Medical Memoranda

A Case of Pseudocyesis

A woman of 41 complained of "peculiar feelings" in the abdomen, pain in the breasts, and morning sickness. She had not menstruated for eight months. The abdomen "felt tight," and she said it was much larger than usual. Two successive pregnancy tests were negative, and she was shown the results; her comment was: "I question the tests. They may be wrong. I think it's an eight-months pregnancy." The sensations in her abdomen were described as "like a mouse."

She had married in 1947, and was most eager to have a child. No contraceptives were employed, yet she did not conceive. In 1953 her husband had an accident and went to hospital. This was the first separation in seven years of marriage; the patient was very disturbed by it, and very glad when he was discharged. "It was marvellous. Felt different. More response at intercourse." From this time forward, she had breast pains, felt sick in the mornings, noticed veins on the breasts, with some discharge, and thought herself heavier and bigger in the abdomen. She was quite convinced that these signs indicated a pregnancy. The urinary gonadotrophins were considerably raised.

About a week after her attendance at the clinic she experienced abdominal pain and had a bout of diarrhoea. "I felt something had burst, something to do with the baby." She felt better, and had more energy. The abdomen was "still fat, but not so hard. I felt the baby did start and now it's finished. Something passed away." Six weeks later she said: "I'm better now. I was certain about the pregnancy, but am satisfied now it wasn't." Her own explanation of the episode was: "Perhaps the shock of the accident altered the glands."

The intercourse with her husband which occurred after his discharge from hospital was more gratifying, and more intensely experienced, than it had ever been, and the patient then felt that at long last a child would be conceived. Her later history seems to show that the desire to conceive, if it is strong enough, can produce the subjective sensations of pregnancy, and some objective physical signs as well, even an alteration in endocrine balance. Her belief in the pregnancy remained unshaken by the negative tests, and persisted until the attack of diarrhoea, which signified to her that the "foetus" had been passed out.

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Acute Osteomyelitis of the Maxilla in Infancy with Cavernous Sinus Thrombosis

Acute osteomyelitis of the superior maxilla in infancy is more common than is generally believed, athough only 21 cases have been described in the English and American literature since 1928. The usual aetiological agent is *Staphylococcus aureus,* penicillin-resistant strains of which have been encountered with increasing frequency of late, especially in hospitals and maternity units (Gairdner, 1954). Treatment with penicillin, although it has been successful in the past (Jacoby and Sagorin, 1945; Haworth, 1947; Macbeth, 1951), may therefore no longer be effective. McCash and Rowe (1953) have already reported two cases of osteomyelitis of the superior maxilla in 4-weeks-old infants caused by penicillin-resistant strains of *Staph. aureus*; in these cases there was further deterioration in the clinical condition after starting penicillin treatment. Both cases required surgical intervention and then eventually responded to treatment with chlortetracycline and streptomycin.

A case of osteomyelitis of the maxilla caused by penicillin-resistant strains of *Staph. aureus* and complicated by cavernous sinus thrombosis is described below. This patient received penicillin for four days before the antibiotic was changed to chlortetracycline and streptomycin, to which the organisms isolated from the pus were sensitive *in vitro.* By this time, however, the infection had spread to the cavernous sinus.

CASE REPORT

The patient, the elder of twins, was born in hospital by breech delivery. Her birth weight was 5 lb. 9 oz. (2.5 kg.). She developed a "mild right-sided ophthalmia" on the third day of life. This was successfully treated with oral sulphadimidine, and she was discharged home aged 17 days. Four days after discharge she presented with swelling of the left side of the face associated with general fretfulness. Cellulitis was diagnosed and treatment with benzathine penicillin ("penidural") by mouth was begun (1,200,000 units daily in four divided doses). There was no improvement, and three days later she was admitted to hospital.

On admission there was an inflammatory swelling involving the left side of the face, nose, and hard palate. Pus was discharging from the gum margin in the left canine region. A diagnosis of osteomyelitis of the maxilla was made.

Crystalline penicillin, 2,000,000 units, was administered intramuscularly in divided doses during the next 24 hours, but the temperature continued to rise and the child vomited frequently. Both eyelids became progressively more oedematous, accompanied by increasing bilateral proptosis. Cultures from the pus grew two separate strains of *Staph. aureus,* resistant to penicillin but sensitive to streptomycin in both instances, and in one also to chlortetracycline and chloramphenicol. The treatment was therefore changed to oral chlortetracycline, 150 mg., and intramuscular streptomycin, 200 mg. daily in divided doses. This was continued for three weeks. Although the swelling of the face receded, and the amount of discharge of pus into the mouth decreased, the ocular signs became more pronounced, indicating thrombosis of the cavernous sinus. Gross oedema of both eyelids was present, with marked bilateral proptosis, diminution of the ocular movements, engorgement of the retinal veins, and slight bulging of the fontanelle.

Treatment with intravenous heparin was instituted as follows: A continuous intravenous drip infusion was begun and 500 units of heparin injected as an initial dose, followed by 250 units after four hours; the total amount given four-hourly was increased by 250 units each time. Thus after 48 hours the dose injected was 2,000 units and the treatment with heparin was discontinued. The clotting-time was estimated twice daily, and at the end of treatment was 13 minutes. The swelling of the eyelids disappeared during the period of heparin administration, but some bilateral proptosis and thrombosis of the right supraorbital vein persisted for another eight days.

After this the progress towards recovery appeared to be satisfactory until, three weeks after the onset of initial symptoms, the oedema of the right eyelids and right-sided ocular proptosis recurred. This was accompanied by a rise in temperature. The antibiotic treatment was changed from chlortetracycline to chloramphenicol, 350 mg. daily in divided doses for two weeks. As anticoagulant, oral phenylindanedione ("dindevan") was used instead of heparin, 20–30 mg. daily in two divided doses being given for 10 days. This dose was effective in maintaining the prothrombin index in the region of 50%. The response to treatment was good. The temperature became normal after 24 hours, the oedema disappeared after 24 hours, and the proptosis of the right eye receded after a week.

Three weeks later proptosis in the left eye recurred. This was followed after four days by an area of inflammation below the inner canthus of the left eye. Another course of phenylindanedione was given for 17 days together with chlortetracycline, of which 250 mg. daily, in divided doses, was administered for three weeks. The inflammatory lesion resolved. The patient was discharged fit and well two months after admission.

Six weeks later, however, she was readmitted with a small hard swelling on the left cheek and oedema of both eyelids. She was again treated with oral phenylindanedione, 30 mg. daily for six days, followed by 20 mg. daily for six days, and chlortetracycline 250 mg. daily in divided doses, which was continued for three weeks. Progress to recovery was uneventful.

Radiological evidence of sequestrum formation could not be demonstrated at any time.

The patient was followed up for six months after her final discharge, during which time she remained well.

COMMENT

The combination of oedema of both eyelids, bilateral proptosis, diminution of the ocular movements, engorgement of the retinal veins, and bulging of the fontanelle made it reasonable to assume that cavernous sinus thrombosis had occurred. Treatment was therefore based on this assumption.

The effectiveness of combining antibiotic and anticoagulant treatment in the majority of adult cases of cavernous sinus thrombosis is well established (Lillie, 1951 ; Shaw, 1952). There appears to be no previous reference to this condition in early infancy.

The case described illustrates the importance of basing the antibiotic treatment on bacteriological sensitivity tests, especially in regard to staphylococcal infections which have been acquired in hospital, where penicillin-resistant strains are likely to be encountered.

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Agranulocytosis from Carbimazole

The initial reports on the use of carbimazole (" neomercazole " ; 2-carbethoxythio-1-methylglyoxaline) in the treatment of thyrotoxicosis suggested that the drug might be free from toxicity in therapeutic dosage (Poate, 1953 ; Doniach, 1953). Further experience has confirmed the clinical efficacy of carbimazole but has revealed that toxic effects occur in a small percentage of treated patients. Painful or swollen joints, pyrexia, and skin eruptions have been described, but the most important complication is agranulocytosis, of which four examples have been published. As carbimazole is a relatively new drug, it is felt that the following account of a fifth case of agranulocytosis is worthy of record.

In the case of agranulocytosis reported by Fraser *et al.* (1954) the patient had previously had severe skin and joint lesions when given methimazole (" mercazole " ; 2-mercapto-1-methylglyoxaline). Recovery ensued when the carbimazole was discontinued, as occurred in the two cases described by Bartels (1953) and Harrison (1954), but in that described by Richardson *et al.* (1954) carbimazole produced a complete and fatal marrow aplasia.

CASE REPORT

A married woman aged 70 was admitted to Bromley Hospital on July 21, 1954, for the investigation of lassitude, diarrhoea, and loss of weight. Examination revealed a nodular goitre of moderate size, a fine tremor of the fingers, and moist palms. The basal metabolic rate was +30% and the sleeping pulse was persistently raised above 120 a minute. Further investigations disclosed no abnormality other than some diverticulosis of the sigmoid colon. The blood count showed a haemoglobin of 90% and a white-cell count of 3,000 per c.mm., with 1,770 neutrophils. The blood cholesterol was 170 mg. per 100 ml. The body weight was 64.5 kg.

In view of the patient's age and the moderate degree of her symptoms, it was decided to treat the thyrotoxicosis with an antithyroid drug. Carbimazole, 30 mg. daily in three divided doses, was started on July 31. After ten days of treatment there was undoubted improvement in her symptoms and signs. In particular the sleeping pulse, which had hitherto been uninfluenced by bed rest alone, fell to a normal level. She was discharged from hospital on August 16, when the dose of carbimazole was reduced to 15 mg. daily, and arrangements were made to follow her progress as an out-patient.

Fourteen days after discharge the patient sent for her practitioner, Dr. R. Colby, of West Wickham, who found her ill with a fever of 103° F. (39.4° C.). Carbimazole was discontinued, penicillin was given, and a blood count confirmed the suspected diagnosis of agranulocytosis ; haemoglobin, 92% ; white cells, 2,000 (neutrophils 140, lymphocytes 1,640, metamyelocytes 160, myelocytes 60). The patient was readmitted to hospital, where she ran a fever of 102° F. (38.9° C.) for the first two days. The tonsils and fauces were inflamed and the submandibular glands were enlarged. Penicillin therapy was continued, and within five days the sore throat and general condition of the patient had greatly improved and the temperature had subsided to normal. The white-cell count showed a progressive rise, and on the sixth day after admission was 5,000 (2,300 neutrophils, 1,600 metamyelocytes, lymphocytes 900, basophils 200 per c.mm.). Examination of the sternal marrow on the same day was reported to show the typical picture of the maturation arrest seen in drug reactions. The haemoglobin percentage and platelet count remained normal throughout her illness and the bleeding-time was not prolonged. She made a complete clinical recovery.

COMMENT

The published reports on the use of carbimazole, apart from that of Bartels (1953), suggest that in therapeutic dosage it is the least toxic antithyroid agent yet available (see Fraser *et al.*, 1954). A second advantage of the drug is that the degree of thyroid gland enlargement that accompanies its use is less than with the thiouracil derivatives. In a series reported by Kirkeby and Rømcke (1955) the thyroid gland increased in size in 34% of the cases treated with methylthiouracil or propylthiouracil, compared with 7% in those treated with carbimazole. A third advantage, claimed by Poate (1953), is that the thyroid gland is less vascular after carbimazole therapy than after the thiouracil drugs.

The results of further studies on the use of carbimazole are awaited before its place in the treatment of thyrotoxicosis can finally be assessed. Meanwhile the present case report is a further reminder that the use of this relatively safe and effective antithyroid drug is not devoid of risk.

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