grade 3). There appears, therefore, to be some
relation between dyspnoea on the one hand and a low M.V.V.
with increased residual volume on the other, but this relation is
not necessarily a causal one, for there are many other variables
of lung function which have not been measured in the present
study. For example, uneven distribution of inspired gas (Gilson
and Hugh-Jones, 1955), disturbances of the mechanical
properties of the lungs (Leathart, 1959), and an increased
alveolar-arterial Pco₂ difference (Brasseur, 1963) have all been
reported in coal miners.
Psychiatric Study of a Consecutive Series of 34 Patients with Ulcerative Colitis

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The purpose of this study is to evaluate the role of psychiatric factors in the causation of ulcerative colitis by survey of a fairly long unselected series. Ulcerative colitis is still one of the most enigmatic of diseases, despite its position, paradoxically, as the “classic” psychosomatic illness. Its cause is believed by most gastroenterologists to be unknown, yet a large number of reports by psychiatrists discuss it as an unquestionably psychosomatic illness. The studies by physicians and surgeons of such medical factors as cause, course, complications, and drug treatment have been for the most part cautious, uncertain, and qualified (Crohn and Yarnis, 1951 ; Michener et al., 1961 ; Hijmans and Enzer, 1962 ; Koreitz et al., 1962 ; Edwards and Truelove, 1963, 1964 ; Rhodes and Kirsner, 1965). On the other hand, reports by psychiatrists of much smaller series (Engel, 1961 ; Finch and Hess, 1962 ; Fullerton et al., 1962 ; Powles, 1964), sometimes random and unselected, have used such terms as “constantly show severe psychopathology,” “uniformly show pathology,” “in every case,” and “the evidence is overwhelming, incontrovertible.”

We are aware that psychiatrists recognize that the disease is one which may be multifactorial in origin, and that some of them regard the psychogenic component as only one factor out of many. Nevertheless, the presence of severe psychopathic disease is so consistently described that the impression is gained from leading psychiatric papers that an emotional component is necessary for the condition to exist. Both sophisticated psychiatric readers and general physicians seem to derive this