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OTOGENOUS NON-PURULENT ENCEPHALITIS*

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AMONG the otogenous intracranial complications, uncomplicated non-purulent encephalitis is the most recent of our clinical experiences in this region. *Borries* was the first to describe this morbid picture in 1921 as an independent clinical entity. As early as forty years ago *Oppenheim*, in some cases, found coincidence of encephalitis and otitis but he did not think that the otitis could be the agent responsible for the encephalitis. In 1900, however, he put forward the suggestion that it was quite possible ("durchaus denkbar") that cases of purulent otitis might be met with which, by hæmatogenous metastasis, bring about encephalitis. Subsequently this question was taken up by different authors, particularly by *F. Voss*, but none of them considered otogenous encephalitis to be an independent affection; it was, on the contrary, always considered a stage preliminary to a cerebral abscess. *Borries* put forward his view on the ground of three clinically proved cases of uncomplicated encephalitis recorded in the literature (two cases reported by *Voss*, one by *Wischnitz*), to which he added two cases observed by himself, convincingly pointing out that, besides the cases in which the otogenous encephalitis was a preliminary or abortive stage of abscess, there were also cases

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of hæmorrhagic encephalitis without any tendency to suppuration, analogous to hæmorrhagic encephalitis attending whooping-cough, scarlet fever, etc.

In a series of subsequent publications *Borries* explained the details of his point of view, proposing the subjoined new division of the intracranial complications of otitis, in which the non-purulent inflammation of the brain tissue, in contradistinction to previous classifications, finds its natural place as an independent disease.

- (1) Inflammations of the sinus.
- (2) Inflammations of the dura.
- (3) Inflammations of the leptomeninges.
- (4) Inflammations of the cerebral tissue.

The inflammations of the cerebral tissue may be either of isolated occurrence or combined with other intracranial complications, besides presenting various degrees of intensity. They are subdivided by *Borries* into the following :—

(a) Simple encephalitis, which is the most benign form, manifesting itself pathologo-anatomically chiefly as microscopically detectable perivascular leucocyte infiltrations. Probably it can be of isolated occurrence, but in the majority of cases it occurs as a complication of other intracranial inflammations.

(b) Hæmorrhagic encephalitis, representing a severer form than simple encephalitis, attended by both micro- and macroscopically visible hæmorrhagic infiltrations of the brain tissue. Frequently it occurs together with, or as a preliminary stage, of abscess, though in some cases it occurs as an autonomous disease, without tendency to suppuration.

(c) Purulent encephalitis = cerebral abscess. This frequently develops from hæmorrhagic encephalitis but it may also follow directly after simple encephalitis.

Since *Borries'* first report, reliable clinical observations of uncomplicated otogenous encephalitis have been published by several authors. Some of them will be briefly recorded.

Lund has described the following unusual case :—

A 16-year-old girl with left-sided otitis media and labyrinthitis after labyrinthectomy developed a high fever rising to 102-104° F. and persisting for a protracted period. Pulse rate 80-100. The cerebrospinal fluid was turbid, with numerous cells. Five weeks

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after the operation the patient developed symptoms from the left temporal region. No abscess was revealed by turning down the dura and incising the brain, but an encephalitis, apparently provoked by aerogenic, anaerobic bacteria was shown.

Wacker records the following case :—

A woman, æt. 32 years, who on account of cholesteatomatous otitis was submitted to left-sided radical operation, after a lapse of three weeks developed high fever and gradually progressive papillo-œdema and right-sided hemiplegia and hemianopsia. Puncture and incision of the brain was negative. Cerebrospinal fluid was normal but under slightly increased pressure. Apart from slight paraphasia, all the symptoms disappeared in the course of three months.

An acute case running a very violent course was reported by *Key Åberg*.

A 4-year-old child, two months after right-sided mastoidectomy developed acute left-sided hemiplegia with coma. The temperature was high ; the pulse rate 110. The patient was submitted to a right-sided radical operation, an extradural abscess in the middle cranial fossa being removed, but, on puncture of the dura, only bloodstained fluid was discharged under pressure. As the fever and the brain symptoms persisted unchanged, the operation was repeated, the brain being punctured without, however, revealing any abscess. The cerebrospinal fluid was normal and under normal pressure. The brain symptoms subsided very slowly, and four months after the last operation the patient still presented paresis and contracture of the left-sided extremities.

Symonds has had the opportunity of observing three cases of otogenous encephalitis. Two of the patients suffering from otitis media presented symptoms of an abscess of the temporal lobe, which could not, however, be detected in one of them. The other patient refused to submit to operation. Information as to temperature, pulse rate and conditions of the cerebrospinal fluid is missing.

The third patient, a 10-year-old boy with right-sided otitis media, three weeks after mastoidectomy developed "slow cerebration", the plantar reflexes being of the extensor type and the abdominal reflexes on the left side reduced. The temperature oscillated between 99 and 105° F., the pulse rate varied from 90 to 120. The cerebrospinal fluid was normal but under increased

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pressure. Exploration of the brain revealed no abscess. The post-operative temperature remained high during a protracted period.

Symonds mentions three cases of pseudo-abscess in children reported by *Adson*. In two of these cases the exploratory operation did not reveal any abscess, whereas the third case was not submitted to operation. *Symonds* asserts that these six cases are doubtless cases of otogenous, non-purulent encephalitis, which he continues to consider a preliminary stage of abscess.

This conception is also adhered to by *Horgan* in his discussion of the following case treated by him.

A 16-year-old boy with right-sided chronic suppurative otitis, one week after the acute onset of the disease developed left-sided hemiplegia and a temperature of 99.2° F. Pulse rate 120. Cerebrospinal fluid normal. After mastoidectomy and exploration of the brain without result, the fever and the symptoms persisted for a protracted period, yielding entirely, however, to a decompression operation.

Bijleveld's case is as follows :—

A 5-year-old boy after bilateral mastoidectomy developed focal symptoms from the right temporal lobe accompanied by high fever. On puncture of the brain, softened brain tissue was aspirated but no pus. The symptoms gradually disappeared entirely after the radical operation.

In all these cases of otogenous encephalitis located in the cerebrum the symptoms subsided very slowly, and the patients were discharged after a period of from one to four months only.

Location in the cerebellum seems to be of far less frequent occurrence. However, *Westergaard* describes such a case :—

A 12-year-old boy, who was submitted to operation on account of left-sided chronic suppurative otitis with perisinus abscess, a couple of days later developed cerebellar nystagmus, marked left-sided dysdiadokokinesia and cerebellar ataxia, besides papilloedema. The temperature and pulse rate were normal. The symptoms disappeared entirely after a lapse of six weeks without exploration of the brain.

Lately, *Shapiro* has published an interesting case of pseudo-cerebellar abscess.

A man, æt. 22 years, during the course of a right-sided chronic suppurative otitis, developed diffuse manifest labyrinthitis, an

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extradural abscess in the posterior cranial fossa and sinus thrombosis. During and following the operation performed on account of these complications the patient had a rise of temperature up to 104° F., which persisted for a protracted period; an apparently septic condition accompanied by increased intracranial pressure with papilloedema and some evidence of inco-ordination suggestive of cerebellar involvement on the right side. The cerebrospinal fluid was under high pressure with 4-8 cells per mm. All the symptoms gradually subsided in the course of four weeks, and the patient was discharged in perfect health.

Shapiro thinks that this case must be classified with the group of more or less obscure cases of intracranial hypertension which sometimes occur as a complication of ear suppurations; however, in his discussion of the differential diagnosis he does not mention that the morbid picture may probably be classified as that of otogenous encephalitis.

Thus, according to the literature, otogenous non-purulent encephalitis is not of frequent occurrence. Possibly this is due to the difficulty of diagnosing a good many of the cases or to their being termed pseudo-abscesses.

In the following notes a typical case of otogenous non-purulent encephalitis will be described.

A woman, *æt.* 30 years, was admitted to the ear department of the Rigshospital 20.8.1936. She had for twenty-six years suffered from chronic suppurative right-sided otitis media following scarlatina. During the last year she has had attacks of giddiness and vomiting. Ten days prior to admission she again had an attack of giddiness, vomiting, pains in her right ear and a temperature of 100.4° F.

Objective examination showed that she was in possession of her mental faculties; no torpor. Temperature 101° F.

Right ear: The auditory meatus contained a moderate quantity of foetid secretion, the deeper part containing a polypus which concealed the tympanic membrane. There was slight horizontal rotary nystagmus toward the left. No tenderness of the mastoid process, no nuchal rigidity, no Kernig's sign, or Babinski's reflex.

22.8.36. Right ear: The polypus was removed. The acoustic function was absent, and the vestibular reaction was reduced though scarcely abolished on the right side. By irrigating the right ear with water at a temperature of 112° F. the spontaneous nystagmus toward the left seemed to be checked, but no nystagmus could be elicited toward the irrigated side. By irrigation of the left ear with water at 86° F. a nystagmus of normal type towards the right was elicited after a lapse of 25" seconds.

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X-ray examination of the temporal bone revealed bilateral impaired pneumatization, without signs of ostitic processes.

During the following days the condition remained stationary, with giddiness, nausea, slight diffuse headache, and a slight rise of temperature: $99.4-99.8^{\circ}$ F. There was a continual slight nystagmus towards the left.

28.8.36: Rise of temperature to 101.2° F.

During the following two days the rise of temperature persisted, and a paresis of the lowest rami of the left facial nerve, paresis and hyper-reflexes of the left arm and leg, and positive left-sided Babinski reflex appeared. The lumbar puncture yielded slightly turbid, sterile fluid; 460 cells per mm. (chiefly polynuclear), with slight hyperalbuminosis, under a pressure of 220 mm.

There was no tenderness on percussion of the skull. No nuchal rigidity or positive Kernig's sign. Ophthalmoscopy normal. No defects of the visual field and no cerebellar asynergies. There was no bradycardia, the pulse rate varying between 100 and 120.

31.8.36: A radical operation was performed on the right side. The bone was hard and acellular. No pus. No cholesteatoma. Apart from a little granulation tissue in the antrum and a carious malleus nothing definitely pathological was found. As it seemed probable that there was an abscess in the right mesocranial fossa, craniotomy was performed, with cerebral puncture in five different directions but without revealing any abscess. The sinus was normal. The craniotomy was extended posteriorly and the cerebellum was punctured in three directions, but also without revealing any abscess.

The patient's condition subsequent to the operation was just the same as before it. The lumbar puncture was repeated three days later but revealed signs of increase of chiefly polynuclear cells up to 1,000 per mm. No micro-organisms.

10.9.36: For the first time the patient showed slight nuchal rigidity, positive Kernig's sign, and dysdiadokokinesia of the left arm, which was not quite reliable, however, as there was paresis of that arm. Repeated examination of the eye revealed normal conditions. The temperature became swinging, with marked remissions, of a more or less pyæmic character. The Widal reaction was negative. No growth on blood cultures. Leucocytes 9,760, neutrophils 65 per cent., eosinophils 2 per cent., monocytes 7 per cent., lymphocytes 26 per cent. WR. negative.

6.10.36: On lumbar puncture turbid fluid was evacuated under pressure of 800 mm., 2,000 cells per mm. The fluid was sterile. A further operation was performed with repeated puncture of cerebrum and cerebellum but no abscess was found. The sinus was denuded, part of the transverse sinus being involved, and after puncture some venous blood was removed by means of the

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aspirator. Wherever the dura became visible it had a normal appearance.

The post-operative temperature continued to swing for about a fortnight, but the patient was markedly unaffected by it. Subsequently the temperature fell to normal.

28.10.36 : The wound was clean and satisfactory in appearance. As the neurological symptoms persisted, the patient was transferred to the nerve department of the Rigshospital. She then developed a slight haziness of the papillae, with striate hæmorrhage in the upper part of the left disc. A slight convergent insufficiency was also detected. The visual acuity and visual fields were normal.

The examination of the cerebrospinal fluid revealed 550 cells, chiefly polynuclear. The pressure was 280 mm., with a satisfactory rise on Queckenstedt's test. In the course of the following three weeks the subjective and objective symptoms gradually subsided.

28.11.36 : The wound behind the right ear was healed. The patient was able to hear whispered voice at a distance of 15 cm. from her right ear. The spontaneous nystagmus had disappeared. She was discharged in perfect health after three months' uninterrupted hospital treatment.

4.7.37 : Readmission for examination.

Right ear : The resection cavity was dry and epidermized. The patient was able to hear the whispered voice at a distance of 15 cm. on the right side, but a thorough acoustic function test could not be carried out. No spontaneous nystagmus. No fistula symptoms. There was definite caloric function on the right side, though weaker than on the left. The eye examination revealed normal conditions.

As a remnant of the previous neurological findings, there are now merely hyperactive reflexes of both legs, with plantar reflexes of the extensor type. The patient's gait is normal. She has no tendency to sway on being submitted to Romberg's test.

Commentary

If we summarize the case report of this female patient there is a question of right-sided suppurative otitis media with destruction nystagmus toward the left owing to labyrinthitis, probably serous labyrinthitis, the function of the labyrinth not being entirely abolished, the temperature apart from the day of admission being subfebrile.

Not until eight to nine days after admission did marked intracranial symptoms become manifest, particularly paresis of the lower rami of the left facial nerve, paresis and hyper-reflexes of the left arm and leg, with positive Babinski sign,

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and turbid, sterile cerebrospinal fluid with pleocytosis under slightly increased pressure. These symptoms, together with the report of attacks of giddiness and vomiting during a year, were so highly suggestive of the existence of an abscess that a right-sided radical operation was resorted to with subsequent cerebral and cerebellar punctures, which were repeated five weeks later at a further operation, the result however being negative. The sinus and dura were normal.

These unproductive punctures of the brain and the favourable course of the disease with complete disappearance of the intracranial symptoms within three months, show that this morbid picture must most probably be interpreted as one of otogenous non-purulent encephalitis with benign collateral meningitis.

The assumption that, in the present case, there should be a possibility of an abscess that has either been absorbed spontaneously, or which has encysted and temporarily subsided, cannot be excluded, though it is improbable, considering that the general and focal brain symptoms have entirely disappeared and the patient has been free from symptoms for a period of eighteen months.

As regards the symptomatology, judging from those cases met with in the literature, the morbid picture may develop either quite suddenly or quite slowly, more often in children and young adults, in connection with both acute and chronic ear suppuration. The diagnosis of otogenous encephalitis without abscess-formation is extremely difficult. In many cases, in which it must be assumed that the pathological processes are located in the silent areas of the brain, or perhaps are very limited in extent, the symptoms are so vague and atypical that the affection is either not recognized at all or misinterpreted. In pronounced cases, such as for instance the one described, the symptoms are exactly like those of cerebral abscess.

Borries, in his clear and adequate manner, has set forth the definition that the encephalitis is characterized by the syndrome "abscess symptoms without abscess". Nor is it probable that there has been a question of an abscess if the operation does not reveal any pus and the patient recovers; or, in case of death, if autopsy reveals encephalitis without any sign of abscess-formation. As a rule the diagnosis will be arrived at *per exclusionem* only with the help of the further course of the

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disease. Thus in all the forementioned cases the diagnosis initially considered the most probable one, was that of cerebral abscess.

Of particular interest in this respect is the temperature of the patients. *Oppenheim*, in 1897, emphasized the importance of high fever for the differential diagnosis between non-otogenous encephalitis and abscess. Only in nine of the fourteen cases recorded above was information available concerning the temperature of the patients. One of them presented a normal temperature, whereas the other eight were highly febrile during protracted periods, the temperature of several of them being of a septicopyæmic character. The two cases reported in 1921 by *Borries* were also highly febrile. How much importance should be attached to this symptom in patients in whom other causes of fever cannot easily be excluded, will be difficult to decide.

The pulse rate attending otogenous encephalitis, apart from a couple of cases with a somewhat slow pulse rate, has always been proportionate to the temperature. Thus no cases with true bradycardia have been observed.

The cerebrospinal fluid presented every picture imaginable, ranging from normal without cells, with or without increase of pressure, to turbidity and containing numerous cells. Probably lumbar puncture will hardly afford any help with regard to the differential diagnosis of cerebral abscess.

As the changes in the cerebrospinal fluid in cases of abscess are known to be due to the attendant encephalitis or meningo-encephalitis, the otogenous encephalitis without abscess-formation may likewise present the contrast between the benign course of the cerebrospinal fluid symptoms and the malignant course of the cerebral symptoms otherwise so characteristic of the abscess (*Borries*).

Concerning the therapy, it can merely be said that otogenous encephalitis has in most cases been treated as a cerebral abscess, because an abscess could never be excluded with certainty. This treatment will no doubt always be the most adequate, as an abscess must be emptied, and the operation is not likely to increase the encephalitis.

The knowledge of otogenous encephalitis as an autonomous affection will, amongst other things, be of importance in the estimation of the prognosis in cases of apparent cerebral abscess, in which the puncture fails to reveal pus. Such cases

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must not be considered hopeless and, on that ground, tempt the surgeon, under the assumption that the patient's only chance of recovery is the evacuation of pus, to incise the brain without restraint in many different directions.

The prognosis can on the whole be considered favourable. It is true that a few cases of otogenous encephalitis with fatal issue are on record, but in the majority of cases the prognosis has been good not only *quo ad vitam* but also with regard to the sometimes very marked intracranial symptoms, which nearly always have subsided entirely.

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Verf. unterzieht die Geschichte dieser Erkrankung einer Durchsicht und führt Fälle an, über die in den letzten Jahren von andern Autoren berichtet wurde. Er gibt eine eingehende Beschreibung eines von ihm behandelten Falles, der vollständig ausheilte. Er glaubt, dass die Auffassung der otogenen Encephalitis als selbständige Erkrankung wichtig ist, unter anderm bei der Bewertung der Prognose in Fällen von scheinbarem Hirnabszess, bei denen die Punktion keinen Eiter ergibt. Solche Fälle dürfen nicht als hoffnungslos angesehen werden und den Operateur veranlassen das Gehirn nach verschiedenen Richtungen zu inzidieren, um womöglich den Eiter zu entleeren. In der Mehrzahl der Fälle ist die Prognose eine gute. Die intrakraniellen Symptome verschwinden in den meisten ausgeheilten Fällen vollständig.

L'auteur passe en revue l'histoire de cette affection et rapporte les cas observés par d'autres auteurs au cours de ces dernières années. Il donne une observation détaillée d'un cas qu'il a soigné

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et qui a guéri complètement. Il considère que la connaissance de l'encéphalite d'origine otique comme une affection autonome, aura, entre autres choses, son importance dans l'estimation du pronostic pour les cas de faux abcès cérébral, lorsque la ponction ne ramène pas de pus. Ces cas ne doivent pas être considérés comme désespérés et induire le chirurgien à faire des incisions multiples du cerveau avec l'espoir d'évacuer le pus. Le pronostic dans la majorité de ces cas est favorable et chez la plupart des malades qui guérissent, tous les symptômes intercraniens rétrocedent complètement.