Masked diabetic mastoiditis

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In the last few years a number of papers have been published about the condition known as 'malignant external otitis'. Meltzer and Kelemen (1959) described a case of pyocyaneus osteomyelitis of the temporal bone in an old diabetic. Microscopical examination of the temporal bone after his death revealed extensive involvement of that bone, although the labyrinth, cochlea and internal acoustic meatus were not involved.

Chandler (1968) reviewed the disease, described 13 patients, and termed it 'malignant external otitis'. He emphasized the triad of pseudomonas otitis externa, diabetes mellitus and old age. He stated that the canal floor is the most important site as the primary route of invasion. Thereafter, more cases were recorded in the literature: Wilson *et al.* (1971) described four cases of this disease and Schwartzman *et al.* (1972) reported that the characteristics of this affliction are unremitting pain and infection with abundant granulation tissue originating in the external ear canal, usually in its floor, at the junction of the osseous and cartilagenous portions, whence it invades the cartilage, bone, nerves and soft tissues.

This paper deals with a further seven cases met with by the authors, with a discussion about the origin and basis of the disease.

Case reports

Case 1

M.G., a male aged 60 years, was seen in June 1967. He felt pain and diminution of hearing in the right ear, which was diagnosed as furunculosis and treated with antibiotics and glycerine-icthyol drops. One month later, the pain was increasing and the swollen external canal not resolving. Culture produced bacillus pyocyaneus. Diabetes mellitus was suspected and laboratory investigations were positive. Anti-diabetic treatment began and heavy antibiotics were administered. Pain was increasing and tenderness could be elicited over the mastoid process. X-ray showed blurring of the mastoid air-cells. A cortical mastoidectomy was done. The pain, however, was not much relieved. Two weeks later, a slight right facial weakness was noticed and a swelling and puffiness in the temporal fossa above the zygomatic arch appeared. A radical mastoidectomy was performed, but the pain was not relieved and weakness of the right side of the palate was noticed. Three weeks later, paralysis of the right vocal cord was seen. The ear discharge continued in spite of repeated cultures and sensitivity tests and administration of the appropriate antibiotics. Four months later, the patient consulted Professor Portman in Bordeaux. There,

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X-ray of the skull revealed rarefaction of the petrous tip, together with the right occipital condyle of the skull and right half of the first and second cervical vertebrae. The patient's condition deteriorated greatly and he died 2 weeks later, i.e. one year from the onset of the disease.

Case 2

M.S., a male aged 65 years, presented to our hospital complaining of right ear discharge, otalgia and right facial paralysis. He was diabetic for ten years. Three months beforehand, he had pain in the right ear which was diagnosed as otitis externa and treated as such. Oedema of the external auditory meatus was present and granulation tissue started to appear in the floor of the external ear canal. Culture produced bacillus pyocyaneus. X-ray showed haziness of the mastoid air-cells on the same side. Tenderness over the mastoid was elicited. A cortical mastoidectomy was performed, but the pain was not much relieved and one week later, swelling of the right temporal fossa above the zygomatic arch was noticed. Palatal paralysis then appeared together with right cordal paresis. A radical mastoidectomy was done two weeks later, but the patient developed bronchopneumonia and died.

Case 3

A.E., was a 70-year-old diabetic man, who complained of right otalgia and ear discharge of one month's duration. Examination revealed external otitis with granulations in the floor of the right external meatus. The responsible organism was bacillus pyocyaneus. A cortical mastoidectomy was advised, but he refused. One-and-a-half months later, he developed right facial paralysis and marked tenderness over the mastoid tip. A radical mastoidectomy was done, but the discharge continued and the patient died six months later.

Case 4

A.Z., a male aged 35 years and diabetic, presented with pain and foul discharge from the left ear of three weeks' duration. On examination, there was oedema of the external ear canal with granulations arising from the floor of the canal and tenderness over the mastoid tip. Culture produced bacillus pyocyaneus. A cortical mastoid operation was done and the pain disappeared. A month later, he developed slight left facial paresis. A radical mastoidectomy operation improved the facial function. Eight months later, the patient developed pain and swelling of the left temporal fossa.

Case 5

M.A.D., a 60-year-old diabetic man presented with severe right aural pain, throbbing in nature, and scanty foetid discharge. On examination, there were granulations in the external ear canal arising from its floor. Culture grew bacillus pyocyaneus. A cortical mastoidectomy showed some granulations in the tip cells. Two months later, he died of inhalation bronchopneumonia.

Case 6

S.S.M., a female aged 50 years, diabetic. She complained of bilateral deafness and yellowish offensive discharge of both ears of two months' duration. Examina-

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tion revealed fleshy granulations filling both external ears. Culture produced bacillus pyocyaneus. X-ray mastoids revealed moderate pneumatization of both mastoids. The patient was operated upon by excision of the external meatal skin bearing granulations and restricted more or less to the cartilagenous portion.

Case 7

M.S., a male aged 75 years, complaining of left ear discharge, left otalgia and deafness of two months' duration. He was diabetic. On examination, there were fleshy reddish granulations filling the left-ear canal. Culture grew bacillus pyocyaneus. X-ray revealed clouding of the left mastoid process. A cortical mastoidectomy was done and the mastoid tip cells were filled with granulation tissue which spared the antrum, peri-antral cells and middle-ear cavity. The patient had an uninterrupted post-operative course and was cured.

Discussion

The seven cases described above fulfil the specifications of the triad given by Chandler (1968) to the diagnosis of 'malignant external otitis', namely: old age, diabetes mellitus, and pseudomonas otitis externa.

It is clear that the follow-up of our patients showed that all the cases, although starting with external otitis, progressed to present with the clinical picture of otitis media and mastoiditis and even progressed in many cases to petrositis, and in four out of the seven cases to a fatal end.

This led us to doubt the process as being an 'external otitis' and to believe that from the start it is related to an infective process in the mastoid.

It is known that air cells of the temporal bone vary considerably in respect to size, number and distribution from one individual to another. These spaces develop in a manner similar to that which produced the cavities of the tympanum, epitympanum and antrum. In reality, the air cells are merely outpouchings of the latter spaces.

The petrous and temporal air cells arise from three sources (Figs. 1 and 2): (1) antrim; (2) Eustachian tube; (3) tympanum.

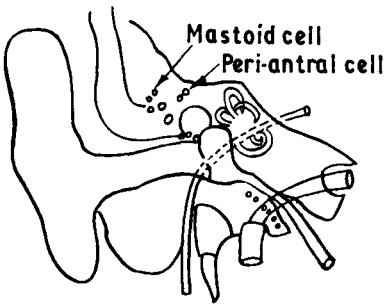


FIG. 1.

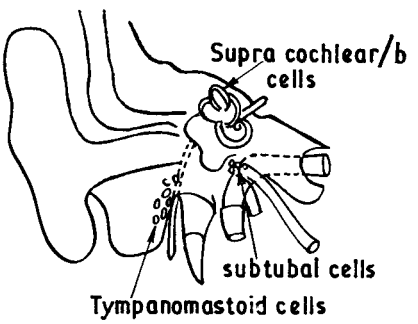


FIG. 2.

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(1) The air cells which arise from the antrum are the mastoid and peri-antral cells.
(2) The tubal cells arise from the floor of the Eustachian tube.
(3) The air cells derived from the middle ear are:
    (a) From the floor of the tympanum, cells extend below the Eustachian tube (sub-tubal cells) and sometimes between the cochlea and jugular bulb.
    (b) From the anterior aspect of the tympanum, cells extend in front of, behind and above the carotid artery (peri-, post- and supra-carotid cells).
    (c) Also in front of and above the cochlea (pre- and supra-cochlear cells).
    (d) Cells frequently grow directly from the tympanic cavity, medial to the facial nerve, into the tip of the mastoid process (Shambaugh, 1967). These cells have not been named before. We suggest the name 'tympano-mastoid cells', to denote that they arise from the tympanum and go directly to the mastoid. Actually this name is very suitable, especially as these cells are close to the tympano-mastoid suture (Fig. 3).

From this anatomical review, it is clear that the cells in the mastoid process do not all have the same origin; some arise from the antrum, and others arise directly from the middle-ear. In our view, this difference in origin has a great bearing on the clinical course of middle-ear infection in diabetics.

Our case 1 will be taken as an example. This patient presented with a furuncle of the external meatus, treated for six weeks, but no improvement was noticed and when diabetes mellitus was discovered, and the culture produced bacillus pyocyaneus, the condition fitted the specifications for 'malignant external otitis'. Heavy courses of antibiotics and rigid control of his diabetes, however, failed to resolve the condition. Later on, the case progressed, and mastoiditis and later petrositis became apparent. This led us to think that the condition from the start was a middle-ear infection, especially as he complained from the start of a sense of decrease in hearing which was explained at first by obstruction of the external meatus by the furuncle and later on by the granulation tissue obstructing the meatus, but can also be explained by a concomitant mild middle-ear affection.

This mild middle-ear infection in the old diabetic may extend to those cells in the tip of the mastoid process which arise directly from the tympanum, and when these cells point superficially they do so in the external-ear canal, exactly in the same way as when marginal cells point in the external-ear canal and give what is known clinically as 'sagging of the external meatus', thus leading to granulations and even fistula

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formation inside the meatus. To facilitate the comparison, one can say that it produces bulging of the floor (Fig. 4). The findings of Chandler (1968, 1972) and Wilson *et al.* (1971) of a dehiscence in the floor of the external canal from which granulations appear in the external canal may be explained either by an actual dehiscence or by a fistula produced by infection extending from these cells.

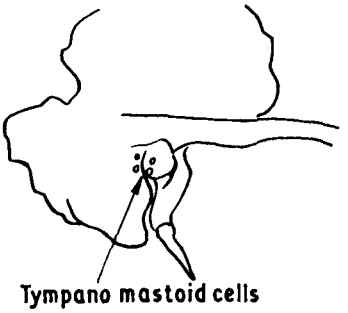


FIG. 3.

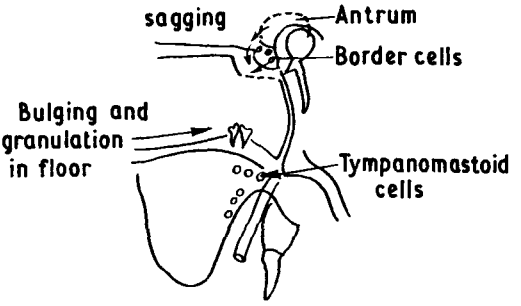


FIG. 4.

Chandler (1972) reported another 26 cases of malignant external otitis, with an attempt to shed some light on the pathogenesis of facial palsy in this disease. We cite here two of these cases.

Case 2 was an 83-year-old male who had been treated for external otitis six weeks previously. Examination revealed granulation tissue in the floor of his canal at the junction of the membranous and osseous portions. Mastoid X-ray revealed moderate clouding of a pneumatic mastoid process. Five days later, he had some ipsilateral facial weakness; and three weeks later, an extended radical mastoidectomy and total decompression of the facial nerve were accomplished. A well-pneumatized mastoid process was encountered with normal antrum and peri-antral cells. There was granulation tissue in the mastoid tip cells adjacent to the digastric ridge.

Case 3 was a 75-year-old diabetic patient with a history of treatment of external otitis for three weeks. Examination revealed marked swelling of the left external auditory canal with granulation tissue bulging into the canal floor at the junction of its cartilagenous and osseous portions. X-ray revealed a cloudy pneumatized mastoid process. At operation, extensive erosion and pitting of the mastoid cortex was encountered. There was an extensive granulomatous mastoiditis which spared the antrum, middle ear and squamosal cells. An extended radical mastoidectomy was completed which revealed extension of infection to the petrous tip through the sub-labirynthine cells.

These two cases show how the so-called 'malignant external otitis' presents from the first beginning with:

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(a) Cloudiness of the mastoid air cells.
(b) Granulation tissue found in the retrofacial cells and mastoid tip cells adjacent to the digastric ridge, leading to the sublabyrinthine cells and petrous apical cells.
(c) Involvement of the facial nerve.

Chandler (1972) believes that infection originating in the external meatus, in weak diabetic patients, causes 'secondary mastoiditis' as a result of direct extension through the mastoid cortex or Fallopian canal. Morgestein and Seung (1971) presented a temporal bone report of a patient who suffered from pseudomonas mastoiditis, petrositis and death, which was 'clearly' of external canal origin (Fig. 5).

In our view, it is difficult to accept the belief that such a superficial infection as external otitis, however 'malignant' it is and however long the diabetes has been present would burrow itself so deeply in the surrounding area as to erode bone and extend to the mastoid process and even deeper to the petrous apex with an unhappy fatal end in many cases. It is more credible that the condition is from the start a 'masked mastoiditis' which begins as a very mild infection in the middle ear cleft and rapidly settles in the mastoid tip cells, which arise directly from the tympanum.

This mastoiditis, modified by antibiotic therapy for the 'external otitis', progressed without the warning signs and symptoms. The neuropathies and vasculopathies associated with diabetes add to this silent progress until it points in the external ear canal, as mentioned before, in the same way as marginal cells produce sagging of the external meatus in cases of acute mastoiditis (Figs. 4 and 6).

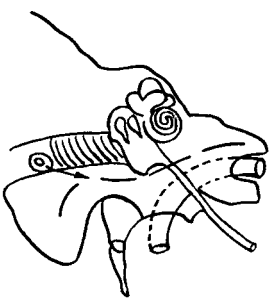


FIG. 5.

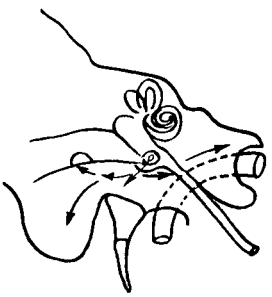


FIG. 6.

In our cases where energetic treatment was taken up early and a cortical mastoidectomy drained the primary infection in the bone, the process was arrested and a cure was sometimes achieved. But in many cases, the patients were treated as external otitis and precious time was

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lost until the condition in the mastoid had established itself and had even progressed to deeper cells in the temporal bone. A cortical or even a radical operation at that time in many cases failed to arrest the disease and it extended to the petrous apical cells with frank signs of petrositis.

Summary

Seven cases of elderly diabetics suffering from the so-called 'malignant external otitis' are presented. Chandler (1968) and others consider that this disease is localized to the external ear canal. We do not agree and believe that it is due to a masked middle-ear infection extending through the tympano-mastoid cells to the mastoid process, sparing the antrum and bulging into the floor of the external meatus. In time, extension to petrous apex results in petrositis and a fatal end in many cases.

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