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Author's reply

Dear Sirs
We would like to thank Dr Oluwasanmi for her letter regarding our paper. Their report of patients and references support our conclusion that tonsillectomy should be performed with minimal trauma to the tonsillar bed and patients should be informed of the risk of postoperative taste disturbance after tonsillectomy. We have usually been following up our tonsillectomized patients at least one month post-operatively, and our case was the first patient with a complaint of taste disturbance following tonsillectomy among 845 tonsillectomy cases operated on in our clinic between 1987 and 2003. We used an elevator for dissection, a wire loop for amputation, gauze tampons and silk ligatures for bleeding control. We do not use electrocautery or bipolar diathermy. As Dr Oluwasanmi mentioned, the use of bipolar diathermy or electrocautery[1] could be a possible cause for this complication, and it pays to do further studies. We conclude that especially the patients with an additional pathology extending into the lower pole of the tonsil may be at high risk for this complication.

Recently, we have contacted our patient by phone and learned that he was still suffering from taste disturbance 22 months after the surgery. He has preferred not to eat sweet foods and fruits. He said that this disturbance did not have an impact on his social life and he did not need to take any medication such as antidepressants. However, we agree with Dr Oluwasanmi and also wrote in our paper that preoperatively, patients should be informed of the risk of postoperative taste disturbance.

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Immunohistochemical and histopathological features of keratosis obturans and cholesteatoma of the external auditory canal. Atypical keratosis obturans *JLO* 2003;**117**:725–7

Dear Sirs
We read with interest the article by R. Persaud, P. Chatrath, A. Cheesman.

Among the external auditory canal diseases, keratosis obturans (KO) and external auditory canal cholesteatoma (EACC) are less known. Their descriptions in journal and books are full of differences. KO is a rare condition characterized by the presence of a keratin plug in the bony position of the external auditory canal without an erosion of the underlaying bone. EACC is defined as a result of a secondary growth of the stratified squamous epithelium, with inflammation and hyperemia with extensive erosion of the underlying bone. Neiborg *et al.* and Piepergerdes *et al.* demonstrated the differences in clinical and pathological presentations between these two diseases.[1,2] The cause of the KO has been related to seborrhoeic dermatitis, furunculosis, trauma, eczema, sympathetic stimulation of the cerumen glands (in bronchiectasis). Aetiology of spontaneous EACC has been poorly known so far. The characteristic finding in EACC is erosion of the bony external canal in the inferior or anterior part, sometimes with sequestration of the bone.[3] In the pathogenesis of the middle-ear cholesteatoma the main role is played by epithelial cell migration, hyperproliferation and differentiation.[4] Histopathological examinations proved, that spontaneous EACC and middle-ear cholesteatoma have no differences. Haematoxylin and eosin staining of an EACC show a thickening of the squamous epithelium and stroma with inflammatory infiltration. Adamczyk *et al.* showed significant statistical difference between the MIB1 immunoreactivity scores in EACC and normal skin.[3] The immunostaining for EGFR and TGFα showed a cytoplasmic staining pattern and was consistently stronger than in normal auditory meatal skin.[3]

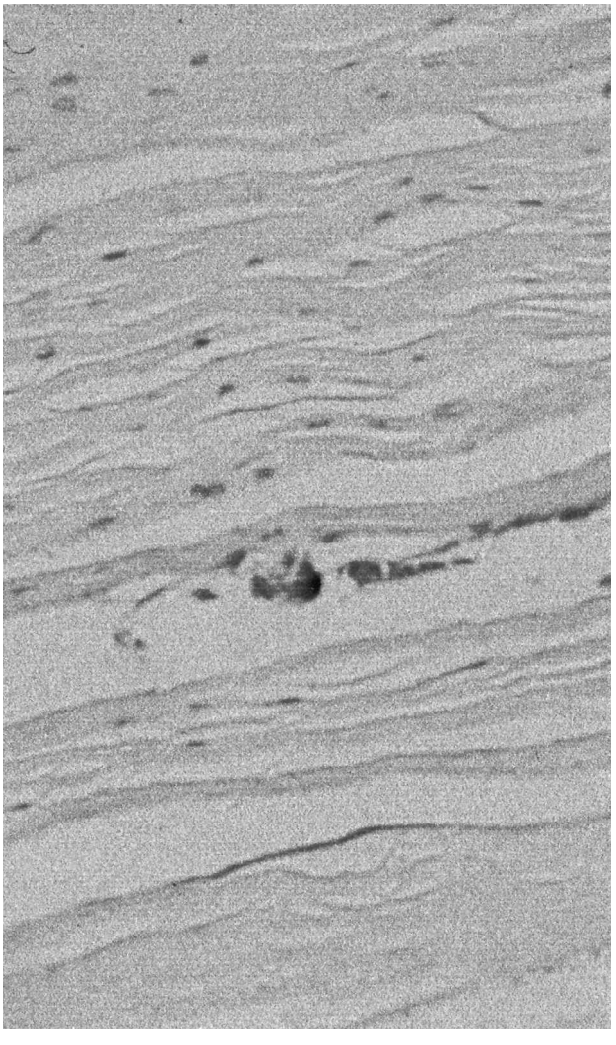

FIG. 1
Keratosis obturans (H&E; ×400).

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New immunohistochemical investigations on KO showed no presence of the growth factors EGFR, TGFβ1 and Ki67 and p53 antigens, responsible for bony resorption. Cytokeratins (CK 5, 6, 8, 17, 19) and tenascin were present. Histopathological investigations proved the presence of single keratinocytes lying among keratin deposits. The differences in histopathological and immunohistochemical examinations show, that EACC and KO are distinct clinical entities.

The case report of 42-year-old woman with KO given is very interesting, although in some points gives diagnostic hesitations. First of all the patient did not complain about hearing loss, pain, pruritus and tinnitus, which are characteristic for KO but not for EACC. Taste impairment resulted from pressure on the chorda tympani and might be present in KO as well as in EACC. In microotoscopy the authors observed 'a large defect in the hypotympanum and the cavity was lined with normal-looking squamous epithelium'. Such a condition is not typical for KO, but it is seen in EACC. The anatomical description of bony lesions is unclear; do authors mean hypotympanum or inferior part of bony portion EC? The presence of bone destruction is characteristic for EACC, but not for KO. That is why we conclude that authors in the described case should diagnose EACC, not KO. This pattern of resorption of the EC bone (not soft tissue) may be due to the pressure and proteolytic activity of EACC. Absence of EC skin inflammation and unilateral occurrence without facial palsy are evidence against KO. Contraindicative for KO is the osteolytic activity, proving the presence of growth factors and proliferation antigens. Regarding all the premises found in the case report we suggest that the patient suffered spontaneous EACC with untypical course. Histopathological and immunohistochemical investigations, not performed in this case, might be the decisive evidence, EACC is a similar disease to middle-ear cholesteatoma, and different from KO (see Figure 1).

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Author's reply

Dear Sirs

We woud like to thank Kuczkowski *et al.* for highlighting some important points pertaining to the dilemma in diagnosing keratosis obturans (KO) versus external ear canal cholesteatoma (EECC). The introduction of immunohistochemical investigations to differentiate between the two conditions suggest that additional tools are still needed to help with the diagnostic problem. It is important to be able to make a definitive diagnosis because each condition requires different managaement strategies. KO can nearly always be managed conservatively with regular aural toilet, but EECC often requires surgery. We feel that the best way to deal with the issues raised by Kuczkowski *et al.* is to evaluate the published evidence.

KO and EECC have been considered as separate entities for the last 20 years, after being regarded as variations of the same disease for at least 87 years. While both disorders are distinct, they do have some overlapping characteristics which may make it difficult to reach a definite diagnosis. In 1980, Piepergerdes *et al.*[1] reviewed the literature and highlighted the differences between the two disorders by comparing the clinico-pathological findings. Patients with KO presented with acute conductive hearing loss, severe pain, a widened ear canal, a thickened tympanic membrane and rarely, otorrhoea. The deafness and pain are usually secondary to the accumulation of keratin in the ear canal. In contrast, EECC is characterized by otorrhoea and a chronic unilateral dull pain secondary to invasion of squamous tissue into a localized area of periosteitis in the canal wall. Hearing and the tympanic membrane are usually normal in EECC. They reported that most cases of EECC occurred unilaterally and in the elderly population. KO is associated with sinusitis or bronchiectasis in 77 per cent of children and 20 per cent of adults.[2] Bilateral occurrences of KO are also more common in children.[3]

Bunting[4] stated that bony erosion of the canal wall can occur in both KO and EECC. In an attempt to differentiate the disorders, Piepergerdes *et al.*[1] described two different erosion patterns. KO is associated with a greatly widened ear canal from circumferential erosion of bone whilst in EECC the erosion is localized to the posteroinferior aspect of the canal. In 1986, Hawke and Shanker reported a case of automastoidectomy caused by KO and suggested that the bony erosion may be due to the pressure exerted by the silently accumulating mass of keratin within the ear canal.[5] However, in EECC the erosion may be the result of proteolytic enzymes and/or inflammation associated with the cholesteatoma in a similar setup to that of middle-ear cholesteatoma with invasion and bone destruction playing prominent parts in the pathology.

In EECC there is bony necrosis or sequestration of the underlying bone[6] and this is distinctly absent in KO.[7] Naiberg *et al.*[7] described the pathological findings in KO as marked inflammation and vascular dilatation in the subepithelial tissue medially in the external ear canal. However, Hawke and Shanker[5] suggested that, in one form of KO, there may be no inflammation in the skin lining the canal.

As both conditions arise in the external ear canal there is an overlap in signs and symptoms. An examination of the cases of KO and EECC presented in the literature revealed that there is no consistent mode of presentation, clinical sign or symptom which reliably differentiates the two conditions. For instance, Piepergerdes *et al.*[1] stated that EECC tends to occur unilaterally and in the older age group, but Smith and Falk[8] described EECC in a child and two young adults. Furthermore, Persaud *et al.*[9] have reported a 34-year-old man in whom both external ear canals were affected with cholesteatomas. Jarvis and Bath[10] described a case of a 79-year-old woman with a chronically discharging painful ear which was associated with a keratin mass. They initially considered the diagnosis of EECC, but a CT scan showed a generally widened ear canal. At surgery a large keratin plug was removed but there was no evidence of ostenonecrosis; the final diagnosis

was KO. This case illustrates that pain and otorrhoea are not consistent differentiating symptoms. Other reports have also confirmed these observations.[6,7] Even in the Piepergerdes *et al.* series,[1] otorrhoea was present in one of the three cases of KO and also in 20 per cent of cases described by Black and Clayton.[3] Hearing loss is also not a reliable distinguishing feature as Heibrun *et al.*[11] reported four patients with conductive hearing loss associated with EECC. Conductive hearing loss was also reported in a case of a giant EECC.[12] Recently, Heilbrun *et al.*[11] described two cases of EECC with circumferential erosion and stated that this pattern of bone destruction is not conclusive in differentiating KO and EECC.

A careful assessment of the reported cases of EECC and KO revealed that osteonecrosis or bony sequestration, found in EECC and not in KO, is the main finding which consistently differentiates the two disorders. In addition, there is a focal loss of the epithelial covering in EECC but not in KO. However, it is conceivable that a severely infected case of KO may result in ostenonecrosis or sequestration of underlying bone associated with epithelial ulceration, but this has never been reported.

In summary, KO and EECC are different conditions originating in the bony part of the external auditory canal. However, it is sometimes difficult to distinguish between the two disorders because of overlapping features. In the future, immunohistochemical techniques may be helpful but currently osteonecrosis, with the focal appearance of sequestrated bone, and the loss of an epithelial covering appear to be the most useful and consistent findings which favour the diagnosis of EECC. These findings were absent in our patient.

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