Discussion

Although the very limited size of the primary growth would seem to favour operative procedures such as lobectomy, the patient's age and rapid decline clearly ruled this out.

The precise cause of death is not clear, but the soft necrotic growth in the lung was probably producing considerable toxæmia, which affected the myocardium of an elderly subject. The route of metastasis was probably haematogenous.

We are greatly indebted to Dr. J. E. McCartney, of the Southern Group Laboratory, Park Hospital, for the photomicrographs of the primary and secondary tumours and to the local hospital photographic service of Messrs. Turnham, of Eastbourne, for the photograph of the specimen. We are also indebted to Messrs. Ilford Ltd. for the x-ray photograph.

REFERENCE

Willis, R. A. (1948). *Pathology of Tumours*, pp. 359, 374. Butterworth, London.

SJÖGREN'S DISEASE ASSOCIATED WITH A NUTRITIONAL DEFICIENCY SYNDROME

BY

R. B. RAFFLE, M.B., M.R.C.P.

In 1933 Henrik Sjögren first described the disease which bears his name. The eye changes were first noted, but the condition was later found to involve a large number of other structures. The non-ocular features have been stressed in later papers by Sjögren and by Weber (1945). The present case, which was seen in general practice and which is unassociated with eye changes, shows certain clinical features which seem worthy of record.

Case Report

The patient, a spinster of 50 working as a charwoman, first reported in January, 1949, complaining of depression and vague "rheumatic" pains affecting the occipital, interscapular, and sacral regions. She felt weak, lacked appetite and energy, and was subject to early morning waking. These symptoms had been present for about three months.

Pronounced facial swelling affected both parotid regions, and on inquiry she stated that this had started first on the left side of the face in July, 1947. The initial attack had been diagnosed as mumps, but the swelling had never subsided completely. Instead there had been recurrent exacerbations with incomplete regression, and some weeks later the right side of the face became similarly affected. Later the patient noted swellings under her chin. There had never been any pain. Some weeks after the onset of the swelling she had noticed dryness of her mouth, which became worse and caused difficulty in swallowing and speaking. The dysphagia had become intensified in the last few weeks, and was then associated with a feeling of food sticking behind the larynx. During this time she had persistent soreness of the tongue.

There were no gastro-intestinal symptoms and no paraesthesiae, but for many years she had been subject to a typical Raynaud phenomenon in the fingers during cold weather. She had also had what seems to have been a distal pulp abscess affecting the left middle finger in November, 1948, since when the finger had been persistently cyanotic and subject to attacks of burning pain on warming. It was involved in the "local syncope" of the Raynaud attacks in common with the other fingers, but the attacks lasted longer in this finger and never recovered beyond the stage of cyanosis. The menopause had occurred two years previously. She had never been subject to eye trouble.

Examination on January 19 revealed a spare, pale, nervous woman, looking older than her years and possessing a conspicuous swelling in both parotid regions. Each parotid gland was enlarged to the size of half an orange, and was firm and nodular but not tender. Both submaxillary glands showed a similar plum-sized enlargement. The whole buccal cavity was dry, and the mucosa was dull red in colour and of a matt texture. The tongue was dry, red, and glazed, the papillae not being evident. She also showed an angular stomatitis and cheilosis. The skin was dry, the hair thin, and the eyebrows normal. The left middle finger was cyanosed and the finger-nails showed koilonychia. The heart, lungs, and abdomen revealed no abnormality. The blood pressure was 135/85. The spleen was not palpable.

Blood examination showed: haemoglobin, 70% (Haldane); red cells, 4,200,000; colour index, 0.83; white cells, 5,000 (polymorphs 72%, eosinophils 1%, lymphocytes 15%, monocytes 12%). E.S.R. (Westergren; 200 mm. column): 1 hour reading, 20 mm. The Wassermann reaction was negative. A chest x-ray film revealed no abnormalities. Urine examination showed nothing abnormal.

Progress.—The patient was at first given vitamin A, 150,000 units daily. On February 7 she was not improved in any way, and vitamin A therapy was stopped and ferrous sulphate, 18 gr. (1.2 g.) daily, was administered. On February 23 the haemoglobin was 85% and the red blood cells numbered 4,500,000. There was no improvement in the buccal condition, but she felt subjectively better. "Beplex" capsules, six a day, were given. This provides a daily intake of 6 mg. of thiamin hydrochloride, 4.8 mg. of riboflavin, and 60 mg. of nicotinic acid.

On March 5 the haemoglobin was 105% and the red blood cells numbered 4,800,000. She felt much more "like herself," looked better, was sleeping well, and was no longer fatigued. The soreness of the tongue had gone and also the angular stomatitis and cheilosis, but the xerostomia persisted and the glandular enlargement was unaffected. Her appetite was improved and the dysphagia less troublesome; she attempted to take a more normal diet, and reported that the sensation of food sticking behind the larynx had vanished. Three beplex capsules and 9 gr. (0.6 g.) of ferrous sulphate were continued daily, and the improvement was maintained.

Discussion

The course of the illness seems to fall roughly into two stages—the first, which had started in July, 1947, comprising the gradual increase in size of the salivary glands together with the xerostomia; the second, beginning in October, 1948, and associated with the complaints of fatigue and general ill-health. During this second period the soreness of the tongue and the finger infection had developed.

The salivary gland changes are compatible with the diagnosis of Sjögren's disease, the ocular features of which are sometimes delayed, xerostomia being the presenting symptom. The age, sex, and psychological type of the patient also point to this diagnosis, and the association with Raynaud's phenomenon has been noted before. As Ellman and Weber (1949) point out, it is doubtful how far the condition can be distinguished from Mickulicz's disease.

The glossitis, angular stomatitis, cheilosis, koilonychia, and hypochromic microcytic anaemia of the second stage are clearly the result of nutritional deficiency. In view of the part allegedly played by vitamin deficiency (e.g., hypovitaminosis A) in the aetiology of Sjögren's disease, inquiry was made into the dietary history. In spite of the fact that the patient was a spinster, living alone and doing part-time domestic work, this was good, and there was no evidence to support the existence of any gross primary nutritional error preceding the onset of the glandular swelling, or, indeed, prior to 1948. During that year, however, the

xerostomia had made chewing and swallowing difficult and she had had to keep largely to liquids. It was therefore considered that the xerostomia had resulted in the nutritional deficiency syndrome of the second stage. The addition of iron and vitamin B complex greatly improved these deficiency features. Iron and riboflavin appear to be the main deficient factors. The picture was very similar to that of the Plummer-Vinson syndrome, though it occurred at a later age period than is usual with the latter condition, which tends to improve spontaneously after the menopause.

It has been suggested (Stannus, 1940; Pollak, 1945) that iron deficiency may condition disturbance in the metabolism of riboflavin and allied factors; the haemin derivatives cytochrome and cytochrome oxidase are linked in tissue respiration with enzyme systems containing riboflavin and nicotinic acid, and riboflavin may be a weak link in the chain. The cause of the nutritional disturbance in this case seems to be the dysphagia secondary to the xerostomia, whereas in the usual younger case of Plummer-Vinson syndrome other factors (e.g., demands of pregnancy, lactation, and menstruation) are responsible. It is interesting to note that in what has been referred to as the second stage of this illness the dysphagia became intensified and was associated with the sensation of food sticking behind the larynx—a feature which is characteristic of the Plummer-Vinson syndrome. The dysphagia in the latter condition seems to result from B-complex (particularly riboflavin) deficiency and does not appear to be the primary factor in the pathogenesis of the condition.

As has also been noted by other observers (e.g., Ellman and Weber, 1949), the glandular enlargement was unaffected by vitamin A therapy. It would seem that the cause is not to be looked for in nutritional deficiency, but, in view of the often striking psychological disturbances which these patients show, investigation from the psychosomatic angle and attention to the aspect of autonomic imbalance may be profitable.

REFERENCES

- Ellman, P., and Weber, F. Parkes (1949). *British Medical Journal*, 1, 304.
Pollak, H. (1945). *Brit. J. Ophthalm.*, 29, 288.
Sjögren, H. (1933). *Acta ophthalm.*, Kbh., Suppl. 2, 1.
Stannus, H. S. (1940). *Lancet*, 1, 352.
Weber, F. Parkes (1945). *Brit. J. Ophthalm.*, 29, 299.

Medical Memoranda

Herpes Gestationis with Hydatidiform Mole and Chorion Epithelioma

Herpes gestationis has been described by Dühring as a condition which differs from dermatitis herpetiformis only in so far as pregnancy is the exciting cause. It is a fairly uncommon condition, and associated with hydatidiform mole and chorion epithelioma is an extreme rarity. Elliott (1938) describes an extensive bullous eruption associated with chorion epithelioma and metastases. A further case is reported by Whitfield, Smith, and Manson (1946).

CASE REPORT

The patient, a woman aged 42, had had two previous pregnancies, in 1934 and 1936, and one miscarriage in 1946. In August, 1948, she consulted Mr. Keith Vartan on account of menorrhagia, and was admitted to hospital on October 21. The Friedman test was then found to be positive. Loss of blood continued, and she was treated by transfusion. On

November 5 a hydatidiform mole was removed from the uterus. Penicillin was given, and on November 12 she was found to have developed a generalized herpetiform eruption, mostly on the limbs and around the waist. The lesions consisted of circular red patches most of which, after a day or two, developed into vesicles.

On November 26 dilatation and curettage were performed, but only one or two small vesicles were obtained in the curettings. On December 2 a Friedman test was negative. I saw the patient on December 17, when the clinical picture was typical of herpes gestationis. She was transferred to St. Nicholas' Hospital on December 22. In view of the stress laid on the danger of chorion epithelioma developing in these cases, it was thought essential that another Friedman test should be performed, but unfortunately the patient discharged herself at Christmas and was not heard of again until some weeks later, when she had been admitted to another hospital on account of recurrence of uterine bleeding. She was there found to have a soft, large uterus, which was curetted, several small vesicles, typical of hydatidiform mole, being obtained.

Histologically, a differentiation between hydatidiform mole and chorion epithelioma could not be made. The Friedman test was positive, and it was therefore decided to remove her uterus, which was found to be moderately enlarged with a thick firm wall. The uterus was filled with a nodular semi-cystic growth which had a mottled haemorrhagic appearance. Macroscopically, it suggested a mole which was invading the uterine wall. The histologist decided that the condition should be regarded as a chorion epithelioma. A Friedman test taken a fortnight after operation was still positive. There was still evidence of a rash when the patient was admitted to the third hospital, and this persisted without change during her stay there. Shortly before her discharge from a fourth hospital, to which she had been transferred for x-ray treatment, it was observed that the rash had cleared up, but that there was residual pigmentation of the skin.

I wish to thank Mr. Keith Vartan, Dr. E. J. Newton, Mr. M. N. Rankin, and Dr. T. S. Leedham for the information they have given me about this patient.

W. G. TILLMAN, M.D., M.R.C.P.,
Dermatologist, St. Nicholas' Hospital, Plumstead, S.E.18.

REFERENCES

- Elliott, Joseph A. (1938). *Arch. Derm. Syph., Chicago*, 37, 219.
Whitfield, J. M., Smith, Leroy, and Manson, R. Campbell (1946). *Va med. Mon.*, 73, 257.

Primary Septicaemia and Meningitis Due to *Salm. Enteritidis*

A basal meningitis due to *Salmonella enteritidis* has been reported on a number of occasions. Smith (1921), Lynch and Shelburne (1930), Stevenson and Wills (1933), and Schulz-Schmidtborn (1934) each report one case. Huang *et al.* (1937) report two cases associated with relapsing fever. The ages of these cases vary from 5 weeks to 30 years.

Salm. enteritidis septicaemia, as noted by other observers, usually occurs in association with a debilitating disease. The following case illustrates this point, a severe degree of anaemia being present for the greater part of a year previously and the patient suffering from a low-grade pyrexia for three months prior to death. The septicaemia was primary and there were no intestinal symptoms until 48 hours before death. Until a positive blood culture was obtained an interesting sequence of events occurred to obscure the diagnosis.

CASE REPORT

The patient, a girl aged 17, underwent splenectomy for an idiopathic haemolytic anaemia in December, 1948. After the operation she developed, first, a thrombosis in the deep veins