TABLE II

	Test Mixtures Incubated at 4° C. for 1 Hour, the Red Cells then Washed Three Times at 37° C.	Coombs Test on Tile
1	5 drops 5% cell suspension in saline	—
2	5 drops 5% cell suspension in saline + 5 drops fresh serum	+
3	5 drops 5% cell suspension in saline + 5 drops serum after incubation at 56° C. for 5 minutes	—
4	5 drops 5% cell suspension in saline made from clotted sample after 1 hour at 4° C.	—

The results of these further tests led us to believe that the patient's serum contained a cold incomplete auto-antibody, which caused the positive Coombs reactions on the patient's cells through the clotted blood samples' being chilled in a refrigerator. Moreover, it seems that the potency of both the cold auto-agglutinin and the incomplete auto-antibody was increased during the period of anaemia, when the erythropoietic system was deficient in the liver factor. This deficiency was also associated with positive Wassermann and Kahn reactions by the serum. The patient's history contains no suggestion of a syphilitic infection, and we can offer no explanation of these findings except to postulate that, as the liver-factor deficiency in some way caused an increased production of a non-specific haem-agglutinin and a presumably non-specific incomplete red-cell antibody, it also caused the production of a non-specific antibody capable of giving positive Wassermann and Kahn reactions. As these latter reactions became negative with no treatment except liver injections, we feel they should be regarded as false. We are aware of a comparable case (Rubinstein, 1948)—one of acute haemolytic anaemia with transient positive Wassermann reactions during a haemolytic crisis. In cases of primary atypical or virus pneumonia the serum often contains strong cold auto-agglutinins and occasionally gives transient positive syphilitic serological reactions (Florman and Weiss, 1945). It seems reasonable to expect that factors causing a rise in titre of cold auto-agglutinins would cause a parallel increase in the cold incomplete auto-antibodies, and so one might find such a combination of serological reactions in a case of virus pneumonia as we found in our case. But the clinical picture of our case at no time suggested a virus pneumonia, and the diagnosis remains as pernicious anaemia in relapse. The patient has now felt perfectly fit and the blood has remained normal for the past six months with maintenance liver injections.

Finally, we wish to draw attention to the added complication of a direct Coombs reaction on the red cells of a patient with suspected acquired haemolytic anaemia, in that one must now ensure that a positive result is not due to cold incomplete auto-antibodies in the patient's serum.

Summary

Details are given of a case of pernicious anaemia in relapse, in which the serum gave false positive Wassermann and Kahn reactions and contained a cold incomplete auto-antibody causing the red cells to give a positive Coombs reaction.

We are grateful to Dr. W. M. Chapman, of Shaftesbury, for allowing us to publish details of his case; to Drs. P. L. Mollison and J. V. Dacie, Postgraduate Medical School of London, for their assistance; and to Dr. C. N. Partington, of Dorchester, for the results of his investigations.

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A CASE OF PAINFUL SWELLING OF THE HAND

BY

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There is no intention to suggest, even by implication, that the surgical procedures described below constitute a cure for more than a very occasional case of poly-arthritis or of neuritis of the upper limb; but so dramatic a release from pain and swelling and so rapid a return of function are worthy of record.

Case History

The patient, a woman aged 56, had been vigorous and healthy before 1941, living in the cold dry air of the north-east of England. In 1941, in an air raid, she received a severe emotional shock, and from this time her general health began to deteriorate. In 1943 she felt tingling sensations in the fingers of both hands. The pain was intermittent and worse at night. A few months after its onset the nocturnal pain became so severe that she was unable to sleep. Her hands were always red and, in her own words, began to look old. She felt as if two heavy weights were tied to her wrists. Because of the swelling of her hands she was advised to become a blood donor, and during the next few years she gave six blood donations. Repeated venesection did not give relief. Very soon the inability to get a good night's rest sapped her general health, and as a result she became emotional and psychoneurosis was suspected.

Within a few months of the onset of the illness the pain had travelled to the palms of the hands, up the arms to the shoulder-joints, and across the front of the chest and back. By 1946 she found it impossible to lie on either side while in bed, the slightest pressure against her shoulders being intolerable. No special position gave relief. From 1946 to 1948 the condition slowly worsened, and by 1948 both her hands were swollen and painful. As the swelling and the pain in the right hand increased, the condition in the left hand improved, and was resolved by June, 1949. Full investigations had been carried out in another hospital, and the blood sedimentation rate was found to be normal on two occasions.

When I saw her in October, 1949, the whole of her right arm was swollen, the shape of the hand being as shown in Fig. 1. Movement at any joint from the shoulder downwards

FIG. 1.—The periarticular swelling of the hand is clearly shown.

FIG. 2.—Radiograph showing osteoporosis of bones of the right hand.

caused severe pain. A diffuse swelling in the right supraclavicular region was more easily seen than felt. There was no objective sensory loss in the skin of the limb. The left arm was normal, and evidence of disease was not found elsewhere in the body or in the central nervous system. Temperature, pulse, urine, blood urea, and blood cell counts were normal. The Wassermann reaction was negative. The radiological report (by Dr. S. Josephs) read: "There is a marked osteoporosis of the bones of the right hand, with fusiform soft-tissue swellings of the fingers. The appearance is that seen in rheumatoid arthritis, but may be accounted for by disuse. No evidence of cervical disk or cervical rib." (Fig. 2.)

Diagnosis

Rheumatoid arthritis had already been diagnosed by an orthopaedic surgeon and by an experienced physician, and Dr. C. E. Kellett, a physician colleague of mine, had also thought the same. However, as the case was not absolutely typical of rheumatoid arthritis I felt that there should be taken into account in the differential diagnosis acroparaesthesia, compression of the root or trunks of the brachial plexus, obstruction of the subclavian vein, psychoneurosis, neuralgic amyotrophy, angioneurotic oedema, and toxic neuritis.

The discomforts of acroparaesthesia are usually rapidly and completely relieved by rest and by simple medical measures. Also, persistent swelling of the hand is unusual in this condition. In cases of compression of the roots of the brachial plexus there is almost invariably evidence of dermatome sensory loss and of dermatome muscle wasting. "Freezing" of the shoulder may obscure the diagnosis, but fixity of the fingers is unknown in cases of, say, a cervical herniated nucleus pulposus. It is true that the swelling in the arm and hand in the present case may have obscured muscle-wasting, but the overall picture was very little like that usually found in cases of root compression. Persistent swelling of the hand and arm is also very rare in the so-called costoclavicular syndrome. In the two cases of obstruction of the subclavian vein that I have seen the swelling of the hand was generalized and there was no pain. There was no evidence of a psychoneurosis, and I have never seen a case of severely painful angioneurotic oedema. Neuralgic amyotrophy is followed by extreme wasting of the muscles of the arm; none occurred in this case. Toxic brachial neuritis is a very painful condition and is associated with tenderness and with oedema, which may be due merely to the patient holding his arm still or to interference with the neurovascular mechanism.

On the clinical evidence alone I think it was impossible in this case to come to a precise diagnosis, for, though on the whole it favoured rheumatoid arthritis, the condition of toxic brachial neuritis could not be excluded with certainty.

Operation

I decided to explore the right supraclavicular triangle and cut the cervico-thoracic sympathetic. The pathological findings and surgical procedures were:

The hibernating pad of fat was found to be unusually thick. As a rule this mass of fat can easily be swept away from the deep tissues by means of a swab with a minimum of bleeding. On this occasion the fat had to be removed by sharp dissection. Histological examination of the fat revealed no abnormality.

The veins of the longus cervicis group were distended with dark-blue blood and appeared varicose. This congestion was due to local factors, since oxygenation of the general blood stream was satisfactory, as shown by the pinkness of the patient's skin and the colour of the blood in the more superficial layers of the wound. The main radicles of the longus cervicis veins were ligated and the distended venules removed. Histologically, nothing abnormal was found in the walls of the excised veins.

The upper two trunks of the brachial plexus were abnormally red and were matted here and there with sticky adhesions to the surrounding tissues: these adhesions were severed. The scalenus anterior muscle was divided to facilitate exposure of the stellate ganglion.

The sympathetic pathways were excised from the lower border of the stellate ganglion above to the chain between the third and fourth thoracic ganglia below. The white rami to the stellate ganglion from the first thoracic root were preserved, and so a post-operative Horner's syndrome was avoided. Histological examination of the tissue removed proved it to be sympathetic ganglion and chain showing no pathological abnormalities.

The next morning all pain had disappeared from the hand and arm; there was active movement at all joints, limited only by the periartritic thickening. Within 24 hours it was obvious that the swelling of the hand was subsiding; within three weeks the whole of the swelling had disappeared and there was a complete return of function in the hand and arm. There has been no sign of recurrence of symptoms up to date.

Discussion

What may have brought about the relief in this case? By scalenectomy and the removal of the supraclavicular pad of fat and of the longus cervicis group of veins possible impairment of the venous drainage of the arm may have been relieved. Neurolysis of the trunks of the brachial plexus is difficult to evaluate, but the findings of perineural adhesions point to the presence of an inflammation. By sympathectomy alone it is possible to relieve pain in certain conditions, the mechanism of the relief being largely one of conjecture. Also, sympathectomy increases the arterial blood supply to the limb.

Unfortunately in this case it is impossible to decide which of the three surgical manoeuvres produced the cure, or whether it depended on a combination of two or three. These cases, in my experience, are rare. Should any other surgeon meet with this type of case and decide to operate when known medical measures

have been tried and failed I should like to suggest that cervico-thoracic sympathectomy be carried out by the posterior route.

Finally, it would be interesting to know whether sympathectomy would influence the action of "compound E" when it is given for peri-arthritis of the upper limb.

This case was referred to me by Dr. J. Hope Poole, and I thank him for his help and detailed observations; Dr. Raymond Whitehead has supervised the presentation of this paper, and I am grateful for his criticism; and much is owing to Miss Norma Harvey.

Medical Memoranda

Spontaneous Subarachnoid Haemorrhage in Children

As very few reports have been published of spontaneous subarachnoid haemorrhage in children from rupture of intracranial aneurysms, three such cases are presented. It is remarkable that more cases of haemorrhage in children have not been described in view of the congenital origin of the aneurysms. Miller (1938) reviewed the literature and refers to Ramsay's case aged 10, Barker's fatal case aged 5 months, and two others at 11 years, and describes five further cases at ages 2, 3, 10, 11, and 12 years. Other cases have been published. Forbus (1930) describes a case aged 8 years. Jefferson (1947), in a series of cases presenting with ocular palsy, includes one at 13 years and one at 14 years, and mentions a baby of 9 months which he states is one of the youngest on record. In a series of 81 cases of subarachnoid haemorrhage Taylor and Whitfield (1936) had only one in a child, aged 10 years, and in Beadles's (1907) series of 555 cases there is no mention of a child. Thus it would appear that rupture of a berry aneurysm is a rare accident in children.

Case Reports

Case 1.—A boy of 12 was seen in 1936 with the classical syndrome of prostrating headache and deepening coma, signs of meningeal irritation, and heavily blood-stained C.S.F. There was no history of trauma or undue strain, and no evidence of any other congenital abnormality. The boy recovered, but had a residual hemianopia. When last seen in 1939 he was an apprentice in an engineering works.

Case 2.—A girl aged 8½ came to the children's out-patient department on February 16, 1949, with a history of vomiting six days previously. Three days later she had been giddy and had fallen, and the next day she was again very sick, after which she was noticed to have weakness of the left side of the body. On examination she had a left hemiparesis with a doubtful extensor plantar response. The fundi were normal and no other abnormal signs were present. A tentative diagnosis of intracranial tumour was made and arrangements for her early admission to hospital were completed. On the day of admission, February 18, she vomited repeatedly, became febrile, and on examination had a tender stiff neck, a positive Kernig's sign, left-sided weakness, and a left extensor plantar response. The disks were abnormally pink and a little swollen, and there were a few scattered retinal haemorrhages. Other systems were normal. She was restless and irritable, with periods of drowsiness. Lumbar puncture showed fluid under increased

pressure, with a normal Queckenstedt test. The C.S.F. was uniformly and heavily blood-stained, and the supernatant layer was xanthochromic. Later reports showed the fluid to be sterile and to have a negative Wassermann reaction.

The next day the patient complained of headache, which was relieved by the removal of about 10 ml. of C.S.F. The fluid looked just as blood-stained as on the previous day. The neurological signs were unchanged. From then she improved steadily and it was soon impossible to restrain her activity. There was considerable recovery of power in the paralysed arm and leg. After three weeks physiotherapy was started and she was discharged from hospital on May 15, 1949.

In June, 1950, her general condition was good and she was attending school. Unless she became excited or tried to hurry there was little disability. On examination she then had some weakness of the left arm and leg and an up-going toe on that side. The fundi were normal. At that time there had been nothing to suggest that any further bleeding had taken place.

Case 3.—A boy aged 9 was admitted to hospital in coma on October 2, 1948. Since an attack of bronchopneumonia at the age of 2 he had had recurrent respiratory infections and had had curious turns which may have been attacks of petit mal. For two years these attacks, which occurred at intervals of many months, had been accompanied by occipital headache and vomiting. On the morning of October 2 his mother had heard him cry out and found him on the floor, struggling aimlessly. He had become drowsy and at times unconscious. The family doctor saw him six hours later and referred him to hospital. When the boy was admitted he could be roused from his light coma, but was restless and resentful of examination. His face was turned to the left, there were no voluntary movements of the left arm or leg, and he had a slow, coarse nystagmus. The pupils were small and equal. The fundi were normal. There was moderate neck stiffness and a positive Kernig's sign. No other abnormality was found on routine clinical examination. Lumbar puncture was done and showed a blood-stained C.S.F. On standing, the supernatant fluid was yellow. The fluid was sterile. The following day he was very restless and the left hemiplegia was marked. The left plantar response was extensor. Lumbar puncture was repeated and the fluid found blood-stained as before. He then became delirious and his general condition appeared to be deteriorating rapidly. On October 7 there was slight papilloedema, left hemiparesis with an up-going toe, and left hemi-anaesthesia. The appearance of the C.S.F. was unchanged. He remained restless and noisy, swallowing little, and with a stiff neck and positive Kernig's sign until the beginning of November. The C.S.F. was examined at intervals and remained blood-stained. On November 5, although there was still some neck stiffness, he was started on physiotherapy because of commencing contractures in the left arm and leg.

During the next two months the boy made steady progress, and after recovering from an acute attack of tonsillitis he was eventually discharged from hospital on April 20, 1949. He attended for out-patient physiotherapy for some time and treatment was then stopped. When seen in June, 1950, he had minimal weakness of the left arm and leg, and an extensor plantar response on that side. He had remained well, and his mother thought that his general condition was much better than before his illness in October, 1948. He had had no further headaches or petit-mal attacks.

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