- ¹⁴ Salmon, S E, *et al*, *New England Journal of Medicine*, 1970, **282**, 250.
¹⁵ Baehner, R L, and Nathan, D G, *New England Journal of Medicine*, 1968, **278**, 971.
¹⁶ Holmes, B, *et al*, *Journal of Clinical Investigation*, 1967, **46**, 1422.
¹⁷ Sbarra, A J, and Karnovsky, M L, *Journal of Biological Chemistry*, 1959, **234**, 1355.
¹⁸ Rous, P, *Journal of Experimental Medicine*, 1925, **41**, 399.
¹⁹ Kakinuma, K, *Journal of Biochemistry*, 1970, **68**, 177.
²⁰ Keitt, A S, *American Journal of Medicine*, 1966, **41**, 762.
²¹ Tanaka, K R, and Paglia, D E, *Seminars in Hematology*, 1971, **8**, 367.
²² Paglia, D E, *et al*, *Journal of Clinical Investigation*, 1970, **49**, 72a.
²³ Löhr, G W, *et al*, *Lancet*, 1968, **1**, 753.
²⁴ Keller, H V, *et al*, *Antibiotics and Chemotherapy*, 1974, **19**, 112.
²⁵ Gallin, J I, *et al*, *Journal of Immunology*, 1973, **110**, 233.
²⁶ Miller, M E, *et al*, *Lancet*, 1968, **2**, 60.
²⁷ Chandra, R K, *et al*, *Lancet*, 1969, **2**, 71.
²⁸ Quie, P G, *et al*, *Journal of Clinical Investigation*, 1966, **45**, 1058.
²⁹ Motulsky, A G, and Yoshida, A, in *Biochemical Methods in Red Cell Genetics*, p 74, ed J J Yunis. New York, Academic Press, 1969.
³⁰ Paglia, D E, and Valentine, W N, *Journal of Laboratory and Clinical Medicine*, 1967, **70**, 158.
³¹ Beutler, E, *Red Cell Metabolism*. New York, Grune and Stratton, 1971.
³² Kaplow, L S, *Blood*, 1965, **26**, 215.
³³ Ng, R P, *et al*, *Lancet*, 1975, **2**, 901.
³⁴ Ng, R P, and Alexopoulos, C, *Journal of Immunological Methods*. Awaiting publication.
³⁵ Espamol, T, *et al*, *Clinical and Experimental Immunology*, 1974, **18**, 73.
³⁶ Klebanoff, S J, *Annual Review of Medicine*, 1971, **22**, 39.

Coeliac disease with farmers' lung

T J ROBINSON

British Medical Journal, 1976, **1**, 745-746

Summary

Two patients with allergic alveolitis due to mouldy hay antigens (farmer's lung) were shown to have malabsorption due to coeliac disease. As similar associations have been found with other alveolar diseases, this association is probably not fortuitous and further population screening should be done.

Introduction

Several recent case reports have shown an association between coeliac disease and diffuse interstitial pulmonary disease. Villous atrophy due to a gluten-sensitive enteropathy has recently been described with extrinsic allergic alveolitis due to avian exposure (bird fancier's lung)¹ and there are reports of sarcoidosis² and fibrosing alveolitis³ occurring with coeliac disease. Apparently patients with coeliac disease have an increased incidence of other autoimmune disturbances, although the meaning of these changes is not clear. Abnormalities of immunoglobulins in both serum and jejunal secretions have been recorded in coeliac disease,⁴⁻⁸ as has an association with Addison's disease,⁹ Sjögren's syndrome,^{10,11} thyroid disease,¹⁰ and ulcerative colitis.¹²

Fibrosing alveolitis is associated with hyperglobulinaemia¹³ and is found in associations with conditions of possible autoimmune aetiology such as ulcerative colitis,¹⁴ Hashimoto's thyroiditis,¹⁵ rheumatoid arthritis,¹⁵ and Sjögren's syndrome.¹⁴⁻¹⁶ The lungs are, of course, often affected in connective tissue disorders, as in rheumatoid arthritis, systemic sclerosis, polyarteritis nodosa, and systemic lupus erythematosus.

I report here two cases of extrinsic allergic alveolitis due to mouldy hay (farmer's lung) occurring in patients with coeliac disease.

Case 1

A hill farmer aged 40 was admitted with a two-month history of progressively increasing shortness of breath, weight loss, and general

malaise. He had close contact with hay and straw as he carried bales on his back each day to feed the cattle in the fields.

He was dyspnoeic at rest and there were widespread fine crepitations heard throughout both lung fields. X-ray examination of the chest and gel immunodiffusion against *Mycoplasma faeni* confirmed farmer's lung. The symptoms quickly improved in hospital without any specific treatment. His chest improved clinically, although it was some months before his chest x-ray appearances reverted to normal.

Four years earlier he had attended a surgical outpatient clinic with mouth ulcers and diarrhoea. Mouth biopsy had shown chronic non-specific ulceration. His haemoglobin had been 10 g/dl and the white cell count $4.6 \times 10^9/l$ ($4600/mm^3$). The red cells had shown a dimorphic picture. No treatment had been given.

Investigation on the second admission showed: haemoglobin 13.6 g/dl, red cell count $5.4 \times 10^{12}/l$ ($5.4 \times 10^6/mm^3$), packed cell volume 49.9%, mean cell volume 76 fl ($76 \mu m^3$). Thyroid function was normal. Serum carotene was 0.80 $\mu mol/l$ ($43 \mu g/100 ml$) (normal 1.1-2.2 $\mu mol/l$ (60-120 $\mu g/100 ml$)), serum calcium 2.1 mmol/l (8.4 mg/100 ml) (normal 2.1-2.6 mmol (8.4-10.4 mg/100 ml)), folate <0.5 $\mu g/l$ (normal 3.5-24.0 $\mu g/l$), serum lactic dehydrogenase 624 U/l (normal 100-225 U/L), total protein concentration 81 g/l (normal 60-80 g/l), albumin 46 g/l (normal 35-50 g/l), and globulin 35 g/l (normal 25-30 g/l). He weighed 53 kg. Jejunal biopsy showed subtotal villous atrophy. He was started on a gluten-free diet with considerable improvement in his general health. At the last review, six months after his admission, he had put on 8 kg in weight, and his chest was clear both clinically and on x-ray examination. Haemoglobin was 15 g/dl and calcium levels were normal.

This man had classical extrinsic alveolitis due to mouldy hay but had a concomitant coeliac syndrome, which had been producing symptoms for at least four years.

Case 2

A farmer's wife aged 58 attended another hospital in 1968 complaining of shortness of breath, weight loss, and epigastric soreness. She was recorded as having bilateral basal pleural rubs and being dyspnoeic at rest but not cyanosed. One chest x-ray picture was reported as being normal. Another several weeks later showed fine miliary mottling. Haemoglobin was 14.6 g/dl with a normal white cell count and differential count. Total proteins were increased, with the albumin globulin ratio at 0.9. Serum electrophoresis showed an increase in α_1 - and γ -globulin.

She was readmitted in 1975, when aged 65, and was found to be acutely dyspnoeic. There were crepitations in both lung fields. She was apyrexial. Sputum culture gave a heavy growth of *Klebsiella pneumoniae* and she was treated with antibiotics. Serial sputum cultures for tuberculosis were negative. Haemoglobin was 12.7 g/dl and white cell count $8.1 \times 10^9/l$ ($8100/mm^3$) with an eosinophil count of 6%. Chest x-ray examination showed widespread chronic inflammatory changes. Tests for extrinsic alveolitis showed a positive gel immunodiffusion against *M faeni*. Farmer's lung was diagnosed clinically and on discharge from hospital she was feeling better.

Craigavon Area Hospital, Craigavon, Co Armagh, Northern Ireland

T J ROBINSON, MD, MRCP, consultant physician

She was admitted to this hospital in April 1975, when she complained of persistent dyspnoea, malaise, and chest pain. She was thin, dyspnoeic at rest, but not cyanosed or anaemic. There were widespread crepitations throughout both lung fields and some expiratory bronchospasm. Investigations were arranged because of a history of diarrhoea and the results were as follows: haemoglobin 13.6 g/dl, white cell count $11 \times 10^9/l$ ($11\,000/mm^3$), total protein 65 g/l, albumin 42 g/l, and globulin 23 g/l. The result of the latex test for rheumatoid arthritis was negative. Serum carotene was $1.0 \mu\text{mol/l}$ ($55 \mu\text{g}/100 \text{ ml}$), serum vitamin B₁₂ 875 ng/l (normal range 150-900 ng/l), and serum folate 0.8 $\mu\text{g}/l$. Within five hours 16% of a 25-g dose of xylose was excreted. There was no antinuclear factor. Calcium concentration was 2 mmol/l (8 mg/100 ml). Faecal fats were normal. Chest x-ray examination showed diffuse fibrotic changes throughout both lung fields—overall appearances that would be consistent with farmer's lung. Jejunal biopsy findings were reported as follows: "Stunting of mucosal villi with an increase in the chronic inflammatory cell content in the lamina propria. The appearances are those of partial villous atrophy."

Discussion

Both of these patients presented initially with respiratory symptoms and gluten enteropathy was only subsequently detected. In retrospect, however, symptoms had been present for some time. Non-specific symptoms are the presenting factors in many adults with coeliac disease and in any population there are probably many people with undiagnosed coeliac disease. Recently Berrill *et al*¹ have suggested that there is a strong case for routine jejunal biopsy in patients with bird fancier's lung. The same may apply to farmer's lung.

In most of the recent reports^{1, 2, 17} avian antigens have been detected in the patients with pulmonary disease associated with coeliac disease but the role of these antigens is not clear. The recent report of Berrill *et al* (incorporating two cases originally described in 1970¹⁸) showed that five out of 16 patients with bird fancier's lung had total or subtotal villous atrophy and had positive avian precipitins with a history of contact with birds.¹ In another report, however,³ avian antigens were not implicated in any of the three cases of diffuse interstitial lung disease found among a population of 24 patients with coeliac disease. In a case of coeliac disease associated with extrinsic allergic alveolitis reported by Scadding² precipitins to pigeon antigens were

present in the blood, and bronchial challenge with pigeon serum produced a febrile reaction after four hours.

Morris *et al*¹⁷ found precipitins against various avian antigens in coeliac patients, although there was no history of close contact with birds. Close contact is not necessary, however, as evidenced by cases of pigeon fancier's lung reported in the wives of two pigeon fanciers who were only indirectly exposed to avian antigens.¹⁹ A patient of my own who is a poultry farmer had coeliac disease first diagnosed when he was referred because of positive avian precipitins without pulmonary disease. Tests for allergic alveolitis were done by his general practitioner because the patient had complained of general ill-health and weight loss. This patient responded well to a gluten-free diet and the avian precipitins became negative.

Further screening of patients with coeliac disease and diffuse lung disease associated with immunological abnormalities including intrinsic allergic alveolitis is necessary to tell how often these conditions co-exist.

References

- Berrill, W T, *et al*, *Lancet*, 1975, **2**, 1006.
- Scadding, J G, *British Medical Journal*, 1970, **2**, 557.
- Lancaster-Smith, M J, Benson, M K, and Strickland, I D, *Lancet*, 1971, **1**, 473.
- Hobbs, J R, and Hepner, G W, *Lancet*, 1968, **1**, 217.
- Asquith, P, Thompson, G A, and Cooke, W T, *Lancet*, 1969, **2**, 129.
- Blecher, T E, *et al*, *Gut*, 1970, **10**, 57.
- Kenrick, K G, and Walker-Smith, J A, *Gut*, 1970, **11**, 635.
- Douglas, A P, Crabbe, P A, and Hobbs, J R, *Gastroenterology*, 1970, **59**, 414.
- Goudie, R B, *et al*, *Lancet*, 1969, **1**, 186.
- Maclaurin, B P, Matthews, N, and Kilpatrick, J A, *Australia and New Zealand Journal of Medicine*, 1972, **4**, 405.
- Pitman, F E, and Holub, D A, *Gastroenterology*, 1965, **48**, 869.
- Salem, S N, and Truelove, S C, *British Medical Journal*, 1965, **1**, 827.
- Hobbs, J R, and Turner-Warwick, M, *Clinical and Experimental Immunology*, 1967, **2**, 645.
- Scadding, J G, *Proceedings of the Royal Society of Medicine*, 1969, **62**, 227.
- Turner-Warwick, M, and Doniach, D, *British Medical Journal*, 1965, **1**, 886.
- Tomasi, T B, jun, Fudenberg, H H, and Finby, N, *American Journal of Medicine*, 1962, **33**, 293.
- Morris, J S, *et al*, *Lancet*, 1971, **1**, 754.
- Hood, J, and Mason, A M S, *Lancet*, 1970, **1**, 445.
- Riley, D J, and Saldana, M, *American Review of Respiratory Disease*, 1973, **107**, 456.

SHORT REPORTS

Inversion of cervix uteri at caesarean section

Partial inversion of the fundus uteri is sometimes observed during caesarean section due to forcible or unwise traction on the cord while attempting to deliver the placenta, but inversion or "invagination" of a partly effaced and dilated cervix into the lower uterine segment at the time of caesarean section is probably never seen by most obstetricians. The extreme rarity of this condition is probably why it has not been reported.

Case report

A 29-year-old primigravida, whose expected date of delivery was 16 June 1975, was admitted to the university obstetric unit on 14 June for her delivery. Since on obstetric examination there was some doubt about the presentation, an x-ray film was ordered which showed a "breech" with extended legs and arms. As the pelvis was adequate and the baby's weight assessed at 2500-3000 g, an assisted breech delivery was contemplated. She went into spontaneous labour on 24 June and an internal examination showed a non-effaced cervix about 6 cm dilated. During labour the fetal

heart rate suddenly became rapid (varying from 160 to 180/min), and since even at this stage (nearly 12 hours after the onset of labour) the cervix was only partly effaced and 6-8 cm dilated a caesarean section was decided on. A routine lower segment caesarean section was performed and the fetus delivered without much difficulty except for some resistance encountered while delivering the head. When the suturing of the uterus was about to start we noticed a pouch-like protrusion, which partly occupied the lower segment. The upper end of the pouch, which extended to almost the level of the uterine incision, was open with a somewhat wavy margin while below it communicated with the vagina (see fig). As the identity of this "pouch" appeared uncertain, and it was lying freely mobile within the lower segment, its open upper margin was partly sutured to the decidua of the lower segment with interrupted catgut. This was wrong as at the end of the operation when the vagina was swabbed we noticed that there was hardly a cervix to be felt, although it had been only partly effaced just before the caesarean section. It was only then that the possibility of invagination (inversion) of the cervix was suspected, which was what had actually happened.

In view of this diagnosis it was decided to divide the sutures anchoring the invaginated cervix to the decidua and reduce the inversion. An attempt towards achieving this was made (nearly four hours after the caesarean section) from below (vaginally), under general anaesthesia, with the patient in the lithotomy position, and the manoeuvre proved successful—the anchoring sutures were identified and divided; the inverted cervix was then hooked with the finger and drawn down, in stages, into the vagina and the inversion corrected. The patient made an uneventful recovery and was discharged on 4 July.