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IDIOPATHIC HÆMOTYMPANUM

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HÆMOTYMPANUM is still a sufficiently uncommon condition for sporadic cases to be reported, and a search of the literature might appear to give the impression that the discovery of a blue drum is a clinical rarity. Credit for the first full description of the condition belongs to Shambaugh who, in 1929, noted two such cases, and illustrated the appearance of the drum with a colour plate. Fowler (1939) made a brief reference to a personally observed case, while O'Donnell (1941) described two cases in children, and suggested the name "idiopathic hæmotypanum" for the condition. Kler (1948) discussed one case, and Ranger (1949) reviewed the literature, and added observations and investigations in two cases. Vior (1951) described a case of blue drum, and Menck-Thygesen (1952) added two cases, one of whom had the condition bilaterally. Johnston (1953), in an interesting article, discussed two cases, while further cases have been reported by Simonton (1955), Bourgeois and Natalie (1955), Terkildsen (1956) and Hall (1957), each of whom discussed one case, while Sheehy and McKibben (1956) described three cases. These, as far as I am aware, are the only published reports of the condition.

Against this total of twenty cases, it may appear to be presumptuous on my part to describe a further eight cases, two of whom showed the condition bilaterally, but the fact that six of them were discovered within a period of twelve months appears either to confirm the view expressed by Menck-Thygesen that the condition may not be as infrequent as has been thought, or to suggest that the ætiological factor may be becoming more common.

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Sufficient is known about the condition to classify it as a clinical entity, not a clinical rarity. It has a well-defined history, diagnostic clinical features, consistent radiographic pictures and operative findings, and characteristic histological appearances from material in the mastoid cells. It cannot be named with accuracy until the ætiology has been finally established, but it must be fitted into our conception of otomastoiditis.

Case Reports

The first two cases may be dismissed briefly as notes are not now available to me, and as no treatment was carried out.

Case I. A boy, aged 14, was examined on account of deafness and recurring attacks of pain in the right ear over some years, but with no discharge. Examination was negative except that the right drum was of an intensely blue colour, but of normal contour and with a cone of light. Tuning fork tests showed a conductive deafness in this ear, and radiographs revealed extensive pneumatization of both mastoids, the cells in the unaffected side being healthy, but those in the right mastoid being uniformly blurred. This boy was examined in a Cottage Hospital in the Border district of Scotland, and the parents were unwilling to allow surgery.

Case II. A youth, aged 19, was examined at the request of the Ministry of Labour and National Service on account of a history of recurring attacks of pain and deafness in the left ear. The left drum was deep blue in colour with a normal light reflex, and radiographs showed a uniform obscurity of a well-pneumatized mastoid process on this side. He was asked to consult his own doctor with a view to being referred back for surgery, but he has not been seen again by me.

Case III. Nicolas W., was examined on 7.3.56 when he was aged 3, on account of recurring earache in both ears, with discharge from the right ear. At that time the right meatus was filled with pus, a swab of which yielded no growth, and the left drum was normal. He had unhealthy tonsils and adenoids, and these were removed on 4.5.56. He was examined again on 21.5.57 on account of further attacks of pain, confined now to the left ear, but with no discharge. The right drum showed a healed central perforation, while the left was intact, blue in colour, and of normal contour. He was again seen ten days later on account of severe pain in the left ear for one day followed by a sudden blood-stained discharge. On examination a little dried blood-stained material covered the drum, and by 4.6.57 the drum was of a normal colour but was bulging. A week later the drum appeared normal. On 2.7.57 there was a further attack of pain in the left ear, and the drum was again bulging and blue in colour. X-rays showed the right mastoid to be normal, while the cells on the left side were obscured and cellular break-down was reported.

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On 10.7.57 the *left mastoid* was opened, and a large coalescence of cells was found near the tip. This necrotic area contained thick brown mucoid secretion. The mucosa of the antrum was thickened. The attic was opened, and the incus appeared to be healthy. *Pathological examination* revealed giant cells, cholesterin clefts and hæmosiderin. He was discharged from hospital ten days after operation, when the drum was slightly indrawn but of normal colour and it has remained thus. The last examination was in May, 1958 when the drum was of normal colour, and there has been neither pain nor deafness in the ear.

Case IV. Raymond T., was examined on 5.6.56 when he was aged 6, on account of recurring attacks of pain and deafness in both ears, treated by his own doctor on several occasions with oral penicillin and sulphonamides. There had been no discharge. His drums were indrawn and of normal colour. His unhealthy tonsils and adenoids were removed on 27.6.56. He reported again on 11.7.57 on account of further attacks of pain and deafness in both ears, treated by his doctor with chloramphenicol. Both drums were intensely blue in colour and were indrawn, and he could only hear a whisper at some 4 in. in either ear. X-rays showed bilateral sclerotic mastoids. On 29.7.57 he came to the operating table, and examination of the drums before operation showed both to be indrawn and of normal colour. It was decided however, to proceed with the operation, and the *right mastoid* was opened. The cortex was dense, but the mastoid contained an area of necrosis, filled with black cellular debris extending from the tip to the antrum and petrosal angle. The lateral sinus plate was involved in this necrotic area, but the wall of the sinus appeared to be healthy. More debris was found in the antrum, and blackish-brown mucoid material exuded through the aditus through which it was aspirated. There was a similar infection in the zygomatic cells. *Bacteriological examination* showed the fluid to be sterile. The *pathological report* showed giant cells, cholesterin spaces and hæmosiderin. On 2.8.57 the *left mastoid* was opened, and the findings were identical with the other side, even to the lateral sinus being exposed by necrosis. The attic was not opened, but aspiration was continued through the aditus until no further black fluid could be obtained. Again the fluid was sterile and *histology* showed giant cells and cholesterin spaces. By 22.8.57 both drums were normal, and on 3.10.57 he could hear a whisper at 12 ft. in either ear.

Case V. Sydney S., was first examined in 1952 when he was aged 5, on account of deafness, especially in the left ear. The left drum was indrawn and of normal colour, and his tonsils and adenoids were removed. He was seen again on 13.6.57 after his third attack of pain and deafness in the left ear within a year. On this third occasion there had been a sudden discharge of blood and pus. The left drum was bulging and of a deep blue colour, and he only heard a whisper close to this ear. The right

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drum was normal, and his nose and throat were clear. X-rays of the mastoids showed both processes to be poorly pneumatized with obscurity and cellular break-down in the left side. On 3.7.57 the *left mastoid* was opened and found to contain black necrotic cells and a thick yellow-green discharge. A break-down of the cells in the petrosal angle was cleared out, and the subdural cells were also necrosed. The mucosa of the antrum was thickened; the attic was opened and the incus was seen to be healthy. Glairy blackish-green fluid was aspirated from the floor of the attic external to the incus. The *pathological report* showed cholesterin clefts and giant cells, but no hæmosiderin and no purulent infiltration. By 22.7.57 the drum was of normal colour, and he heard a whisper at 6 ft. in the ear.

Case VI. Patrick O'N., developed a meningitis in 1951 when he was five years old. He was examined on 27.3.52 because of deafness in both ears. The drums were indrawn and of normal colour, and he could only hear a conversational voice at 4 ft. in the right ear and at 2 ft. in the left. It was noted that he had large tonsils and adenoids, but apparently no action was taken. He reported again on 25.11.54 on account of recurring attacks of pain in both ears. Examination again showed indrawn drums and on 10.12.54 the Eustachian tubes were inflated. Both were patent, but the right tube was relatively clearer than the left. Under the same anæsthetic his tonsils and adenoids were removed. On 29.10.57 he was again seen with a history of recurring otitis media and deafness, worse in the right ear than the left. The right drum was indrawn and of normal colour. The left drum was also indrawn, showed a scar anteriorly, some crusting in the attic region, and was blue in colour. Examination of the nose, nasopharynx and throat was negative. X-rays of the sinuses were also negative, but those of the mastoids showed bilateral sclerosis. On 25.11.57 he came to the operating theatre when examination showed the left drum to be indrawn and of normal colour, with an anterior scar and an attic crust. The *left mastoid* was opened and was diploic in type. Black cellular necrosis was found superficially in the angle between the lateral sinus and the middle fossa dura. The antrum appeared to be healthy, but, on opening the attic, the incus was found to be lying loose, and was removed. Both processes were eroded. A large granulation was removed from the attic. The head of the malleus was healthy, and no cholesteatosis was found. The *pathology* showed that the granulation tissue from the attic contained the elements of cholesterin granuloma—cholesterin clefts, giant cells and hæmosiderin. There was no squamous epithelium and no pyogenic infection. As the right ear had been more markedly involved in the previous pain and deafness, the *right mastoid* was opened on 2.12.57, but contained no black cells. Again, the incus was lying loose, and was seen to be eroded after its removal. On 7.1.58 both drums were indrawn and healthy in colour,

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but he was still deaf. On 16.2.58 the Eustachian tubes were inflated, and, again, both were patent.

Case VII. Graham Y., had his tonsils and adenoids removed at the age of 6 in another hospital in 1956 on account of deafness. The hearing improved thereafter, but he was seen on 18.2.58 on account of recurring pain in the left ear, treated by his own doctor with penicillin. The nose and throat were clear. The right drum was indrawn, and of normal colour. The left drum was also indrawn, but was intensely blue in colour. Rinné's test was positive in the right ear, and negative in the left, and Weber's test was lateralized to the left. He could hear a whisper easily at 10 ft. in the right ear, but only at 1 foot in the left. X-ray of the sinuses showed some mucosal thickening in the left antrum, and that of the mastoids revealed marked cellular break-down in the left mastoid, while the right was normal. On 7.3.58 the *left mastoid* was operated upon. The cortex was dense, but the mastoid contained a cavity filled with thick black mucoid discharge. There was extensive break-down of the mastoid cells, and the sinus plate was included in this necrotic area, which extended from the tip to the petrosal angle. The antrum and attic contained stringy mucus, chocolate in colour streaked with yellow. The mucosa of the antrum and attic was thickened, and the incus was healthy. *Bacteriology* showed the fluid to be sterile, and *histology* again revealed the characters of cholesterin granuloma—giant cells, cholesterin spaces and hæmosiderin. On 8.4.58 the drum was normal, and he could hear a whisper at 20 ft. in this ear.

Case VIII. Barbara D., had her tonsils and adenoids removed in another hospital, when she was aged 5, on account of deafness. She was examined on 10.2.58 because of deafness for a year, and attacks of pain in the right ear. Her tonsils had been well removed, and her nasal airway was good. The left drum was indrawn and blue in colour. The right drum was acutely indrawn and intensely blue. She could only hear a whisper close up in either ear. X-ray of the mastoids showed both to be poorly cellular, with no break-down. On 21.3.58 examination before operation showed both drums to be indrawn and of normal colour. The *right mastoid* was opened and found to be diploic in type with a few black cells near the tip. There was no break-down of the intercellular walls, but the mucosa of the antrum was thickened and stringy mucus was present in the cells throughout. The attic was opened and the incus was healthy. *Pathological investigation* failed to reveal any of the features of cholesterin granuloma. In view of this, the other mastoid was not operated upon.

These case reports, and the operative findings and pathological appearances in Cases III–VIII, bear such a striking resemblance to each other, and to the other published reports that one can, with some certainty, dogmatize on the features of the average case. One may

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assume that Case VIII showed the condition in the earliest phase with only a few black cells and negative pathology, while Case VI, in whom the incus was eroded and a large granulation was present in the left attic, represents the most advanced pathology.

History

A history of *pain or discomfort* in the affected ear has been a feature common to all cases. Earache, of course, is a frequent symptom in childhood, occurring bilaterally or in either ear at different times, but recurring attacks of pain confined to one ear are much less common and require particular investigation. Two points emerge from the histories detailed above. In the first place it is noticeable that in many of the children there was a history of bilateral otitis media, and that, after removal of tonsils and adenoids, the pain in one ear disappeared, and at a later X-ray examination this mastoid appeared healthy. On the other hand, the operation had no effect on the recurring attacks of earache in the ear in which the drum eventually became blue. This is in contradistinction to O'Donnell's experience in which removal of tonsils and adenoids cured the condition of blue drum.

The second point emerging from the history is that these attacks of earache have been treated by the family doctor with oral antibiotics. Efforts have failed to ascertain the exact state of the drum during these exacerbations, the exact antibiotic used, and the exact dosage of that antibiotic. One feels that the antibiotic currently popular at the time of examination has been the one used, irrespective of the sensitivity of the causal organisms. One feels, too, that the dosage may possibly have been inadequate, and that no very definite criteria of cure may have been observed. Masking may thus follow, and the symptoms may well recur, due to a focus of infection lying within the mastoid cells, the middle ear or the Eustachian tube.

Aural Discharge. This is not an invariable feature, and was only noted in two of my cases. In each it was described by the mother as being sudden, short-lived and blood-stained. The fluid is presumably the same as that obtained at paracentesis or aspiration of the middle-ear cavity, and this has been shown by Ranger to be essentially altered blood with a positive Van den Bergh reaction. Culture of such fluid is usually sterile, but Shambaugh found *Staphylococcus albus* in his cases. My two cases in whom there was a history of spontaneous discharge were not seen until the discharge had ceased, and no swab could be taken.

Deafness is a complaint common to all the histories, and it is noteworthy that this persisted in some measure between the attacks of pain. It varies from a muffled feeling reported in adults to a deafness complained of by the parents of the children. My own tests suggested that it was conductive in type, and this view is supported by O'Donnell, and Sheehy

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and McKibben. Ranger's audiograms show a fairly flat curve, as do those of Kler, Johnston and Simonton. Menck-Thygesen's audiograms, on the other hand, suggest a mixed type of deafness with a greater loss for the higher notes. Improvement in the audiograms is slight after myringotomy and more marked after mastoid operations.

Examination

As a rule, examination has been negative apart from the blue drum in the affected ear and the radiographic evidence of mastoid involvement on that side. In some cases, however, the history or clinical features in the nose or nasopharynx have led me to have the sinuses X-rayed, but the films did not show any consistently present abnormality.

The blue drum is characteristic of the condition, and is typically of a dark colour. Sheehy and McKibben have emphasized the distinction between this and the gun-metal blue of serous otitis media. Shambaugh remarked on the fact that the blue colour was confined to the pars tensa, while the pars flaccida was of normal colour, and this is not to be wondered at in view of the cause of the discoloration. All varieties of contour of the drum have been observed in association with this blueness, indrawing, normality and bulging. The jugular vein on the same side should be compressed as a diagnostic measure, because if the colour were due to the jugular bulb an alteration in the appearance of the drum should be observed.

I examined three of my cases on the operating table immediately before operation, after they had spent 24 hours in bed, and in none was the blue colour evident. Yet the characteristic black cells were present in the mastoids of all three cases at operation. This raises the question, which will be discussed later, as to whether the blue appearance is transitory or not.

The Eustachian tube. It is not surprising that the Eustachian tube should have been blamed for the deafness in these cases, and, indeed, for the source of the blood, even though many authors have commented on the ease with which tubal inflation could be carried out. Shambaugh and Fowler both declared that the tube was patent, Kler stated that the Eustachian tube could be inflated easily but noted that this produced no improvement in the deafness. Menck-Thygesen reported that there was no tubal occlusion at the outset of one of his cases, but that this developed later, and was therefore not the cause of the deafness. Johnston also noted patent Eustachian tubes, while in two of Sheehy and McKibben's three cases the tubes were clear. The tube was blocked in the third case.

Blood investigations have been carried out by a number of authors and have been largely negative. The presence of cholesterol clefts in the histological sections has led to the serum cholesterol being investigated.

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The difficulty of this is that there is a wide normal variation depending, among other things, on the technique employed, and differing in different laboratories. Ranger pointed out this normal variation but concluded that his patients showed no disorder of the blood which would give an increased tendency to bleed. Johnston found a normal blood picture and blood chemistry apart from a high prothrombin concentration of 70 per cent. (normal 30 per cent.). His patient had a serum cholesterol of 215 mg. per 100 ml., the normal being 110–390 mg. per 100 ml. Kler's patient had normal bleeding and clotting times, normal prothrombin and platelet count, and the tourniquet test did not reveal any fragility of the capillaries. Hall, who thought at the operation that the affected mastoid area was unduly vascular, could find no corroboration of this histologically or angiographically.

Mastoid radiography. It has been the universal experience of those who have had the mastoid processes X-rayed that the films have shown cloudiness on the affected side. Most of the films showed cellular processes, but in some pneumatization was not prominent. In the cellular mastoids, the cells were homogeneously opaque, and not infrequently there was well-marked evidence of destruction of the intercellular walls.

Treatment

Removal of Tonsils and Adenoids. O'Donnell stated that his two cases were cured of their deafness, and the blue colour of their drums gradually disappeared, following removal of the tonsils and adenoids. No other author has had a similar experience. It will be seen that six of my cases (Cases III–VIII) had had their tonsils and adenoids removed for a history of bilateral earache and deafness, and that at the time of that examination the drums were not blue. The operation relieved the condition in one ear, as one would expect, but the recurring attacks of earache and deafness persisted in the other ear, and this drum subsequently became blue. From this it would appear that the removal of tonsils and adenoids can have had no bearing on the progress of the condition, assuming that it was developing at that time, and that the source of the blue drum does not lie in infection of the lymphoid tissue of Waldeyer's ring.

Inflation of the Eustachian tubes. Inflation of the Eustachian tubes alone has no effect on this condition. As has already been stated, many of the authors mentioned that the tubes were patent, and Kler has asserted that tubal inflation does not improve the hearing. Again, O'Donnell is in the minority as he states that there was some obstruction to inflation by catheter. On inflation he reported that râles were heard, and afterwards the hearing for a whisper improved from 6 inches to 10 feet. The colour of the drum changed to a dull grey with brownish patches.

Myringotomy. This was the sole method of treatment until 1949. The drum was incised and thick chocolate coloured fluid was aspirated.

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Tubal inflation assisted in expelling the thick secretion from the middle-ear cavity. Analysis of this fluid has shown it to be altered blood, and most authorities have found it to be sterile. Shambaugh reported *S. albus* from the secretion. The result of myringotomy was a temporary relief from the symptoms and a slight improvement in hearing. The drum either remained blue (Hall) or quickly returned to its blue colour, and the process had to be repeated, either by myringotomy or by aspiration through the drum by means of a needle. Menck-Thygesen noted that the drum became anæsthetic, and aspiration could be performed without any local analgesia. Hall on the other hand states that the drum was so tender that pain was experienced on touching it with cotton wool, but this he ascribed to neurosis on the part of the patient.

Mastoid operation. The failure to effect a permanent cure by these methods led inevitably to an attempt at mastoid surgery, and Ranger, who was the first to describe this, gives a clear account of the removal of black debris from the mastoid cells, leaving an apparently healthy cell lining. He performed the cortical (Schwartze) operation and found that one of his cases had an extensively pneumatized mastoid, and the other a smaller acellular process. Since that date (1949) Menck-Thygesen, Bourgeois and Natalie, Johnston, Terkildsen, Sheehy and McKibben, and Hall have all used the cortical operation with success, while Simonton and Sheehy and McKibben have had to proceed to the radical operation, in one case each, in order to obtain a cure. My own experience has been confined to the cortical mastoid operation in Cases III–VIII without any previous attempt at myringotomy. This treatment has been based on the clinical findings of a blue drum together with radiological evidence of mastoid involvement. I can see no place for myringotomy in the treatment of the condition if, as seems certain, the source of the blood is the mastoid process. The extensive involvement of the mastoid process, and the cellular necrosis found at operation makes it obvious that nothing short of mastoid surgery can cure the condition. The results obtained by the operation appear to justify this view. The hearing improves, the pain disappears and the drum returns to its normal colour and remains thus.

Bacteriology

Bacteriological examination of the secretion in the mastoid cells shows it to be a sterile fluid. In the main the cases have been operated upon in the quiescent phase, and it may be argued that this sterility has been due to the antibiotic drugs used by the general practitioners. Case VII was operated upon immediately after an exacerbation of pain, but the secretion was sterile.

Pathology

Histological investigations of the mastoid cells and their black cellular content have consistently shown the presence of giant cells and

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cholesterol spaces. Less consistently, hæmosiderin has been found. Such a pathological picture is now known as cholesterin granuloma, and is not necessarily confined to patients with blue drums. I have previously described a number of cases exhibiting true cholesteatosis in the attic and cholesterin granuloma in the subcortical cells (1956).

Discussion

Johnston and Vior have discussed the possible causes of a blue appearance of the tympanic membrane, and the following have been mentioned (1) a fracture of the base of the skull, (2) the mechanism of the decompression chamber, (3) varices of the middle ear, (4) hæmangioma of the middle ear, (5) protrusion of the jugular bulb and (6) hæmophilia. There is no doubt that any of these could result in a blue drum, and there may be many more similar conditions, but it is quite obvious from the eight cases which I have recorded, and from the majority of the cases reported in the literature, that the condition described has a separate ætiology, and can be readily differentiated from the above by the history, blood examination and radiography.

It is generally agreed that the colour of the drum is due to the presence within the middle ear of a thick fluid which is essentially altered blood. The source of this blood must be the Eustachian tube, the middle-ear cavity or the mastoid cells. Earlier opinion favoured the tube or the middle ear, and it was only after similar fluid was found in the mastoid cells that it came to be recognized that these cells were the most probable site of origin. Hæmorrhage in the mastoid, as Ranger pointed out, would produce sufficient fluid so that it would be difficult to clear it by aspiration through the drum, whereas hæmorrhage arising in the tube or middle ear might easily be so cleared. Again, hæmorrhage in the middle ear or tube would not be cured by mastoid operation. The destruction of the intercellular walls, and the extent of involvement of the mastoid cells is such that it appears obvious that the mastoid is primary site of trouble.

One must therefore discount earlier theories, based on the assumption that the source of the blood is in the tube or middle ear. Kler's view that there is a congenital anomaly of the veins draining the tympanic membrane to permit an escape of a minimal amount of blood into the membrane and middle-ear cavity, and Johnston's theory of an area of varicosity or even hæmangioma in the middle-ear cleft which fails to undergo complete tissue repair, but which repeatedly breaks down to permit of the escape of fresh blood, may account for the occasional blue drum, but cannot be accepted as the common cause. Ranger examined O'Donnell's theory of a *hyperæmia ex vacuo* and rejected it on three counts (*i*) Eustachian obstruction is common but hæmotympanum is rare, (*ii*) Eustachian obstruction is not found in all cases of hæmotympanum and (*iii*) very few cases of *hyperæmia ex vacuo* have been

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found in acute otitic barotrauma occurring in aviators or experimentally produced in cats.

Menck-Thygesen places the origin of the bleeding in the mastoid cells, as do Sheehy and McKibben who added that surgery had so far failed to uncover the source of the bleeding. Hall thought that the affected areas in the mastoid were unduly vascular, but was unable to confirm this histologically.

I have been interested to find that examination of three of my patients before operation failed to show the blue drum which had previously been apparent in the out-patient department, and, that in spite of this, typical appearances were found in the mastoid process and typical sections were obtained histologically. It is evident from this observation that the fluid is not sealed within the middle ear, but is able to escape, either along the relatively wide Eustachian tube of the child or backwards through the aditus into the mastoid antrum.

Continuing with this line of thought, it might therefore be possible for the condition of cholesterin granuloma to exist in the mastoid of an ear in which the drum was consistently of a normal colour on examination. If this were so, it would be natural to expect that, if a number of mastoids were to be opened in children with a similar history and similar radiological appearances to those described, but with normally coloured tympanic membranes, one should be able to demonstrate black cells and cholesterin granuloma in a significant percentage of them. Ranger, indeed, quoted such a case of secretory otitis media in which cholesterol clefts were found histologically, but in an amount less than in cases of hæmotympanum.

During the past two years I have operated upon twenty children with histories of recurring attacks of pain and deafness in one ear, with radiological evidence of opacity of the mastoid cells and break-down of the intercellular walls in the same side, but whose drums, on repeated examinations have been entirely devoid of any blue discoloration. Space will not permit of a detailed description of each case, and, indeed, such descriptions would be largely repetitive. Two may be cited as typical examples:

Douglas S., was seen on 27.4.54, aged 5, with a history of recurring attacks of bilateral otitis media and deafness, treated by his own doctor with oral penicillin. His tonsils were small and slightly congested, and he had enlarged cervical glands. His nose was clear. Both drums were indrawn, but of normal colour. He could only hear a whisper at 1 foot in either ear. On 10.5.54 the Eustachian tubes were inflated, and were found to be patent, and his adenoids were removed. Six months later he was seen on account of further bilateral otitis media and deafness. The right drum was acutely indrawn and congested and the left drum was normal. On 18.2.55 his tubes were again inflated with ease, and his

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tonsils and some adenoid tissue were removed. A year later he again reported on account of several further attacks of right-sided otitis media. Again, the right drum was acutely indrawn and congested and an X-ray of the mastoids showed diploic processes with cellular break-down in the right side. On 30.3.56 the *right mastoid* was opened and the cells were found to contain clear, faintly yellow, tenacious mucus. There was an area of cellular necrosis from the tip to the antrum. *Bacteriology* showed that the fluid was sterile. *Pathological investigation* revealed œdematous mucosa infiltrated by lymphocytes and plasma cells, but with scanty polymorphs. No features of cholesterin granuloma were present. Eighteen months later, on 28.11.57, he had had no further trouble with the right ear, the right drum was only slightly indrawn, and he heard a whisper at 16 feet in this ear.

Sadie G., had her tonsils and adenoids removed elsewhere in 1952, when she was aged 4. She was examined on 21.10.54 with a history of colds, catarrh, earache and deafness. Her throat was clear but her nose contained mucopus. Both drums were indrawn and healthy in colour. X-ray showed both antra to be opaque. On 1.11.54 a double proof puncture yielded no pus, and some adenoid remains were removed. On 5.5.55 she reported back with a similar history, and the clinical examination was unchanged. X-ray on this occasion showed an opaque left antrum, and this was punctured on 20.5.55, again yielding no pus. She was examined on 30.1.56 on account of deafness and recurring pain in the left ear. The left drum was congested and indrawn and an X-ray showed blurring of the cells of the left mastoid. On 6.5.56 the *left mastoid* was opened, and the cells were found to be filled with thick, sticky mucopus, which was sterile. Pale, adherent granulations were present in a necrotic area extending from the tip to the antrum. The antral mucosa was thickened, and, on opening the attic, a healthy incus was found embedded in thickened mucous membrane. *Pathological* features were identical with the former case. On 11.7.57 her mother reported that Sadie was in much better health, her school work had improved, and her hearing had greatly increased. Both drums were slightly indrawn and of a normal colour.

In none of this type of case has a single black cell been found in the mastoid process. In none has there been any suggestion of cholesterin granuloma in the histological sections. All have shown evidence of infiltration of the mucosa by lymphocytes, polymorphs and plasma cells, while in some cases fibrosis has been noted. The picture is that of a chronic non-specific inflammation.

Thus, in spite of the similarity of the history, in spite of the similarity of radiological appearances, in spite of the mastoid necrosis involving the same groups of mastoid cells, the two conditions of hæmotympanum and secretory otitis media are different. I am persuaded, however, that the ætiology of the two conditions must be closely allied,

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because the only difference is the presence of blood in the one, and not in the other.

Robison (1942) investigated catarrhal otitis media and mastoiditis with effusion, and divided it into two types. The first is due to some demonstrable lesion in the upper respiratory tract, correction of which brings relief of the aural symptoms. The second type does not appear to be associated with upper respiratory tract infection. The tympanum is entirely or partly filled with fluid which may be seen through a retracted membrane. According to Robison this membrane has a yellowish or bluish colour depending on the colour of the fluid in the tympanum. The Eustachian tube is usually partly closed on inflation. Investigation of the total protein in the fluid aspirated from the middle ear convinced him that it was a transudate and not an exudate. He advised mastoid surgery, if other treatment fails, on account of an impending loss of hearing, and with this opinion I am in entire agreement.

Cody (1941) described four cases of catarrhal oto-mastoiditis very similar as to operative appearances and histological findings to the twenty on whom I have operated. He found that their Eustachian tubes were patent, and concluded that "it was obvious that the middle ear and Eustachian tube were the passageway, not the place of origin, of the mucous secretion". My experience in these secretory cases, which come to mastoid operation, leads me to support this opinion. The source of the fluid lies in the mastoid cells, and not the middle ear or tube.

Secretory otitis media, like hæmotympanum, has become much more common in the past fifteen years, and Eagle (1946), commenting on a series of 172 cases seen in the previous four years writes, "We have no definite causative factor for the increased frequency of the disease . . . It is possible that we are dealing with a virus disease; since the disease is almost in epidemic form, there is reasonable ground for this suspicion. We have been unable to correlate the use of sulphonamides in recent years with the disease . . . We do not admit that the disease is necessarily associated with upper respiratory disease. We are reluctant to admit that the disease had been prevalent prior to 1941, and that our eyes are now seeing pathology which they had previously failed to see."

Bateman (1957), reporting 67 cases of secretory otitis media, found that in extreme cases the drum appeared blue (presumably the gun-metal blue noted by Sheehy and McKibben). He found that the fluid from the middle ear was sterile, and was of the opinion that it was a secretion and not a transudate, an opinion which conflicts with that of Robison. He also noted that puncture-aspiration through the drum could be performed with little or no surface anæsthesia.

In secretory otitis media, as in hæmotympanum, the source of the fluid appears to be the mastoid cells. Why does it not drain away? If the Eustachian tubes were blocked in every case it would be easy to under-

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stand this, but Cody asserted that they were patent, Robison stated that they were partly blocked, and Bateman found that, while his patients could not clear their own tubes, they could be inflated by Politzerization. The majority of opinion in hæmotympanum, as we have seen, is that the tubes are patent.

Is it that the fluid is produced in too great a quantity? If one performs a cortical mastoid on a patient with an intact drum, and closes the post-auricular incision completely, the blood within the mastoid cavity will clot, but some of it must have entered the middle-ear cavity and Eustachian tube. Therefore, the drum should appear blue at the end of the operation, and possibly for a day or two thereafter. Yet it is uncommon to find a blue drum when the mastoid is dressed a week after operation, and such blueness quickly vanishes. I have invariably closed completely the incisions of those mastoid operations performed for hæmotympanum and catarrhal otitis media, and I have never seen a drum appearing blue on the patient's discharge from hospital 10–14 days later. One must, therefore, assume either that the operation has allowed the tube to open, or that the Eustachian tube, whether partly blocked or not, can drain from the middle ear and mastoid cavities the fairly considerable volume of blood produced at operation.

The one noteworthy feature of the fluid in the mastoid cells at operation for secretory otitis media or hæmotympanum is its viscosity, and it may be that this quality hampers normal drainage through the Eustachian tube in these cases. Once removed at mastoid operation, it must not re-form, or the condition would surely recur.

It seems reasonable to suppose that something might be learned about these conditions from microsurgery if the drum is reflected as for a mobilization of the stapes, but Sheehy and McKibben's third case, in whom the drum remained blue after operation, was so examined and the middle-ear cavity and tubal entrance were found to be healthy.

Ætiology of hæmotympanum still remains uncertain in the typical case in which angioma, fractured skull, jugular bulb abnormality, or blood dyscrasia can be excluded. Should Eagle's suggestion that a virus may be responsible for secretory oto-mastoiditis be correct, it is possible that a virus which stimulates a hæmorrhagic response in the tissues may account for hæmotympanum. The virus of influenza springs to mind as a possibility, as it is associated with hæmorrhagic bullae on the drum, with epistaxis, and even, in the recent epidemic, with spontaneous septal hæmatoma. Such a virus infection would not be fully controlled by penicillin, although this antibiotic would prevent invasion by secondary pathogens, which might account for the sterility of the fluid. Repeated exacerbations from upper respiratory tract infections might be responsible for the mastoid necrosis which develops.

It is easier to refute theories than to propose new ones, but I think

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that one may discount the view that the source of the blood is either the Eustachian tube or middle-ear cavity. One may also, on the evidence of published literature, discard the idea that a blocked Eustachian tube is responsible for the condition. The mechanism is not that of acoustic barotrauma. Nor, as far as can be ascertained, can an abnormal serum cholesterol be demonstrated. It is a confession of ignorance that the term "idiopathic" must still be applied to the condition.

Prognosis

One is left to contemplate the prognosis had mastoid surgery not been undertaken. Would the necrotic area in the mastoid process heal, or would the condition progress slowly? In 1956 I reported a series of 21 children in whom cholesterin granuloma (then mistakenly called black cellular cholesteatosis) was present in the mastoid cells. Two of these cases showed no perforation: four had developed a postero-marginal perforation with granulations: fourteen showed a perforation of the membrana flaccida with the formation of granulations or a polypus: while one patient had a large polypus, the exact site of origin of which was not determined. Of these 21 children, frank cholesteatosis, with typical histology, was present in ten. One of these children was the older sister of Case III in the present series.

The most advanced pathology of the cases here reported was found in Case VI, which showed a blue drum with an anterior scar and a crust over Shrapnell's membrane. The incus was eroded and was lying loose, and from the attic was removed a large granulation, section of which revealed the typical features of cholesterin granuloma. It appears to be within the bounds of possibility that, if this mastoid had not been operated upon and the granulation removed, perforation of the membrana flaccida and eventual immigration of squamous epithelium with the production of cholesteatosis might have taken place.

It is too great a coincidence to ignore that black cells and cholesterin granuloma may be present with an intact blue drum and also with perforation and cholesteatosis. I am convinced that mastoid operation on these children not only cures the condition and improves the hearing, but may prevent perforation in Shrapnell's membrane, or near the drum margin, with all its consequences.

Summary

The condition of hæmotympanum may not be as uncommon as would appear from a review of the literature. Eight new cases are reported, six of which have been seen in children in the past year. The similarity of their histories, clinical features, radiological findings, operative appearances, and pathological reports classifies them as forming a clinical entity.

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All have been cured by cortical operation in preference to myringotomy.

Many conditions may cause a drum to appear blue, but not all are associated with radiological mastoid opacity, and cellular breakdown with black cells in the mastoid process, and with histological evidence of cholesterin granuloma.

The various ætiological theories are examined. The source of the blood is the mastoid process. Comparison with secretory catarrhal otomastoiditis leads one to suspect a common ætiology, and, if a virus infection is a possible cause of this condition, a virus resulting in a hæmorrhagic response may account for hæmotympanum.

It is suggested that, if the condition is not treated surgically, cholesterin granuloma may form in granulation tissue in the attic, and lead to perforation, immigration of squamous epithelium, and attic cholesteatosis.

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