# IDIOPATHIC HÆMOTYMPANUM

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IT is not uncommon to find blood in the middle ear but there is usually an obvious and adequate explanation of its presence. Occasionally blood is found in the middle ear for no apparent reason and it is with this condition that this paper is concerned.

In 1929 Shambaugh[1] discussed the various causes of a blue discoloration of the tympanic membrane and reported seeing three cases due to the presence of a chocolate-coloured fluid in the middle ear not associated with any infection and occurring without any obvious cause. He gave details of two cases, one aged 55 and the other 52. This appears to be the first reference to the condition and since then very few cases have been reported.

Fowler[2] mentioned one case but gave few details.

In 1941 O'Donnell[3] reported two cases in children who were cured by the removal of their tonsils and adenoids. He suggested the title of idiopathic hæmotympanum for the condition.

In January, 1948, Kler[4] reported one case in a girl aged 17. The drum was still blue but the middle ear free from fluid at the end of treatment which included a myringotomy on two occasions.

These seem to be the only references in the literature to this condition, although other cases have occurred. Mr. J. F. Simpson[5] has very kindly supplied details of a case which he treated in 1944.

In view of the rarity of this condition the following two cases are reported. They were recently treated in the Middlesex Hospital by Mr. J. P. Monkhouse.

## CASE I

S. S., a girl aged 9, was first seen in February, 1948. She had had a catarrhal otitis media on the left side at the age of four and had complained of some pain in that ear at times since then. One month before she was seen she had had a sore throat and discomfort in the left ear. There was no history of any trauma nor of any abnormal tendency to bruising or bleeding.

On examination the left tympanic membrane appeared a deep blue colour but was otherwise normal. There was a conductive type of deafness present on that side. The right ear was normal. She had a large mass of adenoids. X-rays showed that both mastoids were cellular but the left appeared less well pneumatized than the right. The cells on the right side appeared normal but those on the left were hazy.



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She was admitted to hospital in March and under an anæsthetic the middle ear was aspirated through the postero-inferior portion of the tympanic membrane. Some thick, mucoid, dark-brown fluid was withdrawn and following this the posterior portion of the drumhead returned to a normal colour. The needle was reinserted in front of the handle of the malleus and further aspiration cleared the anterior portion of the middle-ear cavity. In all 0·5 c.c. of fluid was aspirated. Eustachian catheterization was attempted at the same time but no air was heard to enter the ear.

The fluid which had been aspirated was found to contain debris and scanty cells in the proportion of polymorphs 35 per cent., lymphocytes 40 per cent. and epithelial cells 25 per cent.: no organisms were seen and the fluid was

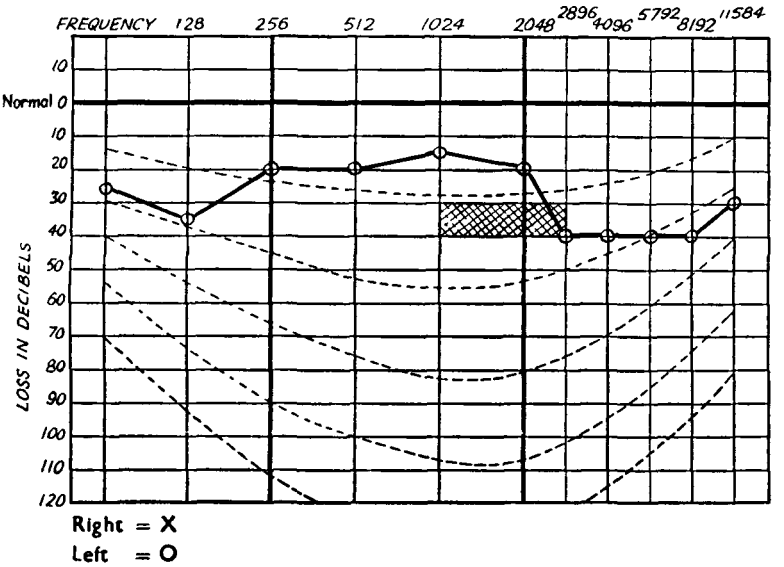



FIG. 1.
Audiogram of Case 1 before mastoidectomy.

sterile on culture. The Van den Bergh test gave a fairly strong indirect reaction.

The day after aspiration the tympanic membrane had returned to its original blue colour and remained unaltered during the next week while the patient was treated by inhalations and by argyrol and adrenaline nose drops. The aspiration was then repeated and 0·4 c.c. fluid obtained with similar results. This time only one puncture, below and behind the umbo, was necessary to clear the whole tympanic cavity. On eustachian catheterization a little air was heard to enter the ear and examination showed some bulging of the posterior portion of the drum with partial return of the blue colour. A large pad of adenoids was removed.

A week later the aspiration was repeated, yielding 0·6 c.c. fluid. Eustachian catheterization gave a good air entry with bulging of the posterior



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portion of the drum. After eustachian catheterization had been completed it was found that the middle ear was again full of fluid and further aspiration yielded another 0·6 c.c. The aspiration and eustachian catheterization were repeated on three further occasions at weekly intervals with similar results but without any permanent effect on the fluid in the middle ear or any improvement in the hearing.

As no benefit had been obtained from the above measures spread over six weeks and as X-rays showed that the mastoid cells were hazy the mastoid was explored. It was more cellular than appeared from the X-rays and there was no evidence of any infection. The cells at the periphery were full of a solid brown debris while those nearer the mastoid antrum contained a thick brown fluid. After the contents had been removed the cells appeared normal to the naked eye. A cortical mastoidectomy was performed and the mastoid cavity drained through the posterior meatal wall.

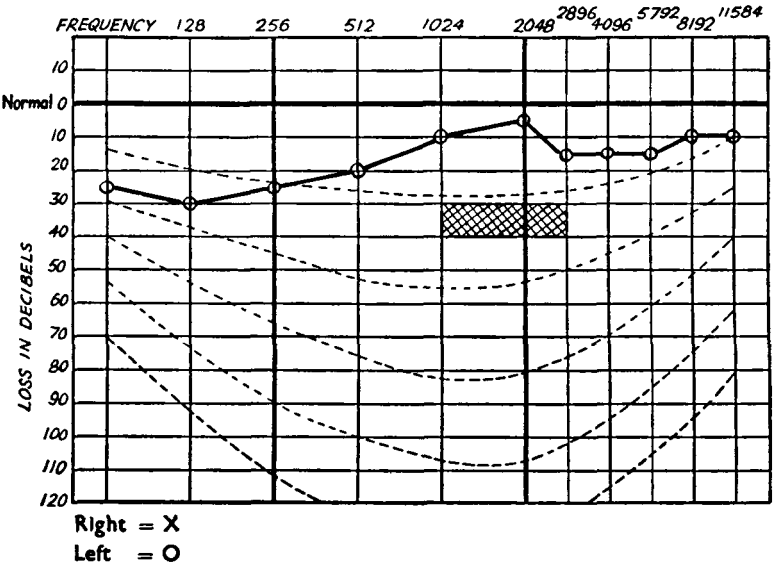



FIG. 2.
Audiogram of Case 1 in January 1949, 9 months after mastoidectomy.

Two pieces of bone which were sent for histological examination were reported on as follows: " (B.S.I. Surg. Hist. 1038/1948.) Section shows bone containing air cells. There are areas of cholesteatomatous tissue present with cholesterol crystal spaces surrounded by foreign body giant cells."

Her post-operative course was completely satisfactory and there has been no return of the blue colour. When the patient was last seen in January, 1949 (nine months after operation) the tympanic membrane was a normal colour without any blueness. Her hearing had improved considerably, particularly in the higher frequencies, and her audiogram at that time is shown in Fig. 2.



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While the patient was in hospital various laboratory examinations were carried out but no abnormality was found. The platelet count was 500,000, the clotting time and bleeding time normal and vitamin C excretion normal.

CASE II

A. M., a man aged 22, was first seen in January, 1948, on account of deafness and tinnitus which had been gradually increasing for about five years. In addition he had had short attacks of vertigo. He heard better in a noisy than in a quiet place. There was no history of any trauma or of any tendency to bleed excessively or bruise easily. The right tympanic membrane was a deep

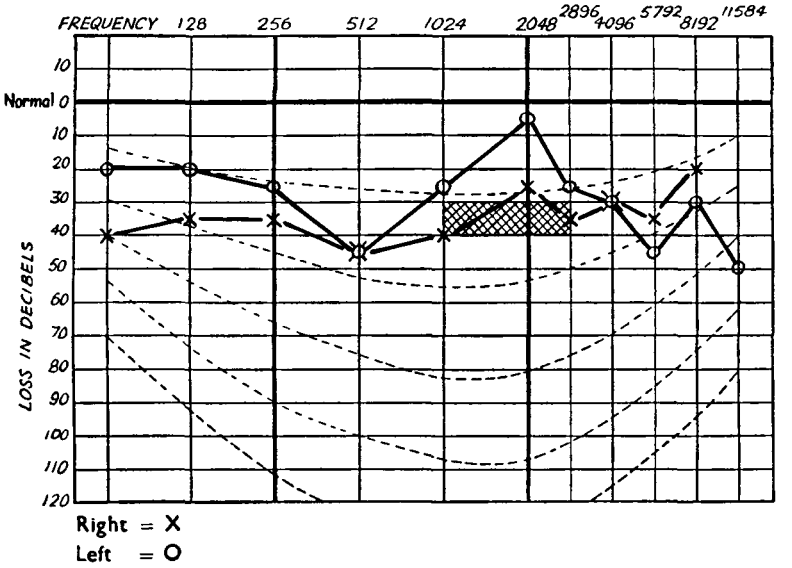



FIG. 3.
Audiogram of Case 2 on admission to hospital.

blue colour but otherwise normal. The left tympanic membrane was normal. There was a moderate degree of deafness of the conductive type present on both sides. Weber's test was lateralized to the right. An audiogram showed a loss of 30 to 50 decibels throughout the scale and little difference between the two ears (Fig. 3). The patient had an obvious facial asymmetry, which was a congenital abnormality, and X-rays showed a marked enlargement of the glenoid fossa on the left side with diminution in the size of the tympanum. The left ascending ramus of the mandible was very poorly developed. Both mastoids were comparatively acellular but there were less cells on the right and the middle ear was opaque on that side.

The patient was admitted to hospital in March and the right middle ear aspirated under an anæsthetic. A dark brown fluid was obtained similar to that in Case I. Following the aspiration the right eustachian tube was catheterized and air was found to pass easily into the ear. This procedure was



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repeated a week later as the fluid had re-accumulated almost immediately. Following this the middle ear was aspirated and the eustachian tube catheterized daily for a period of a week. This was carried out in the ward without any anæsthetic, either local or general, and caused the patient no discomfort. The middle ear was cleared by every aspiration but by the following day it had refilled again. The amount of fluid varied and on some occasions there was only a trace present. In this patient the fluid reappeared more slowly than in the first case.

During the next three weeks aspiration and eustachian catheterization were repeated at various intervals but at the end of that time there was still fluid in the middle ear. In view of this and of the findings in the previous case the mastoid was explored. It was small and acellular. The mastoid antrum contained a quantity of brown debris some of which was sent for section along with pieces of mucosa. A cortical mastoidectomy was performed and the mastoid cavity was drained through the posterior meatal wall.

The histological report was as follows: "(B.S.I. Surg. Hist. 1135/1948.) Section shows granulation tissue containing numerous cholesterol crystal spaces and foreign body giant cells together with granules of hæmosiderin."

A month after operation there was a return of a little fluid but this was cleared by one aspiration and has not returned since. When seen at the end of December, 1948 (eight months after operation) the drum was a normal colour except for a very small area posteriorly where there was a bluish tint in the tympanic membrane. An audiogram showed practically no change from the original in either ear (Fig. 4).

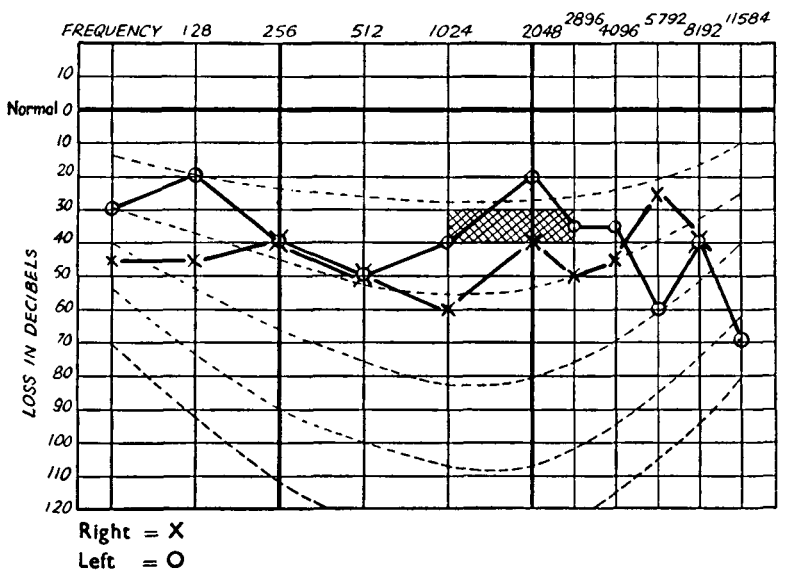



FIG. 4.
Audiogram of Case 2 in December 1948, 8 months after mastoidectomy.



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Investigations while the patient was in hospital showed a normal bleeding and clotting time and a platelet count of 200,000. Vitamin C excretion was somewhat diminished. Blood cholesterol was 336 mgm. per 100 ml.

### Comments

The positive Van den Bergh reaction on the fluid aspirated from the middle ear and the presence of granules of hæmosiderin in the sections of the mastoid confirm the fact that the fluid filling the middle-ear cleft is essentially altered blood.

In Case I there was a collection of altered blood in the left middle-ear cleft but the other ear was normal. Although the tympanic membrane was not indrawn there was some evidence of eustachian obstruction and it was only on the third inflation of the eustachian tube that air entered the ear freely. This obstruction may have been due to the large pad of adenoids which was present, but their removal did not produce any improvement in the condition of the ear. X-rays showed a cellular mastoid with haziness of the cells and at operation the mastoid cells were found to be filled with debris and the same type of fluid as filled the middle ear. The presence of fluid in the cellular mastoid would easily account for the rapid re-appearance of the fluid after aspiration and the inability to clear the condition by any procedure other than mastoidectomy.

Case II presented other abnormalities as well as the blue drum and there was a bilateral deafness even though there was fluid on one side only. In this patient the mastoid was not cellular and the space available as a reservoir, as seen at operation, did not seem sufficient to contain the total volume of fluid which was withdrawn during treatment.

In both cases there was no evidence of any infection. Culture of the fluid was always sterile and infection did not supervene in spite of the repeated aspirations.

Laboratory investigations in both cases showed no evidence of any disorder which would produce an increased tendency to bleed.

In both these cases, as in all the other reported and known cases, the condition was unilateral. In such a small number of cases it is impossible to attach much importance to this fact but it is worth recalling that the clear serous effusions in secretory otitis media are often bilateral. Hoople and Blaisdell[6] found that this condition was bilateral in 44 per cent. of their series of 748 cases, and Eagle[7] reported 172 cases of which 30 per cent. were bilateral. Otitic barotrauma also is bilateral in about 25 per cent. to 50 per cent. of cases[8].

The histological appearances in both cases were characterized by the presence of cholesterol crystal spaces surrounded by foreign body giant cells. There are three possible sources of the cholesterol.



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(a) *Deposition from the blood stream.* This is the least likely possibility. Cholesterol is not usually deposited in normal tissues although deposits may sometimes occur in the spleen and other parts of the reticulo-endothelial system in conditions in which there is a raised cholesterol content of the blood. Although the blood cholesterol in Case II was above the figures usually given as the upper limit of normal, the difference is not great and it is difficult to attach any real significance to it.

(b) *Degeneration of the epithelium.* The evidence is against this because, at operation, after the blood and debris had been removed from the cells the lining epithelium appeared normal and in addition the histological examination showed no epithelial degeneration.

(c) *From the fluid filling the middle-ear cleft.* The laboratory investigations carried out in these two cases confirm the fact that the fluid is essentially altered blood. Cholesterol is an important and insoluble constituent of blood and may crystallize out in cases of hæmorrhage. Accordingly the fluid filling the middle-ear cleft must be regarded as a very likely source of the cholesterol found in the histological sections. However, Professor R. W. Scarff is of the opinion that the cholesterol present could not have resulted from a single hæmorrhage and suggests that there might have been repeated hæmorrhages. This suggestion is in keeping with the finding in Case II that the fluid continued to refill the middle ear after repeated aspirations in spite of the fact that the mastoid was small and did not appear large enough to act as a reservoir from which the middle ear could continually refill. It seems likely that in this case, as in others, several hæmorrhages occurred. Shambaugh reported that X-rays of his second case showed complete absence of pneumatization of the mastoid process and yet he failed to cure the condition by repeated incisions of the drum.

Recently a patient with secretory otitis media was treated by mastoidectomy as she failed to clear after repeated aspirations spread over a long period. In this case also histological examination showed cholesterol crystals in the mastoid but the amount was less than in the two cases with hæmotympanum reported above. This is in keeping with the fact that the cholesterol in serum is in a more soluble form than that in the red corpuscles and is, therefore, less likely to crystallize out from serum than blood.

## Discussion

*Etiology and Pathology.* The direct cause of this condition is a collection of altered blood in the middle-ear cleft but the cause of the hæmorrhage is obscure.

Shambaugh and O'Donnell both stressed the presence of eustachian obstruction and O'Donnell suggested that the hæmorrhage was the result of a " hyperæmia ex vacuo ". Although eustachian obstruction



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may be a factor in some cases it does not seem to be the essential cause of the condition and it is considered that eustachian obstruction alone would be insufficient to produce a hæmorrhage into a normal middle-ear cleft. There are three reasons for making this statement :

(1) Eustachian obstruction is very common but this condition is rare.

(2) Eustachian obstruction has not been present in all cases and it was not evident in Case II reported above.

(3) The most definite cause of " hyperæmia ex vacuo " must occur in acute otitic barotrauma, but in that condition hæmorrhage into the middle-ear cavity occurs in only a small proportion of cases and is always accompanied by other signs such as congestion of the drum and interstitial hæmorrhages. Teed's figures[9] show that in one series of 239 cases of otitic barotrauma there were less than 9 with blood in the middle-ear cavity. Dickson, McGibbon, Harvey and Turner[8] found only one hæmotympanum among 89 cases of acute otitic barotrauma occurring in 1,000 cadets tested in the decompression chamber. Also, in otitic barotrauma experimentally produced in cats, Dickson, McGibbon and Campbell[10] found that congestion and mucosal hæmorrhages were present in all cases having a hæmotympanum. The ears showing the most marked hæmorrhages into the middle-ear cleft and the least mucosal hæmorrhages occurred in cats subjected to an " explosive descent " in which the external pressure was increased from 350 mm. Hg to 760 mm. Hg in one second. This seems significant as these conditions are the ones most unlike those which would occur as a result of eustachian obstruction in patients not subjected to abnormal changes in external pressure.

It is considered, therefore, that eustachian obstruction alone is not the cause of the hæmotympanum in the patients we are considering even though no alternative explanation of the condition can be given.

All the reported cases have not responded to treatment in the same way. In some the condition was cured comparatively easily while in others the fluid persisted in spite of repeated incisions of the tympanic membrane or aspirations of the middle ear. As it has been suggested above that there were several hæmorrhages in the cases reported here, this must be considered as one explanation of the difficulty of clearing the fluid in some cases. However, it seems very doubtful if any case would be cured easily if the mastoid were filled with blood even from a single hæmorrhage. It is considered, therefore, that the extent of the fluid must vary in different cases and this might well depend on the site of the bleeding.

If a hæmorrhage occurred in the middle ear or eustachian tube the mastoid might not be filled and in that case there would be only a small amount of fluid present which could be cleared fairly easily. On the other hand a hæmorrhage occurring in the mastoid would be much more



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difficult to clear by aspiration of the middle ear and it would be practically impossible to cure the condition by that procedure if the mastoid were large.

It seems, therefore, that O'Donnell's two cases and Kler's case were cured fairly easily because the mastoids were not filled. Although there is no record of an X-ray of the mastoids in those cases to confirm this, Kler considered that in his patient the bleeding occurred from vessels in the tympanic membrane and it is probable that the mastoid would not fill in such a case.

In the two patients reported here it is considered that the hæmorrhage probably occurred in the mastoid itself for it seems less likely that it would be filled as a result of bleeding in the tympanic cavity or eustachian tube. This suggestion is also supported by the results of treatment. However, at operation, no abnormality was found in the mastoid to explain the condition.

*Treatment.* O'Donnell's cases were both cured by removal of the tonsils and adenoids. Kler's case was cured by myringotomy on two occasions and by eustachian inflation. In the other cases the condition failed to respond to these measures and a possible explanation of this has been given above. The two cases reported here failed to respond to repeated aspiration but have remained clear for over eight months following mastoidectomy. In view of the suggestion that there were several hæmorrhages before operation it seems probable that mastoidectomy removed the source of the bleeding as well as removing the debris and fluid which filled the mastoid.

The important aspect of treatment is the effect on the hearing, and in Case I there was a considerable improvement following mastoidectomy. It seems most unlikely that such a result would have been obtained by leaving the fluid or by continuing the aspirations indefinitely. In Case II a marked improvement was not to be expected in view of the other abnormalities which were present.

It is possible that mastoidectomy might not cure some cases. If there were recurrent hæmorrhages from the middle ear the source of bleeding would be unaffected by the operation and the condition might recur after it. However, it seems impossible at present to determine the site of the hæmorrhage and it is considered that a mastoidectomy is indicated if the condition fails to respond to more conservative measures and if X-rays show mastoid involvement.

**Summary**

Two cases are described in which there was a blue appearance of the tympanic membrane as a result of altered blood in the middle-ear cleft which had occurred for no apparent reason.

The condition is uncommon and only seven other cases appear to have been reported. All the cases have been unilateral.



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The etiology of the condition is obscure but eustachian obstruction does not appear to be the essential cause.

In the cases reported here there was some evidence that there were recurrent hæmorrhages and that the site of bleeding was in the mastoid.

If the condition fails to clear as a result of repeated aspiration (or myringotomy) and if X-rays show mastoid involvement it is considered that mastoidectomy is indicated. Both cases reported here have remained clear for eight months since mastoidectomy. In one case audiograms show a considerable improvement in hearing as a result of operation. The other case was complicated by other abnormalities and a marked improvement in hearing could not be expected.

I wish to express my thanks to Mr. J. P. Monkhouse for permission to report these two cases and for much information about this and similar conditions, and to Professor R. W. Scarff for information about the pathology. I also wish to thank the Bland Sutton Institute of Pathology for the histological and bacteriological investigations, the Courtauld Institute of Bio-chemistry for the biochemical examinations, Dr. H. Graham Hodgson and the X-ray Diagnostic Department of the Middlesex Hospital for the X-rays, and Miss P. McCloughry and Miss N. C. Gatehouse of the Ferens Institute of Oto-laryngology for the audiograms.

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