

A CASE OF GENERALIZED STREPTOTRICHOSIS WITH EXTENSIVE LESIONS IN THE CENTRAL NERVOUS SYSTEM.

BY W. W. C. TOPLEY, M.B.CANTAB., M.R.C.P.

Clinical Pathologist and Bacteriologist to Charing Cross Hospital; late Demonstrator of Morbid Anatomy at St. Thomas's Hospital.

THE case recorded here derives its interest mainly from the lesions found in the brain and meninges, streptotrichial infection in this region being an occurrence of some rarity. Howard [11], writing in 1903, finds only eighteen cases in the literature. A case reported by Delépine [7] in 1889 is not included in Howard's list, which gives details of all the eighteen cases, and from which many of the following particulars have been obtained, and since 1903 two further cases have been described, one by McDonald [13] in 1904, and one by Beevor and Buzzard [4] in 1903.

If we include the case under consideration the total number of recorded cases is twenty-two.

Considering these in a little more detail we find that in five cases the lesions in the central nervous system were described as primary, that is, no further lesions of a streptotrichial nature were discovered, while in the remaining seventeen cases lesions of a similar nature were found elsewhere in the body.

The lesions found in the central nervous system consisted of cerebral abscesses, almost always multiple, and meningitis.

In thirteen cases cerebral abscesses were present without meningitis.

In three cases the meninges were affected without abscess formation.

In six cases both abscesses and meningitis were present.

The abscess formation observed in these cases shows considerable similarity of character, and need not be further noticed here. With regard to those cases in which the meninges were affected, however, the lesions seem to have differed considerably.

Thus in a case recorded by Almquist [2], on the true nature of which Howard throws considerable doubt, the lesion found was a cerebrospinal meningitis.

Dolore [8] records a case in which a cerebrospinal pachymeningitis of streptotrichial origin was the only lesion present.

In Beevor and Buzzard's [4] case a basal leptomeningitis was present, and this was the type of meningeal lesion found in the present case, and in the remaining cases described, excluding a case described by Ponfick [14] in which the lesion commenced extracranially and perforated the skull, producing a localized meningitis and abscess formation.

Batten [3], writing on meningitis, says: "Streptothrix infection of the meninges is a rare condition. It is most commonly found as an extension from a focus in the skull or soft parts of the face and throat, but some cases of primary infection have been recorded. Sometimes true metastatic foci have been found in the meninges secondary to infection in other parts of the body."

On examining the nine cases mentioned above, however, we find that in two cases only has infection of the cranial bones been noted, namely, one case reported by Ponfick [14] and alluded to above, and the case described by Beevor and Buzzard [4] in which caries of the sphenoid was found.

In Almquist's [2] doubtful case the meningeal lesion was considered as primary, while in the remaining six cases the meningitis was part of a more general infection.

Thus in Dolore's [8] case the original lesion was one affecting the neck and face.

In Chiari's [6] two cases the brain lesions were secondary to a streptotrichial bronchiectasis. In Eppinger's [9] case the bronchial and supraclavicular glands were primarily affected. In the case described by Keller [12] an abscess in the thoracic wall was the only other lesion present, while in the case under consideration the liver and lungs were the seat of streptotrichosis.

Indeed, if we extend the list to include the remaining thirteen cases, in which the meninges were unaffected, we find no further mention of disease of the cranial bones, so that metastatic infection would seem to be the rule in these cases.

In this connexion certain facts observed in the above cases are of some interest. Thus, dealing with the microscopical features of the organisms present in the twenty-two cases, in seven cases clubs and rosettes are mentioned as being absent or very rare. In only one case, that described by Bollinger [5], were typical colonies with club formation definitely stated to be present, while in the case under con-

sideration very definite club formation was present in the colonies in the central nervous system.

Délépine [7], in describing the lesions present in the case recorded by him says: "In a few vessels bacilli were also found, but it was not possible to determine whether they were adventitious products or not. They looked, however, not unlike segments of the more typical filaments found in the abscesses, and were probably early forms of the organism." No such appearances could be observed in the present case.

In the majority of cases in which an accurate description of the organism is given it is described as consisting of a few branching thread forms, sometimes in twisted masses, usually showing little beading and with a marked absence of club formation.

Dealing briefly with the relative frequency of streptotrichosis of the central nervous system as compared with a similar condition in other parts of the body the following figures taken from an article by Acland [1] may be given. Duvan, writing in 1902, and quoted by Acland, gives the following details of 257 cases:—

Face and neck	70	Central nervous system	..	19
Thorax	65	Generative organs	..	19
Liver..	40	Æsophagus	..	7

In the same article Acland gives the following table showing the situations of the lesions in 109 cases of streptotrichosis occurring in the United Kingdom between the years 1884 and 1906:—

Abdomen (including liver)	..	68	Brain and spinal cord	..	5
Face, neck, tongue, &c.	...	50	Acute general	..	9
Chest..	..	29			

The case under immediate consideration will now be described, and finally a few special points in connection with it considered.

While in the hospital the patient was under the care of Dr. Hawkins, and later of Mr. Makins, and it is through their kindness that I am enabled to record it here.

The patient, a man, aged 45, by trade a horsekeeper, was admitted to St. Thomas's Hospital on January 31, 1911, with a large tender swelling in the lower part of the abdomen, and gave the following history.

On January 7 he first noticed a feeling of tenderness over the lower part of the abdomen. No swelling was then present. A few days afterwards a small hard swelling appeared, which gradually increased in size. The tenderness and pain continued, and were increased after a heavy meal or on coughing.

The patient stated that he had never vomited, but that he had always suffered from a slight tendency to constipation.

Inquiries as to his family history yielded no information of importance. The only facts of interest in his past history were as follows:

He contracted syphilis at sixteen years of age. The chancre was followed by a rash, but no further lesions developed.

From 1886 to 1903 patient served in the Army, and from 1886 to 1889 was stationed in India, where he had repeated malarial attacks.

He had lost "about a stone" in weight during the six months preceding admission.

The following was the state on admission:

The heart, lungs, and nervous system were all found to be normal. The urine showed nothing abnormal.

A tumour could be seen and felt, occupying the hypogastric region, and extending slightly into the umbilical and left iliac regions.

The percussion note over the tumour was impaired; the tumour possessed well-defined edges; it did not pulsate; no fluctuation could be made out, and the skin was nowhere adherent over it. The swelling increased in size, and a week after admission the skin over its most prominent part became adherent, and fluctuation was felt.

On the eighth day the patient, who had been running an irregular temperature, varying between normal and 102° F., had a rigor, and the temperature rose to 103.5° F.

The following day an incision was made into the swelling, and about half a pint of foul-smelling pus of a yellow colour was removed, and the cavity drained.

The pus was examined in the Clinical Laboratory, and returned as sterile.

About the same time the patient's serum was tested for the Wassermann reaction with a positive result.

The wound continued to discharge for seven weeks, during which time the patient's condition showed little change, and then, on April 1, he had another rigor, the temperature then returning to the slightly irregular course, varying between 97.5° F. and 100° F., which it had followed since the first operation.

The blood was examined for malarial parasites without result. A count of the white-cells revealed a leucocytosis of 16,600 per c.mm.

On April 24 an exploratory laparotomy was performed. The appendix was found behind the cæcum. It was about 5 in. long, and showed signs of old disease, but there was no obvious connexion between it and the abscess in the anterior abdominal wall. It was removed.

This operation was followed by a temporary improvement in the condition of the patient, but on April 12 he had another rigor, and the temperature continued of a pyæmic type until June 6, accompanied by frequent rigors.

On May 15 the percussion note over the base of the left lung was noted as impaired, and a few crepitations were audible in this region. These signs gradually extended, and eventually involved the greater part of both lungs.

A frequent cough developed with expectoration, and an examination of the sputum revealed the presence of elastic fibres.

A blood culture taken about the same time was found to be sterile.

From June 6 to June 23 the temperature remained at a lower level, never rising above 101° F.; but on June 17 patient had a series of fits beginning in each case with clonic movements of the left arm and leg, which then became generalized, and were succeeded by a period of rigidity. The patient became noticeably cyanosed during these attacks, and on several occasions bit his tongue. Unconsciousness usually lasted for ten to fifteen minutes, and after regaining consciousness the patient usually slept for several hours.

These attacks were repeated on June 18 and 19.

On July 3 paralysis was first noticed affecting the left arm and leg, and more marked in the former.

Both knee-jerks were slightly exaggerated.

The right planter reflex was extensor, the left indefinite. On the same date the patient was noted to be suffering from a left hæmianopia.

Two days later incontinence of urine and fæces developed, the paresis became more profound, and the patient died on July 8. Frequent rigors occurred from June 23 to the time of death.

POST-MORTEM EXAMINATION.

The *post-mortem* examination revealed the following conditions. Scars indicating the incision into the abscess and the laparotomy were present. The lower end of the former scar still showed a red granulomatous area.

On opening the abdominal cavity the intestines immediately adjacent to the laparotomy scar were found to be adherent to the abdominal wall, but the peritoneal cavity appeared otherwise healthy.

The liver.—Throughout the upper part of the right lobe were scattered 12 to 20 small white nodules varying in size from about $\frac{1}{2}$ in. in diameter down to the size of small shot. The smaller of these were of comparatively firm consistence, but the larger were breaking down in the centre, and on pressure yellowish pus exuded. No granules were present.

The lungs were riddled throughout with small shotty nodules having about the same range of size as those seen in the liver, but the smaller considerably predominating. All the larger and most of the smaller nodules yielded on pressure pus of the same type as that found in the liver, and here again no granules could be seen.

The brain.—The base was covered with thick yellowish pus over an area extending from the under surface of the cerebellum posteriorly to the optic chiasma anteriorly. The pus did not reach to the lower

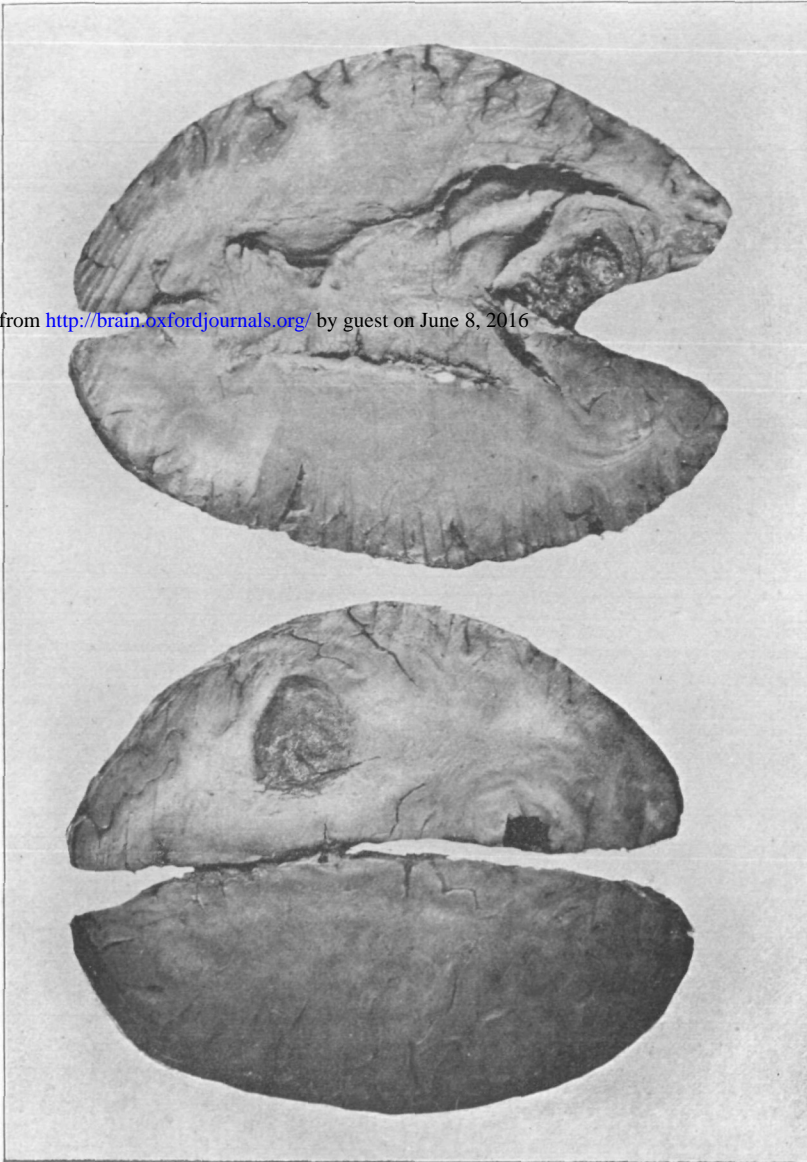


FIG. 1.—Showing a large single abscess lying beneath the right ascending frontal convolution. A small portion of brain-tissue has been removed from the right occipital lobe for microscopical examination.

FIG. 2.—Showing a smaller more irregular abscess situated in the right occipital lobe just anterior to the calcarine fissure.

part of the medulla, or upper part of the cervical cord ; the remainder of the cord was not removed.

The obvious deformity of the brain revealed the presence of extensive suppuration within its substance ; one large abscess indeed burst during examination, but only a minute quantity of pus escaped.

The brain was then hardened for twenty-one days.

The spleen was large, dark red in colour, and of soft consistence.

The appendix had been amputated.

The kidneys, heart, intestines, and remaining organs showed no naked-eye changes.

After hardening, the brain was further examined, with the result that the following areas of suppuration were discovered (figs. 1 and 2).

(1) A large, single, irregular abscess-cavity lying deep to the upper half of the right ascending frontal convolution, right Rolandic fissure and right ascending parietal convolution and extending forwards beneath the hinder part of the superior and middle frontal convolutions.

In its lower part it lay altogether in the white matter, in its upper part the cortex itself was excavated, and in one place, in the upper half of the right ascending frontal convolution, only a very thin shell of cortex covered the cavity (figs. 3 and 4).

(2) A smaller, more irregular and somewhat loculated abscess situated in the right occipital lobe just anterior to the calcarine fissure, the cortex in the neighbourhood of which was extensively invaded.

(3) A very large, multilocular abscess, consisting of two large pockets and many smaller ones affecting the left frontal lobe, which was very largely destroyed. This cavity was much distended with pus, and the right frontal lobe was markedly displaced and compressed as a result of the enlargement of the opposite lobe. Scattered around this large abscess cavity were many small foci of suppuration, not directly connected with the main cavity.

The pus in all these abscesses was of the same type as that observed in the liver and lungs, and though granular debris was present no bodies of the "mustard seed" type could be observed. The walls of the cavities were ragged and showed no attempt at encapsulation. On opening the ventricles they were found full of a slightly turbid fluid, but sections taken from the various parts of their walls showed no evidence of extension of the disease in this direction.

Microscopical sections were prepared from the liver, lungs, cerebral tissue and meninges ; tissue being chosen in each case, which, to the naked eye, appeared to be in the earlier stages of the disease—i.e., the

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FIGS. 3 and 4.—Showing a large multilocular abscess situated in the left frontal lobe. The two main cavities communicated through the aperture seen in the right-hand figure. Note the extreme deformity of the whole right hemisphere, especially the frontal lobe.

sections passed through the smaller foci where breaking down of tissue and pus-formation was least advanced. The sections were stained by Gram's method in each case.

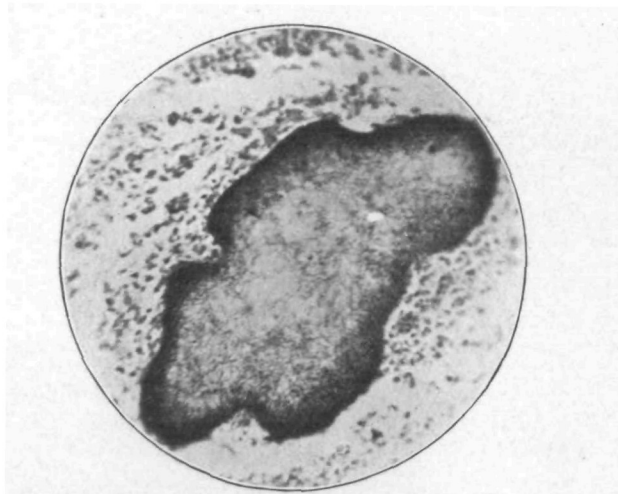


FIG. 5.—A streptothrix colony from a section obtained from the liver. Note the complete absence of radial clubs.

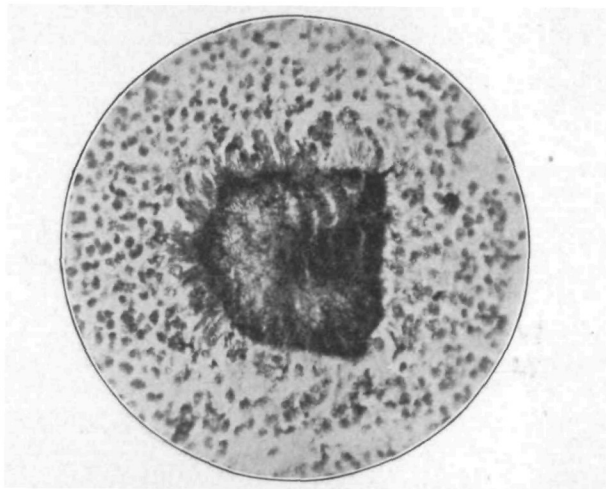


FIG. 6.—A streptothrix colony from a section obtained from the wall of a cerebral abscess. Note the well-marked radial clubs.

Typical colonies of a streptothrix were obtained in a certain number of sections from all the lesions.

A remarkable difference, well shown in the accompanying microphotographs, was observed between the colonies in the brain and those elsewhere. (Figs. 5 and 6.)

Whereas the colonies in the liver, for instance, showed the appearances usually found in human streptotrichosis, with little or no attempt at radial club formation, the colonies in the cerebral lesions showed the most marked radial club-formation, resembling that seen in the bovine disease, save that in no case did these clubs retain the stain by Gram's method.

The threads themselves showed marked beading in almost every case, and many coccus-like bodies were present in the majority of colonies.

The cellular reaction was largely of the mononuclear type, with a considerable admixture of polymorphonuclear cells in most cases.

An attempt was made to cultivate the organism from the pus from the liver abscess, but only a mixed staphylococcic growth resulted. The cultural characteristics could not, therefore, be made out, and it is impossible to relegate the organism to any special class.

One of the main points of interest in this case is, perhaps, the question of the original site of infection.

The appendix was found at the operation placed retro-cæcally, and was slightly thickened and fibrosed. The thickening was slight only, and there was no sign of any caseative or suppurative process, that is, the organ showed none of the lesions which are usually associated with streptothrix infection in this region. But Foulerton [10] says, "The tendency to natural cure must also be remembered as a possible explanation of cases in which streptotrichial lesions occur only in parts which would seem not liable to become the seat of a primary infection from without; thus in a single focus of infection in the liver it is always possible that there may have been a primary infection of some part of the intestinal canal, the initial lesion having meanwhile become healed."

It is thus possible that the condition of the appendix represented an old, healed streptotrichial lesion.

The next question is whether the abdominal abscess was of the same nature.

There are two facts which point in the opposite direction: (1) In the course of the examination of the pus from this abscess which was sterile on culture, the possibility of actinomycosis was thought of, and the organism searched for with negative results; (2) although streptotrichial lesions may heal under certain conditions, this is true, in most cases, only of slight lesions; and it is certainly exceptional for a streptotrichial lesion of the extent of the abscess noted to heal as completely as was the case in this patient.

The obvious alternative is that the abscess was in reality a breaking-down gumma, and additional weight is given to this view by the history of syphilitic infection and the presence of a positive Wassermann reaction.

Whatever the seat of the primary infection, there seems no doubt in this case that the final distribution occurred via the blood-stream, and was of the nature of a true pyæmia.

The reason of the marked radial club-formation is by no means obvious.

The cerebral lesions were certainly terminal in character, and it is improbable that they were of long standing. A clue is perhaps afforded by some further remarks of Foulerton's [10]; after pointing out that infection seems usually to be spread in the body by short rod segments which then branch and not by the spore forms, where further development would occur in the typical radial manner, he says, "In some circumstances, as when a suppurating cavity in the lung is in communication with the air, spore forms may be present in abundance, and being conveyed elsewhere may doubtless originate fresh foci of infection."

In this case the clinical findings make it almost certain that the pulmonary lesions preceded the cerebral.

CONCLUSIONS.

The case here recorded presented extensive lesions in the central nervous system, consisting of a basal lepto-meningitis, and multiple cerebral abscesses.

These lesions were proved to be streptotrichial in nature by the demonstration of colonies of the organism in microscopical sections.

There was no disease of the cranial bones or of the soft tissues of the head or neck, but extensive streptotrichial lesions were found elsewhere in the body. It is therefore clear that the cerebral lesions resulted in this case from an infection of a pyæmic type, and not as the result of extension from neighbouring parts, and a study of the literature would suggest that infection by the blood-stream is the rule in cases of streptotrichosis in this situation.

The colonies of the streptothrix found in the cerebral lesions showed certain marked differences from those found in the liver and lungs, and these differences may, perhaps, be accounted for by the different forms in which the infection may have been carried, in the one case as

true spore-forms from the pre-existing pulmonary lesions, in the other as short filaments, which would appear to be the more common method.

REFERENCES.

- [1] ACLAND. Article on Actinomycosis in Clifford Allbutt's "System of Medicine," Second Edition, 1906, vol. ii, part i, p. 324.
- [2] ALMQUIST. *Zeitschr. f. Hygiene u. Infektionskrankh.*, 1890, vol. viii, p. 189.
- [3] BATTEN. Article on Meningitis in Clifford Allbutt's "System of Medicine," Second Edition, 1910, vol. viii, p. 165.
- [4] BEEVOR and BUZZARD. *Trans. Path. Soc. Lond.*, 1903, vol. liv, p. 320.
- [5] BOLLINGER. *Münch. med. Wochenschr.*, 1887, vol. xxxiv, p. 789.
- [6] CHIARI. *Zeitschr. f. Heilk.*, 1900, xxi, Abth. Path. Anat., p. 351.
- [7] DELÉPINE. *Trans. Path. Soc. Lond.*, 1889, vol. xl, p. 420.
- [8] DOLORE. *Gaz. Hebdom. de Méd. et Chir.*, 1896, N.S., vol. i, p. 496.
- [9] EPPINGER. *Beitrag. zur Path. Anat. u. Allg. Path.*, 1891, vol. ix, p. 287.
- [10] FOULERTON. Article on Pathology of Streptothrix Infections in Clifford Allbutt's "System of Medicine," Second Edition, 1906, vol. ii, part i, p. 302.
- [11] HOWARD. *Trans. Assoc. Amer. Phys.*, 1903, vol. xviii, p. 427; also in *Journ. Med. Research*, 1903, vol. lx, p. 301.
- [12] KELLER. *Brit. Med. Journ.*, 1890, vol. i, p. 709.
- [13] McDONALD. *Scott. Med. and Surg. Journ.*, 1904, vol. xiv, p. 305.
- [14] PONFICK. "Actinomykose des Menschen," Berlin, 1882.