KERATOSIS OBTURANS

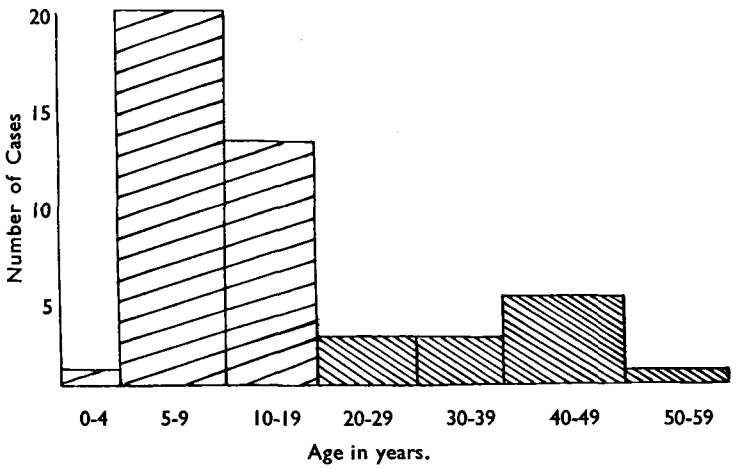
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THE purpose of this paper is to analyse a series of cases of keratosis obturans drawing attention to the association of this condition with bronchiectasis and sinusitis, and attempting to explain the relationship on an ætiological basis.

The series comprises fifty consecutive cases which had advanced to such an extent that general anæsthesia was necessary for the complete removal of the cholesteatoma-like accumulations. The presenting symptom, for which relief was sought, in 80 per cent. of the cases was pain; deafness, otorrhœa and tinnitus were the other complaints in order of frequency. In most there was expansion of the bony external auditory meatus with ulceration and the formation of granulation tissue, while, in a few, the disease had extended to involve the tympanic attic, the aditus and mastoid antrum. The facial nerve was exposed in one case, and the tympanic membrane, though often displaced, was usually found to be intact. The more extensive lesions were encountered in adults. Four patients had had keratotic "masses" removed on a previous occasion.

Analysis

Age Distribution. This figure shows that keratosis obturans is commonest in the first two decades. For future descriptive purposes the fifty patients have been divided into two age groups, viz. 35 Juveniles under 20 years and 15 Adults over 20 years.



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Sex Distribution. Male 29 cases (58 per cent.). Female 21 cases (42 per cent.).

Association with Bronchiectasis and Sinusitis.

TABLE I.

Association	Number of Cases	Number of Juveniles	Number of Adults
(*a*) Bronchiectasis	20	18	2
(*b*) Chronic sinusitis	10	9	1
(*c*) Nil	20	8	12
Totals	50	35	15

TABLE II.

Association	Per cent. of 35 Juvenile Cases	Per cent. of 15 Adult Cases.
(*a*) Bronchiectasis	51·4	13·3
(*b*) Chronic sinusitis	25·7	6·7
(*c*) Nil	22·9	80
Totals	100	100

Twenty patients have bronchiectasis confirmed by clinical examination and radiology. To date, three juveniles have had lobectomies performed and the remainder are being treated conservatively by postural drainage and breathing exercises. It is interesting to note that the majority of the patients with bronchiectasis are in the juvenile group (Tables I (*a*) and II (*a*)), and that chronic sinusitis has accompanied bronchiectasis in all but three of this group necessitating some form of surgical sinus drainage or repeated antral lavage.

Ten patients were found to have chronic sinusitis and again they were mostly juveniles (Tables I (*b*) and II ($\bar{b}$)). Early bronchiectatic changes were suspected clinically in seven of these children but not confirmed radiologically.

Twenty patients, the majority of whom were adults, had no clinical evidence of bronchiectasis or sinusitis (Tables I (*c*) and II (*c*)). It may be significant that one adult had congenital pulmonary stenosis.

Unilateral or Bilateral Keratotic Lesions.

The keratosis was bilateral in 22 and unilateral in 28 cases. It is significant that a definite relationship existed between the distribution of the bronchiectasis and the side or sides involved by keratosis obturans. Of the 20 bronchiectatic cases, in 15 both lungs and both ears were affected and in 4 one lung and the ipsolateral ear. One exception only to this relationship was noted.

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Discussion on Aetiology

The condition was first described by Toynbee (1860), who considered it to be a molluscous tumour having its origin in the dermis of the meatus. He described one case and found ten of these "tumours" in 1,013 post-mortem dissections of diseased ears. In 1874 Wreden called the condition Keratosis Obturans. In 1933 Green presented three cases and reviewed the literature to that date. He considered, like Wreden, that the underlying factor was a persisting hyperæmia of the skin of the external meatus and this view is currently held in most textbooks of otology. According to Green, Scholefield described cholesteatoma of the canal due to a foreign body; Schwartze thought the process of formation was initiated by seborrhœa of the ceruminous glands; Bezold described a cast of the meatus in association with otomycosis; and Politzer, who agreed with the chronic hyperæmic theory, qualified this by stating that it regularly accompanied inflammation of the tympanic cavity, parotid gland or structures adjacent to the ear.

During the last twenty years writers on this subject have expressed divergent opinions as to the ætiology. Altmann and Waltner described one case in detail and came to the conclusion that the formation of cholesteatoma in the external auditory meatus was the result of secondary ingrowth of stratified squamous epithelium of the canal into a cavity formed by sequestration of a superficial area of the bony wall. Circumscribed periostitis, occurring during one of the exanthemata, was considered the cause of this necrosis. Epidermis plugs were thought to be a sequel, rather than a cause, of bone necrosis. Mayer and Fraser, in their monograph on "Pathological Changes in the Ear in Late Congenital Syphilis", described sequestration of bone of the external meatus, inducing epidermal proliferation as part of the healing process and the consequent development of cholesteatoma. Costello and Fried published a case caused by chronic desquamative dermatitis. Biber, in a comprehensive review, discussed the ætiology and histogenesis. He presented three case histories and concluded that primary cholesteatoma of the external canal did not exist and was secondary in nature, mostly due to primary middle-ear inflammation. Tumarkin did not doubt the development of keratosis obturans in the external canal, even when the middle ear and attic were extensively involved. Gardère, Van Canaghem and Campos Peña (quoted by Biber) all found keratosis obturans accompanied by chronic sinuso-rhino-tracheo-bronchitis and blamed chronic Eustachian obstruction for initiating the cholesteatoma formation. Harpman (1951) noted that many patients with bronchiectasis and chronic sinusitis developed keratosis obturans, and later (Harpman, 1953) he expressed the opinion that the underlying cause was a defect of the mucous membranes of the sinuses, lungs and of the epithelium of the surface of the ear drum.

In this present series there were no patients with a history or evidence of past or present otitis externa, otomycosis, foreign body or syphilis. Two patients had a past history of acute suppurative otitis media in the unaffected ear and one of acute suppurative otitis media on the same side.

A relationship between keratosis obturans and bronchiectasis is a definite clinical entity, especially in the younger age group. It cannot be said that sinusitis is, in itself, significant, but associated with early bronchiectasis it

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assumes some import; 25 out of 35 juveniles (71 per cent.) have this common factor.

Three clinical observations are worthy of note in discussing the ætiology of these "masses". Firstly, all grades of severity are encountered from the so-called epidermis plugs consisting of a central core of cerumen with peripheral layers of desquamated squamous epithelium and minimal widening of the canal without ulceration, to gross local destruction by invasive cholesteatoma. Secondly, from a study of past hospital records in these patients and from the interval between recurrences emerges the fact that these keratotic "masses" may take many years to develop. The younger patients tend to have epidermis plugs and the adults more advanced disease. Thirdly, the well-known cough reflex induced by stimulation of the auricular branch of the vagus has been observed to be very active in these patients.

I suggest that the essential local predisposing cause is an excessive secretion of cerumen. The ceruminous glands are described as modified sweat glands and are histologically similar to apocrine glands. Their secretion may be expected to be activated by the sympathetic autonomic system, and these nerves, therefore, would be cholinergic in type. Collins concluded that this was probable when carrying out certain observations on the external auditory meatus. If the auricular branch of the vagus were secreto-motor to ceruminous glands then a connecting link would be established.

The hypothesis, therefore, envisages that the sequence of events starts with bronchial dilatation; the presence of pus stimulates vagal afferents from the tracheo-bronchial tree, producing reflex cough and reflex secretion of cerumen. Mechanical obstruction of the external auditory meatus follows and with the continued desquamation of surface epithelium an epidermis plug is formed. In time, pressure effects arise such as absorption of bone, expansion of the canal and ulceration. Once ulceration is established it is followed by inflammation and repair; granulation tissue is formed and epithelium, in an attempt to cover denuded areas, may invade bone and surround sequestrae. The cholesteatoma-like accumulation is then present.

The association of keratosis obturans and bronchiectasis has been noted by other observers, but no previous attempt has been made to relate the association on a physiological basis. Though the hypothesis submitted may not account for all cases, the clinical relationship in this series is too significant to be ignored.

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