RHINITIS CASEOSA

By ALEX. R. HARPER (Glasgow)

RHINITIS CASEOSA is an uncommon, if not rare, condition, and the cases which have been described in the literature have been reported either as isolated instances or in very small series. Few examples may be seen in a lifetime, even by those with wide experience, and for this reason the case reported here may be of interest.

The condition is characterized by the formation of masses of caseous material in the nose and sometimes in the maxillary antrum, though it appears that the antrum is rarely if ever involved alone. In fact, there is only one possible case of this reported in the literature (Cartaz, 1908). As a rule only one side of the nose is affected, there being no more than three cases described with bilateral involvement (Nyulasy, 1892; Schleicher, 1890; Laocarret, 1898). The condition is accompanied by nasal discharge which is often fœtid, and in the later stages by intranasal and extranasal deformity due to destruction and expansion of the tissues by pressure of the caseous mass.

Historical

Rhinitis caseosa was first described as a distinct clinical entity by Duplay in 1868, though the date is more often reported as 1874, when he correlated the reports of five cases described in the literature in the previous 20 years. A few isolated cases were described over the next 20 years, and these were reviewed by Potiquet in 1889. Reports of several series of cases, usually very small in number, followed thereafter from time to time, principally during the last decade of the nineteenth century and the earlier years of the twentieth. The literature on the subject was reviewed in 1936 by Meyersburg, Bernstein and Mezz, and again an excellent review with a very full and comprehensive bibliography was produced by Polson in 1942, analysing the reported cases. This last exhaustive review of the literature revealed information of some kind concerning about 158 cases, including those only briefly mentioned by various clinicians in discussion, but detailed reliable records were available of only 75 cases. Since then the only reference to the subject seems to have been by Farrior in 1942, who described one of the only three cases presented in the American literature.

Aetiology

Various theories have been postulated from time to time to account for the condition, the principal ones being as follows:

https://doi.org/10.1017/S0022215100047800 Published online by Cambridge University Press

1. *Tuberculosis.* There was thought to be a causal relationship between pulmonary tuberculosis and the pressure of the caseous material in the nose, the caseation being supposed to be due to decay of the nasal epithelium arising from infection by tubercle bacilli. Support was lent to this theory by the description by Cozzolino (1884) of an organism resembling the tubercle bacillus in the caseous material.

2. *Syphilis.* The condition was also considered to be in some way connected with tertiary syphilis and the destruction caused by the formation and breaking down of gummata.

3. *Erysipelas.* It was suggested by Duplay (1868) that extension of erysipelas from the face to the nose caused epithelial desquamation with the consequent collection of a mass in one of the nasal passages, a theory which was evidently founded on the coexistence of the two conditions in one case, and which received little support.

4. *Cholesteatoma.* The condition was later thought by Cozzolino (1889) to be analogous to aural cholesteatoma, and for several years it was referred to under this description. Histological and chemical examination by Kelson (1904) showed definite differences between the two conditions, notably the absence of any cholesteatomatous membrane or the typical laminae and the absence of cholesterin from the nasal material, but the theory retained its popularity for the first two decades of this century.

In supporting the cholesteatoma theory, Heimendinger (1907) and Winkler (1910) each separately described the condition as arising from two independent causes. The first, " true cholesteatoma ", was held to be a developmental anomaly of fœtal origin where some disturbance of the normal embryonic growth caused proliferation of epidermis into the bones of the face. This later acted as a nidus for the formation of a cholesteatomatous tumour, deep seated and involving the overlying bone, which was assumed to be a factor in causing subsequent suppuration.

The second type, " false cholesteatoma ", was thought to be a product of existing sinus suppuration, and to be due to the encroachment into the sinus cavity of diseased atrophic nasal mucosa which had undergone metaplasia to the squamous type. It was assumed that the squamous epithelium, meeting the inflamed ciliated epithelium of the sinus cavity, was unable to unite with it and was thrown off and died, another layer soon taking its place. This condition of " false cholesteatoma " was said to be more superficial than the " true " type, involving only the mucosa and not the adjacent bone.

5. *Specific infection.* Infection by the strepthrix alba was believed by Massei (1896-7) to be the specific cause of the disease, and the presence of the organism was demonstrated by several other observers, but the fact that many other workers failed to find the streptothrix in material from their cases suggested that in cases where it was present the organism was a secondary invader.

https://doi.org/10.1017/S0022215100047800 Published online by Cambridge University Press

Other bacteria and fungi have also been demonstrated in the material from time to time, usually in single cases or in extremely small series.

6. *Polyp degeneration.* It was suggested by Bories in 1889 that the caseous material was the product of degeneration of mucous polypi in the nose, as he found similar appearances on microscopic examination of the caseous material and of polypi which had undergone degeneration. This theory met with little acceptance, however, and furthermore the presence of polypi is by no means an invariable accompaniment of caseous rhinitis in an analysis of the reported cases as carried out by Polson. Polypi were a prominent feature in the case described below, and acted as an obstruction which completely concealed the mass of caseous matter, and thus doubtless contributed to its collection.

7. *Suppuration complicated by obstruction.* The most generally accepted view today is that the phenomenon of caseous rhinitis is due to the retention in the nose and sinuses of the products of suppuration owing to some form of obstruction, under certain conditions, bacteriological and physical, which favour its inspissation. The process was described by Avellis (1900) as " Verkasung ", or metamorphosis of the secretion into a caseous mass, which he assumed to take place during the course of chronic sinusitis when the lining membrane recovered gradually till it was able to overcome the attacks of the infecting organism. The secretion contained in antrum, being prevented from escaping by some form of mechanical obstruction, became stagnant, lost its moisture, and fatty degeneration of the pus cells seemed to result, so that after a while the mass became more or less solid with the consistency of soft cheese.

The factor of obstruction by some means or other has been mentioned in a large number of the reported cases, though not always emphasized, as also has the presence of chronic suppuration in the nose. The suppuration has been described in several cases as being due to a foreign body or a rhinolith, or in others to chronic sinusitis. As is apparent from the various theories held in the past regarding the cause of the condition, it has not always been clear whether the suppuration constituted a causative factor or an effect of the caseous rhinitis.

Polson found that sinusitis was noted as being present in only 21 per cent. of cases, too low an incidence to permit the conclusion that it is an important factor, but it may be pointed out that in many cases the condition of chronic sinusitis could easily have been hidden and overlooked in the presence of the grosser phenomenon of the caseous mass. He also drew attention to the fact that sinus suppuration is common, and that therefore rhinitis caseosa should be commoner than it is if the two conditions have a causal relationship, but it must be remembered that in addition to suppuration and obstruction to the escape of secretions there must be other favourable conditions present for the development of rhinitis caseosa. Inspissation must occur, probably due to stagnation

https://doi.org/10.1017/S0022215100047800 Published online by Cambridge University Press

followed by evaporation of the moisture of the secretion by air currents, and presumably the bacteriological conditions must be exactly right. These favourable conditions are largely hypothetical, and the unlikelihood of all the necessary factors occurring together would account for the rarity of the disease.

The cause of mechanical obstruction to the secretion has varied in the cases which have been sufficiently fully described for this to be assessed. Polypi have been present in many cases, but by no means invariably. In other cases large septal crests or deflections have been blamed, or foreign bodies, granulation tissue, neoplasm situated in the anterior part of the nose, gumma, septal hæmatoma and perichondritis, hypertrophic changes in the mucosa, especially of the turbinates, have all been present to cause obstruction. In the case reported here there seemed to be no doubt but that the mass of polypi in the right ethmoidal region constituted a most effective obstructive factor.

Pathology

The morbid changes found in the nose are as various as the numerous conditions described above as giving rise to obstruction and suppuration.

The caseous material itself has been described as yellowish or grey, semi-solid, resembling the contents of sebaceous cysts, or likened to rotten cheese or putty. Its quantity varies greatly in different cases, and in most cases its offensive odour has drawn comment. Specimens examined histologically have been found to consist chiefly of pus cells and amorphous debris. Fatty cells and some crystals have occasionally been described, and a trace of cholesterin has been found on a few occasions.

The organisms discovered at different times have varied widely, and include streptothrix, fungi and other bacteria as already discussed under the heading of ætiology.

The pathological changes in the mucosa and structure of the nasal passages depend on the degree of suppuration which is present and on the duration and extent of the condition. They vary from lesser degrees of inflammatory infiltration and thickening in early cases to complete mucosal degeneration and destruction, while in the later stages of the disease the accumulation and expansion of the caseous mass leads to slow destruction by pressure of the adjacent soft tissues and bone resulting in external deformity and fistulæ.

Symptomatology

It is important to bear in mind that rhinitis caseosa is not a disease in itself, but is an incidental development in the course of nasal sepsis when other factors such as obstruction and inspissation of the secretions are also present. The history is most likely to be one of increasing unilateral nasal obstruction, often associated with offensive discharge which may

https://doi.org/10.1017/S0022215100047800 Published online by Cambridge University Press

either be purulent or serous. There may be frequent exacerbations of infection of the upper respiratory tract, but on the other hand the infection may be a low grade one and the general symptoms may be confined to vague complaints such as headache and lassitude arising from the chronic sepsis.

On examination of the nose the caseous material may be directly visible through the anterior nares, or it may be seen on posterior rhinoscopy obstructing the choanæ. In other cases the mass may not be immediately apparent owing to obstruction of the view by a septal deflection, hypertrophied turbinates or whatever abnormally caused retention of the secretion in the first place. In the case described below for example, the caseous material was entirely concealed from view by the mass of ethmoidal polypi, and was not discovered until these had been removed at operation.

In addition to the caseous material, nasal examination will reveal signs of chronic suppuration, whether arising from the paranasal sinuses or from some source in the nasal passages themselves such as a foreign body. A purulent or serous discharge will usually be found either in the nares or postnasal space, the nasal mucosa may be engorged or hypertrophied, and crusting may be present. Foetor of the breath appears to be a common accompaniment of the condition, though it is by no means invariable and it was not present in the case reported here.

In the later stages of rhinitis caseosa when the mass has caused extensive destruction by erosion various degrees of facial disfigurement may be seen, such as swelling of the face, broadening or collapse of the nose, external fistulæ near the inner canthus of the eye, or ulceration through the palate. Such extreme damage will not arise, however, unless the nasal condition has been neglected over a long period, and it is probably unlikely to be seen today. While the case with external deformity is usually one of long standing and slow development, however, it is not always so, and Farrior's case was remarkable for the comparatively rapid development of the whole condition. There was apparently only one year between the beginning of any nasal complaint at all, in the form of an attack of acute sinusitis which failed to resolve satisfactorily, and the development of noticeable external deformity which took the form of unilateral swelling and broadening of the nasal bridge, and which remained permanently after the disease had been dealt with.

Treatment

Once the caseous mass has been discovered it may be dealt with by simple means, such as scooping out with a spoon followed by repeated irrigation to ensure its complete removal, and from the published descriptions of several cases it seems that adequate removal of the mass and of any obvious obstruction in the nose is not followed by reaccumulation of

https://doi.org/10.1017/S0022215100047800 Published online by Cambridge University Press

the caseous material. In order to achieve a satisfactory cure of the condition, however, it must be remembered that the collection of material in the nose is only an incidental occurrence in the presence of some form of suppuration, and attention must be directed to carrying out the appropriate treatment for this, whether the trouble be in the nose itself or in the paranasal sinuses.

Case Report

MRS. A.B., aet. 30.

The patient was seen at the Ear, Nose and Throat Department of Glasgow Royal Infirmary, complaining of intermittent nasal obstruction of about a year's duration, and frequent attacks of purulent discharge. On examination, the right nostril was found to be obstructed by polypi growing from the ethmoidal region, while the left middle turbinate was distinctly œdematous and polypoid. There were a few fine streaks of muco-pus in her nasal passages and post-nasal space, and the posterior pharyngeal wall showed only very slight chronic inflammation. Radiological investigation of the paranasal sinuses showed complete opacity of the right antrum and distinct thickening of the lining mucosa of the left antrum. There was marked opacity of the whole ethmoidal region on the right side, the left ethmoid appeared to be clear and the frontal sinuses showed no abnormality.

Three weeks later, the antra were opened through the canine fossæ and the lining membranes of both were found to be in a state of marked polypoid degeneration. These diseased linings were stripped and removed and a nasal antrostomy was completed below each inferior turbinate. The ethmoidal regions were then explored and thoroughly cleared out both through the antra and through the nose. The right ethmoidal labyrinth was entirely replaced by a mass of polypi, while in the left ethmoid the slightly polypoid state of the middle turbinate suggested that it was the seat of a similar diseased state, though still in an early stage.

After removal of these polypi from the right ethmoid the nasal cavity in this region was found to contain a large amount of inspissated caseous material, greyish in colour and with rather a foul odour, which had not previously been visible. A quantity of this material was saved for microscopic examination along with a piece of the ethmoidal polyp. The caseous material was found to consist of eosinophilic structureless debris with some inflammatory cells in places, while the ethmoidal tissue had the structure of a simple fibroadenomatous polyp with much inflammatory change.

After dismissal from hospital the patient continued to attend for treatment as an out-patient at intervals for six months until the nose was brought into a completely satisfactory condition. The left ethmoidal region soon healed up firmly and cleanly with appropriate treatment, but the right ethmoid required unremitting careful attention to achieve a similar satisfactory result. It tended at first to become polypoid again, and for some time remained rather soft and boggy before it was induced to heal in a firm healthy fashion.

A year after operation, the patient was asked to report again to note any further development. Her nose had remained in a satisfactory condition both

https://doi.org/10.1017/S0022215100047800 Published online by Cambridge University Press

subjectively and on examination, and she felt that her general health and wellbeing had benefited considerably.

My thanks are due to Mr. Stephen Young, in whose wards the patient was treated, for permission to publish the details of this case.

REFERENCES

AVELLIS, G. (1900) *Arch. f. Laryng., Berlin,* **10**, 271.
BORIES, E. (1888) *Congrès Franç. de Chir.*, **3**, 595. Abstr. in *J. Laryng. and Otol.*, (1890), **4**, 23.
CARTAZ, A. (1908) *Traité de Méd. et de Therap.*, Paris, **27**, 96.
COZZOLINO, V. (1884) *Boll. delle Malatt. dell'Or*, **2**, 5.
——— (1889) *Ann. des. Mal. de l'Or.*, **12**, 645.
DUPLAY, S., and FOLLIN, E. (1868) *Traité Elémentaire de Path. externe*, Paris, **3**, 806.
FARRIOR, J. B. (1942) *Arch. Otolaryng., Chicago,* **36**, 238.
HEIMENDINGER, A. (1907) *Arch. f. Laryng., Berlin,* **19**, 381.
KELSON, W. H. (1904) *Brit. med. J.*, **2**, 1753.
LAOCARRET (1898) *Ann. Policlin., Toulouse,* **16**, 51.
MASSEI, F. (1896) *Arch. Ital. Laryng.*, **16**, 51.
——— (1897) *J. Laryng., Rhin. and Otol.*, **12**, 478.
MEYERSBURG, H., BERNSTEIN, P., and MEZZ, D. (1936) *Arch. Otolaryng., Chicago,* **23**, 449 ; (bibliography of 71 references).
NYULASY, F. H. (1892) *Aust. med. J.*, **14**, 332.
POLSON, C. J. (1942) *J. Laryng. and Otol.*, **57**, 160 ; (bibliography of 92 references).
POTIQUET (1889) *Gaz. Hof.*, **62**, 121.
SCHLEICHER, W. (1890) *Ann. d. Mal. de l'Or.*, **16**, 449.
WINKLER (1910) *Zeit. Laryng.*, **2**, 251.

https://doi.org/10.1017/S0022215100047800 Published online by Cambridge University Press