

higher are usually accepted as evidence of toxoplasma infection. It cannot, however, be claimed that the positive serological tests are absolute proof that the heart condition in the cases we have reported was due to toxoplasmosis. Nevertheless we suggest that the circumstantial evidence is very strong. Case 1's attacks may have been embolic from an intraventricular thrombus, but the c.s.f. and mental state suggested encephalitis. In addition there were definite fundal changes. The unexplained heart-failure and sudden death in a young mother and brother and sister, together with splenomegaly in two of them, makes an unusual story, and the necropsy report from Dr. Lavertine at the Whittington Hospital would fit the histological picture described in previous cases where toxoplasmas have been found in the myocardium.

Sabin et al. (1952) maintain that in the congenital form of toxoplasmosis no more than one sibling is ever involved. In a previous communication (Hart et al. 1951) one of us (J. W. P.) reported two successive siblings affected. Campbell (1953) reported a family in which the mother and two children appeared to have had a chronic form of toxoplasmosis for several years, in which maculopapular rashes, joint pains, conjunctival injection, and headaches were a feature. All three were shown to have meningo-encephalitis, nerve-deafness, swollen discs, or optic atrophy. The infection persisted for several years without killing the patients. The mother died recently and was found to have an abscess in the spleen. Campbell and other workers suggest that pregnancy may cause a latent chronic toxoplasmosis to flare up. It certainly seems likely that this happened with case 1's sister, and the whole family history appears to support the idea of infection present for perhaps twenty years. Hitherto it has been thought that the congenital form of toxoplasmosis results from infection of the mother during pregnancy. This view may have to be revised, and a more serious view taken of a disease which, though widespread, has been thought in the main to be benign. Earlier diagnosis and a wider appreciation of the protean manifestations, ranging from repeated abortion to mental backwardness and headaches, seem to be required and, above all, effective treatment.

Pinkerton and Henderson (1941) emphasise the fact that toxoplasmas were only found in the myocardium after a most prolonged search, and in a case described by Gilmore et al. (1942) and at first thought to be sarcosporidiosis only three pseudocysts were found in 220 sections of heart-muscle. The organism has often been found more easily, but it is as well to recognise that diagnosis remains difficult and, with culture methods still unreliable, will at present have to depend in the main on serological tests; if these are positive in obscure myocarditis with any consistency, it should amount to proof that toxoplasmosis is the cause. The possibility that we are dealing with a strain of toxoplasma with a special affinity for the myocardium may have to be considered.

The fact that very low standards of hygiene and of nutrition were present in the early childhood of case 1's family may give food for thought. It is tempting to think that the state of nutrition may influence the host's chances of overcoming toxoplasmosis as other illnesses, notably tuberculosis and malaria, and that low standards of hygiene will probably encourage cross-infection among human beings.

#### Summary

Focal myocarditis due to toxoplasmosis has been reported several times as part of the generalised acquired disease. It has not received recognition as a single presenting manifestation.

We suggest that toxoplasmosis should be excluded in all obscure forms of myocarditis, endomyocardial fibrosis, familial cardiomegaly, and idiopathic cardiac hypertrophy.

Three cases of myocarditis are described which are believed to be due to toxoplasmosis. Positive serological tests, abnormal electrocardiograms, and frank heart-disease were present in other members of two of these patients' families, which would normally have been regarded as instances of familial cardiomegaly.

The cases are believed to be examples of chronic toxoplasmosis, probably acquired rather than congenital.

We are much indebted to Dr. I. A. B. Cathie for the serological investigations, Dr. A. L. Jacobs and Dr. J. D. O'D. Lavertine for details of case 1's sister, and Mr. Dawson, the coroner for Ipswich, for reports of two of the persons who died some time ago.

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## EPILEPSY WITH FETISHISM RELIEVED BY TEMPORAL LOBECTOMY

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THE term "affect epilepsy" was first used by Bratz (1906) to indicate certain brief emotional outbursts which he considered to be epileptic. Two decades later "affective epilepsy" was applied to those reflex epilepsies in which the exciting agent is of emotional significance (Wilson 1928). The case described below should properly be placed within Kinnier Wilson's grouping because the epileptogenic stimulus was a sexually charged object or fetish, which in this instance was a safety-pin. It is of interest not only because of the unusual association of this fetish with temporal-lobe epilepsy but also because an anterior temporal lobectomy relieved both the epilepsy and the fetishism.

#### Case-report

For as long as he could remember, a man, aged 38, enjoyed what he described as a "thought satisfaction" when looking at a safety-pin. This was highly pleasurable and he sought the seclusion of the lavatory to indulge it, since he felt, even as a child, that this was an odd and potentially embarrassing habit. At some time between the ages of 8 and 11 years the "thought satisfaction" began to be followed by a blank period; but, since the phenomenon was kept secret, the first witness to it was his wife, who accidentally observed him, aged 23, holding a safety-pin and oblivious of his environment.

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During the next few years she observed more of these attacks, and the pattern became clearer. The patient would stare at the pin for a minute and then appear glassy-eyed. Next he would make a humming noise for a few seconds and for a few further seconds sucking movements with his lips. Finally he stood immobile and unresponsive for two minutes. Recovery was then immediate. Just before some of his attacks his right pupil would dilate. By the age of 31 the immobility was followed by marking time and later by marching backwards while his right hand plucked at his left sleeve. Brief postictal confusion became evident, during which he usually proceeded with the task at hand but occasionally dressed himself in his wife's clothing.

Every fit was produced by a safety-pin and was therefore largely voluntary. A "bright shiny" whole pin was essential, and often several of them were more effective than one. Before his marriage the point of the pin need not have been protected by a hood, and from the age of 14 until 17 scissors were added as a less desirable stimulus. Aside from these minor variations the trigger was specific. Fantasies and dreams of (or accidental exposure to) a safety-pin were adequate as well.

He never injured himself during an attack although he occasionally tripped and fell when marching backwards. His employment entailed maintenance work at considerable heights, but he never fell. He was never incontinent or significantly cyanosed. Initially the seizures were a month apart but by the time of our observation they occurred in groups of two or three every 7-10 days. In the intervening period the sight of a safety-pin was innocuous. Sexual stimulation and anxiety-producing situations were the most common states in which the desire to look at a pin arose. Most frequently the fits occurred soon after awakening when, with a full bladder, adult sexual outlets were sought but refused by a frigid wife. At other times erections were not concomitant, but occasionally during either sexual intercourse or masturbation he had a fit if he fantasied his fetish.

During the last year the patient had been psychotic for 2 or 3 days on three occasions. The first two episodes definitely followed fits, but the last one probably developed without a seizure on the night before his operation was first scheduled. On each occasion he was grandiose, paranoid, and loudly religious. In one attack he claimed that he was related to the King and had discovered a cure for cancer. In the last episode he stated that he received messages from God, that he had a special mission, and that the operation was inadvisable. As evidence of the validity of his claims he showed us two pictures. One was an erotic illustration of the Decameron, and the other was a magazine cover of a small boy surrounded by more than 70 objects spilled from his mother's purse. Among these objects were two very small safety-pins, one open and one closed.

*Personal History.*—The patient is the only son of an over-affectionate mother and a distant and older irresponsible father. The mother fostered an extraordinarily close relationship with her son and brought him up to be the antithesis of his father. He became a model effeminate child and youth and has maintained the deep attachment to his mother throughout his life. His birth and physical development were normal. There was no history of a head injury, nor was there an epileptic or psychotic family history. After an average schooling he was apprenticed to a carpenter and joiner and has conscientiously worked at this trade ever since. He married at the age of 21 and has one healthy child aged 7 years. He had psoriasis for some years. He has vivid memories of collecting and playing with "bright shiny safety-pins" as a very young child. These were always in his pocket in great

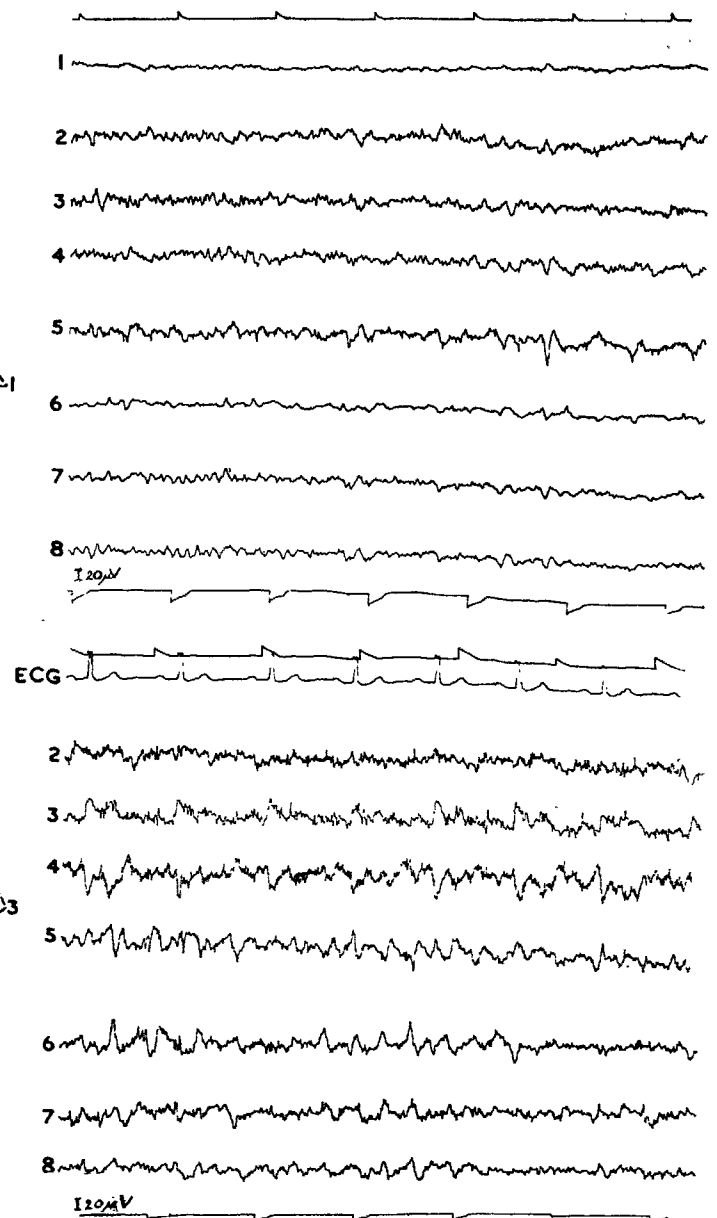
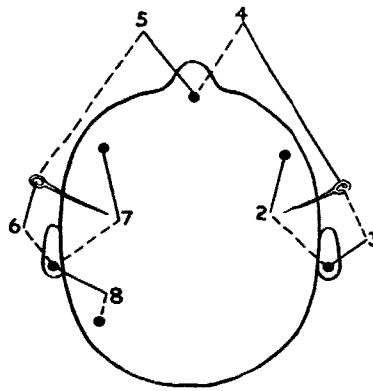
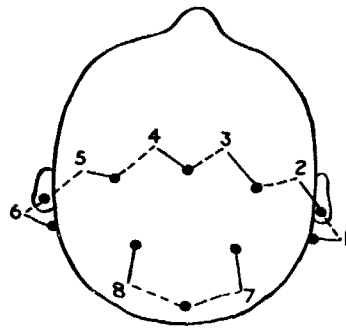


Fig. 1.—Routine electro-encephalogram from convexity of head (above) shows occasional sharp waves from left temporal electrode (channels 5 and 6). In the basal recording (below) similar but more frequent firing is seen at the left sphenoidal electrode (channels 5 and 6), the discharge spreading to the electrodes over the convexity—e.g., channels 7 and 8.

numbers, and his favourite game was to join them in a chain and pull it along the floor. He connected them with napkins and with his mother, and clearly remembers seeing one in her discarded underclothes. The "thought satisfaction" was considered the greatest experience of his life—"better than sexual intercourse." Other aspects of his sexual history are auto-eroticism, adolescent homosexual mutual masturbation, and voyeuristic tendencies, with emphasis on women's breasts. During the earlier years of the marriage sexual intercourse had been performed satisfactorily about twice monthly. In the last five years he had become increasingly impotent, claiming that the safety-pin had replaced his need for a genital outlet. He had always been overtly passive, narrow, conscientious, preoccupied with money, and childlike in speech and deportment. His goal was to be a model husband and father.

*Previous Treatment.*—Before coming to this hospital he had been given supportive psychotherapy, one hour monthly (1942-46). Extensive notes taken at the first interviews, for which we are indebted to Dr. J. C. Smyth, conclusively rule out suggestion as an important factor in elaborating the history. He undertook this treatment when he was conscripted, his object being to gain exemption. All the usual anticonvulsants had been used without benefit. A left antitemporal focus was found on routine electro-encephalography, and he was referred to one of us (D. H.) by Dr. R. W. Tibbetts, who realised the rôle of the epilepsy. He was admitted to Maudsley Hospital on Jan. 2, 1953.

*On examination* he was intelligent, effeminate, passive, and narcissistic. Preoccupation with his problems of sexuality and epilepsy hindered any substantial rapport, but contact

was not impaired by paranoid or aggressive mechanisms. Physical examination revealed no evidence of disease other than extensive psoriasis. Neurological examination, including speech and visual field, was negative. The patient was right-handed. His blood-pressure was 130/75 mm. Hg and Wassermann reaction negative. Blood-counts, cerebrospinal fluid, straight skull radiographs, and a left carotid arteriogram were normal. Electro-encephalography and a lumbar pneumo-encephalogram, however, gave significant findings.

*Interictal Electro-encephalography.*—Routine records (pharyngeal, sphenoidal, thiopentone sodium) showed a clear spike focus at the left basal electrode spreading to the left convexity (fig. 1).

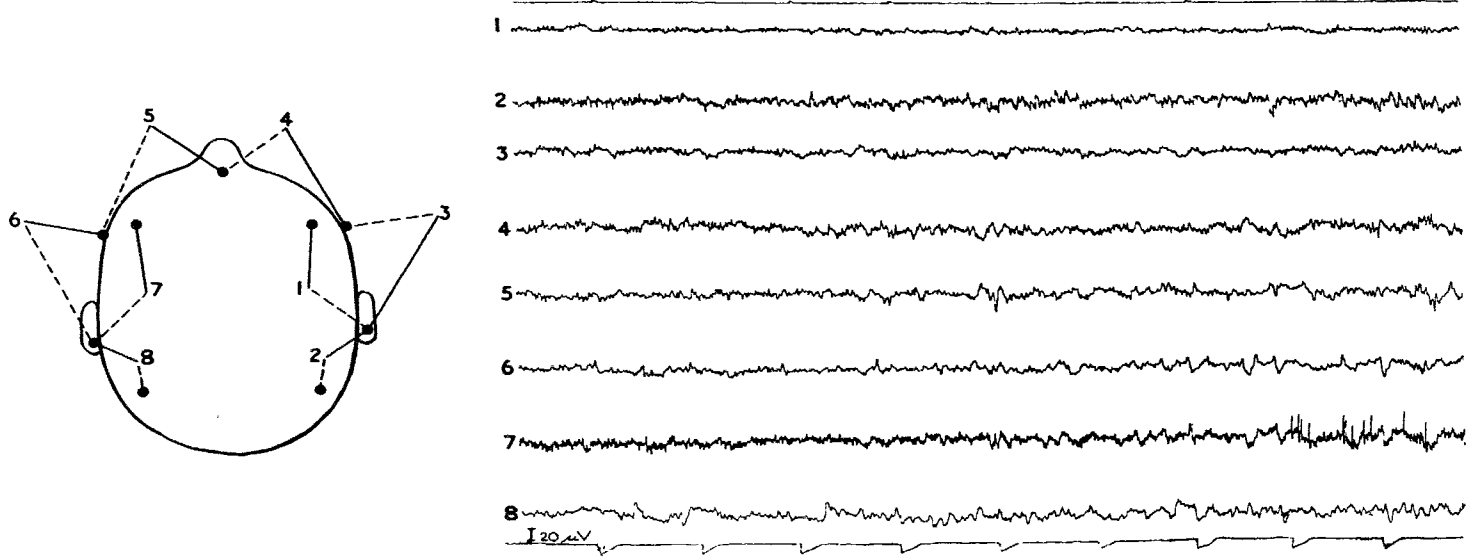
*Ictal Electro-encephalography.*—Simultaneous records were made by electro-encephalography, electrocardiography, and measurement of the palmar skin resistance as well as by two leads on the zygomatic arches. The exhibition of a safety-pin produced a rise in the pulse-rate from 70 to 80. A minute later the exhibition of four safety-pins further raised the pulse-rate to 90, lowered the skin resistance, and produced frequent firing, best seen from the scalp electrodes over the left frontotemporal region. A clinical fit occurred 30–40 seconds later, followed by irregular fluctuating high-voltage slow activity, which was bilateral at first, but became confined to the left side, where it was focal in the fronto-temporal region (fig. 2).

*Pneumo-encephalography* revealed a focal dilatation of the temporal horn of the left lateral ventricle, although the remainder of the ventricular system appeared normal (fig. 3).

*Operation.*—On March 17, 1953, one of us did a left lateral craniotomy under local anaesthesia. No macroscopic abnor-

malities were seen in the exposed temporal lobe, but electro-corticography revealed epileptic discharges in the anterior temporal region. The temporal lobe was then resected, the extirpation including the anterior 2 cm. of the superior temporal gyrus and thence passing obliquely to a point on the inferior border of the lobe 7 cm. behind the temporal pole. The uncus and the anterior part of the hippocampus were included in the specimen (see Falconer 1953). Intravenous thiopentone sodium was used during the later stages of the operation.

*Histological Findings.*—Prof. A. Meyer reported as follows: “Macroscopic inspection of the specimen did not reveal any important changes. The length of the specimen was 7 cm. and the greatest width 6 cm. No gross atrophy or other significant abnormality was seen after transverse dissection into six blocks, of which three (nos. 1, 3, and 5) have been embedded in celloidin and investigated in serial sections. Sections were stained by cresylviolet, Heidenhain’s myelin stain, van Gieson-haematoxylin, and Mallory’s phosphotungstic acid haematoxylin. (The Holzer method was used but proved, as this is often the case in biopsy specimens, unsuccessful.) In block 1 (region near the pole) the meninges and blood-vessel walls were slightly thickened, the latter particularly in the white matter. There was moderate to considerable marginal gliosis. No focal loss of cells was seen in the cortex, but the number of macroglial nuclei was definitely increased in the upper layers of the cortex. The white matter also showed increased glial cellularity. Similar changes were seen in blocks 3 and 5. The Ammon’s horn was investigated in block 5. It showed no typical sclerosis of the Sommer sector, but there was a slight reduction of nerve-



RESTING RECORD BEFORE PRESENTATION OF SAFETY-PIN

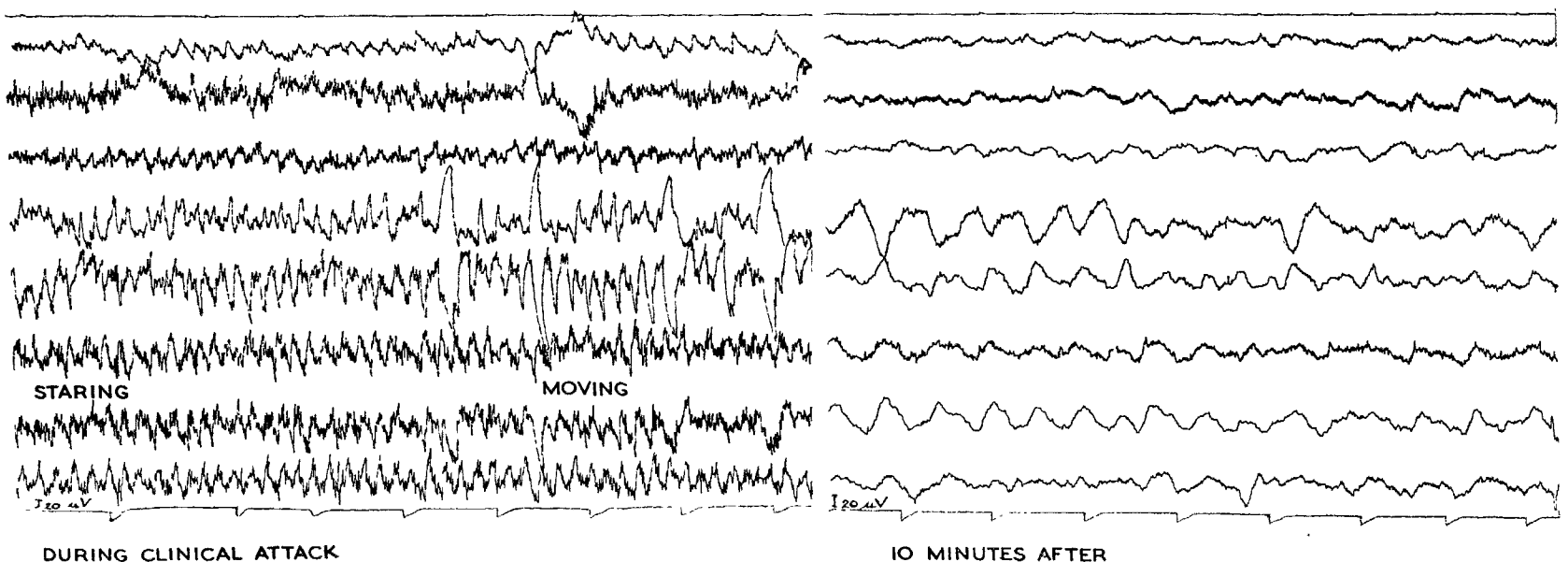


Fig. 2—Electro-encephalogram taken before and during a clinical seizure induced by the presentation of a safety-pin.

cells in the end-plate (h5), and the macroglial nuclei were in excess. Measurements of the width of the cortex were taken in the hippocampal, fusiform, and inferior temporal gyri and compared with normal temporal gyri. The figures obtained were slightly, but probably significantly, below the minimal width in the normal

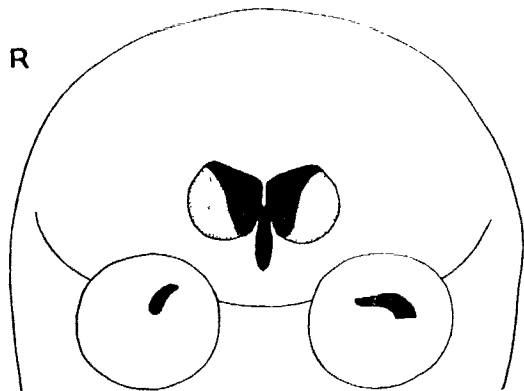


Fig. 3—Tracing of anteroposterior pneumo-encephalogram shows dilated left temporal horn projected within outline of left orbit.

controls. There is thus slight atrophy of cortex and white matter, with compensatory gliosis in all convolutions available for examination. The changes in the Ammon's horn were slight. No sclerosis of the Sommer sector was found."

*Follow-up.*—The patient was readmitted to hospital for further study 16 months after his operation. In the meantime he had no further fits, had no desire to look at a safety-pin, and had become as potent as in early marriage. He had taken small doses of phenobarbitone irregularly. In contrast to his preoperative state he seemed much more mature and spoke of his past disabilities objectively. During the year he had worked harder than ever before. The only other evidence of personality change was that he passed mucus per rectum when involved in irritating situations with his wife or work-mates. He had gained almost a stone in weight, slept well, and had a good appetite. His relationship to his wife had improved. Mr. V. Meyer reported that the patient's intellectual abilities, measured by the Wechsler tests, remained unchanged from before operation, but there was some decrease in his ability to learn and retain new verbal material. However, the patient did not complain of any learning difficulties.

*Postoperative Electro-encephalography.*—Routine and sphenoidal records were made fourteen months after operation. The general characters were normal, and the exhibition of a safety-pin did not change the record, although the pulse-rate rose from 54 to 72 in anticipation, falling to 60 after the exposure was begun. No firing took place after the intravenous injection of thiopentone sodium 0.5 g., but on the waking record an occasional low-voltage spike and some fast activity were observed in the left mid-temporal areas posterior to the operation site.

### Discussion

It is well known that epilepsy of temporal-lobe origin is commonly associated with personality disorder and with psychosis (Gibbs et al. 1948, Hill 1953). In such cases distressing emotional tension or behaviour disorder may precede and lead up to a seizure which can temporarily relieve the abnormal psychological state. Occasionally the morbid preictal mental state includes preoccupations with specific emotions and memories, but hitherto no case has been published in which a desire for a fetish object and gratification of the desire have led to an epileptic seizure. In this respect the case described is, we believe, unique. There can be no doubt, however, that intense emotion can precipitate seizures, but this is usually sympathetic (Wilson 1928) rather than fearful, anxious, or erotic. Moreover precipitating emotions are usually unpleasant—not, as in this case, pleasant and sought after. Occasionally a specific but meaningful stimulus, such as music (Critchley 1937), acts as the only and invariable trigger for epilepsy, and the rôle of emotion in these cases is undoubted (Shaw and Hill 1947). In yet other cases of epilepsy starting in childhood an occasion of severe emotional trauma precedes, sometimes by months, the onset of the disorder (Gowers 1881). More recently a study of temporal-lobe epilepsy has shown that the memory of distressing circumstances which preceded the onset of the epilepsy may be built, as it were, within the pattern of the seizure

and are then re-experienced as an ideational-perceptual aura—a hallucinated dream-like state which can be subsequently recalled (Hill 1953).

None of these phenomena can be precisely identified in the present case. Rather it seems that a perverse form of erotic gratification had developed in early childhood in this patient long before the first epileptic seizure. The fetish is unusual, but the relationship between this type of perversion and character disorder is well known, and the patient's passive effeminate personality is in keeping with the immaturity of his psychological development. Psycho-analytical studies of fetishism suggest that the object is chosen because of its sexual symbolic significance, from an otherwise innocuous memory of the fifth or sixth year of life (Freud 1928). These memories fall into the category of the very persistent "screen" or "concealing" memories commonly experienced by normal people and useful as a disguise for something less acceptable (Freud 1914). In a study of the ideational-perceptual auras of temporal-lobe epilepsy unpleasant "screen" memories have occasionally been noted, but none in which the memory was experienced with pleasure (Hill and Mitchell 1953).

The mechanism whereby the gratification of the fetish-need either became attached to the mechanism for the onset of the epileptic seizure or became the immediate precipitant thereof must, of course, remain obscure. Much the same applies to musicogenic epilepsy when intense emotion precedes the fit. Since, however, a seizure never occurred during marital sexual intercourse unless a safety-pin was fantasied, the direct physiological results of coitus and the physiological accompaniments of erotic excitement per se can be excluded as ætiological factors. The combination of a certain emotional state, mental set, and ideational content (the fetish) were necessary, and in this patient, as in musicogenic epilepsy, the operation of a conditioned reflex may be postulated (Shaw and Hill 1947). The similarity to musicogenic epilepsy is seen in the fact that sufferers from this condition are invariably musical and music lovers (Critchley 1937) but usually give up their musical activities when this causes them to suffer epilepsy. Such patients, however, are not compelled by the force of instinctual drives as in the present case.

The relief of temporal-lobe epilepsy by surgery has been previously reported, notably by Penfield and Flanigin (1950), Bailey and Gibbs (1951), and Green et al. (1951) in North America and by Falconer (1953) in this country. It seems, both from published reports and from our own experience, that the best results are obtained in those patients in whom a strictly unilateral and single electrographic focus is found at operation to be associated with a discrete pathological lesion lying within the resected part of the temporal lobe. No macroscopic lesion was found in this patient, but histological changes were observed subsequently. It is attractive to consider that these changes were causally related to the seizures, especially since the epilepsy improved after their removal. Possibly the changes may have been secondary to the epilepsy, but this patient had never had a major convulsion nor been significantly cyanosed during his seizures. The cessation of seizures after temporal lobectomy was anticipated from our experience with other cases of temporal-lobe epilepsy, and surgery was recommended to prevent not only further epilepsy but also further psychotic episodes and consequent social deterioration. However, the excellent result of surgery in the present case, assessed a year after the operation, has included, greatly to our surprise, relief from the fetishism. This patient no longer feels the need to obtain gratification from the viewing or fantasizing of safety-pins, and with this change have come a turning of his libidinal interest towards his wife and an increase of potency and well-being.

### Summary

A case of temporal-lobe epilepsy is described in which the viewing of a fetish object precipitated seizures and became the invariable trigger.

Relief not only of the epilepsy but also of the fetishism followed temporal lobectomy.

The relationship between the epilepsy and the fetishism is discussed.

We wish to thank Dr. R. W. Tibbetts for kindly referring this case and for the very helpful information he had obtained; Prof. A. Meyer for the pathological report, Dr. G. Pampiglione for the electrocorticographic studies, Dr. R. D. Hoare for the radiological examination, and Mr. V. Meyer, B.A., for the psychological testing.

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## LONG-TERM OXYTETRACYCLINE (TERRAMYCIN) THERAPY IN ADVANCED CHRONIC RESPIRATORY INFECTIONS

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MANY chronic infections of the lung benefit from short courses of antibiotics, both during acute episodes, when they may be life-saving, and also when the chronic symptoms are severe. The improvement is, however, usually short-lived. More prolonged therapy may be essential in the treatment of lung abscess (Sutherland and Grant 1950) and chronic suppurative pneumonia (Nicholson 1952) and is successful in most of these cases, the infection often being entirely eliminated and not recurring. The problem in bronchitis, infective asthma, and bronchiectasis is different, since, when the treatment of whatever length has been stopped, the infection is almost certain to become re-established sooner or later. It is therefore only by repeated short courses or continued treatment that it may be possible to prevent relapses.

Franklin and Garrod (1953) used chloramphenicol for periods of up to five months in children with bronchiectasis with encouraging results, but abandoned this method of treatment when one patient died of aplastic anaemia. McVay and Sprunt (1953) treated 21 patients for an average of eleven months with 0.25 g. chlortetracycline (aureomycin) twice a day and compared the results with a control series of 9 patients treated with placebo tablets. They found a reduction of respiratory infections of more than 50% in the antibiotic group as compared with the controls.

The development of resistant organisms and the danger of toxic drug reactions have been the main reasons that this form of treatment has been so little used in

the past. The present series was started following the excellent response of case A to continued oxytetracycline.

### Clinical Material and Method

The cases selected were all advanced with considerable persistent disability. The assumption that the condition was infective was founded in the first instance on the macroscopic appearance of the sputum rather than the bacteriological findings. The dosage scheme of oxytetracycline was modified as the series progressed, but all patients started on at least 2 g. daily in three or four divided doses until the infection had been apparently eliminated or suppressed, when the dose was reduced to a level which was found sufficient to maintain the response; the usual maintenance dose was 1-1.5 g. in two or three doses. Recently the initial dosage used has been 3 g. daily given as 1 g. eight-hourly. The assessment of response was made on the patients' general condition and symptoms, the quantity and quality of the sputum, and results of bacteriological and cytological investigation. All patients who responded favourably have remained on treatment to the present time, providing the improvement has been maintained and unpleasant side-effects have not intervened.

### Bacteriological Techniques

In order to avoid sampling errors due to irregular distribution of organisms in the sputum (May 1953a), all cultures were made after preliminary homogenisation of sputum specimens by pancreatin (Rawlins 1953), and, in an attempt to avoid failure to isolate some organisms owing to their irregular appearance in different sputum samples (May 1953a), at least three specimens from each patient were examined where possible before therapy was begun.

The quantity of pus in sputum specimens was estimated partly by macroscopic inspection and partly by cell counting (Rawlins 1954). Films of the liquefied sputum were stained with hæmatoxylin and eosin in order to distinguish between true pus and "pus" composed of eosinophils (Rawlins 1954, May 1954).

### Results

TABLE I—RESULTS OF TREATMENT OF 38 PATIENTS WITH OXYTETRACYCLINE

Diagnosis	No.	Good early response	Remaining on oxytetracycline with benefit
Chronic staphylococcal bronchopneumonia	1	1	1 (30 months)
Bronchitis .. ..	17	15	9 (7-20 months)
Infective asthma ..	13	2	0
Bronchiectasis ..	7	6	4 (6-8 months)
Total .. ..	38	24	14

### Chronic Staphylococcal Bronchopneumonia

**Case A.**—A 4-year-old girl had had a cough for a year with several pyrexial attacks which had been treated with penicillin, sulphonamides, and antral wash-outs. Her parents had noticed that her faeces were pale and bulky and that she did not tolerate fats well. On admission to hospital she was extremely ill, cyanosed, dyspnoeic, and febrile. There were coarse crepitations throughout both lungs, and radiography of the chest showed diffuse coarse mottling and patchy consolidation. The sputum was purulent and *Staphylococcus aureus* was persistently cultured from it. The results of investigations of faeces for estimation of pancreatic activity were indefinite. The patient was treated in an oxygen tent with penicillin, streptomycin, and sulphonamides, followed by chloramphenicol. She did not respond to penicillin and the sputum culture contained resistant organisms. She improved for a short time on streptomycin and chloramphenicol, but the organisms rapidly became resistant and she again became very ill. She was then started on oxytetracycline 0.5 g. six-hourly. On the fourth day she began to improve progressively,