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Clinical Records

Mucoepidermoid carcinoma of the middle ear – a case report

K. B. K. Soh, F.R.C.S. (Ed.), F.R.C.S. (Glas.)*, H. K. K. Tan, F.R.C.S. (Ed.)*,
R. Sinniah, Ph.D.(Belf.), M.D.(Dub.), F.R.C.P.(I), F.R.C.R.A. (Aust.), F.R.C.Path. (Lond.)†

Abstract
We report a case of mucoepidermoid carcinoma of the middle ear, a site which does not appear to have been previously described. A discussion of four possible theories of pathogenesis are presented.

Key words: Middle ear neoplasms; Carcinoma, mucoepidermoid

Case report

A 51-year-old Chinese male was first seen in April 1991 for right-sided otalgia which was associated with blood stained discharge and hearing loss. The symptoms had been present for six months, but the patient delayed seeking treatment until the pain became quite intense. He had no giddiness or other symptoms referable to the ear, nose and throat regions.

On examination, a friable lesion was seen over the postero-superior aspect of the deep portion of the right external auditory meatus and adjacent tympanic membrane. There was impaired hearing in the right ear, and tuning fork tests suggested a conductive deafness on the affected side. An audiogram revealed a mixed right-sided hearing loss of 100 dB. There were no cranial nerve deficits. The rest of the upper aerodigestive tract and head and neck were normal. Systemic examination was unremarkable.

Our initial investigations were carried out to exclude nasopharyngeal carcinoma in view of its high incidence in the Chinese population of Singapore. Postnasal space biopsy was performed twice although there was no lesion visible. The results were negative on both occasions. IgA antibody levels against the Epstein-Barr virus (viral capsid antigen and early antigen) showed normal titres.

Examination of the ear under anaesthesia revealed a large vascular and friable mass over the postero-superior aspect of the deep part of the external auditory meatus. The tumour had eroded the tympanic membrane. Multiple large biopsies were obtained for histological examination. These showed an infiltrating tumour composed of a mixture of squamous and intermediate type cells intimately admixed in some areas with mucin-secreting cells. The tumour showed both solid and cystic areas with mucin in the lumen (Figure 1). The intermediate and epidermoid cells were arranged in sheets and islands; intercellular

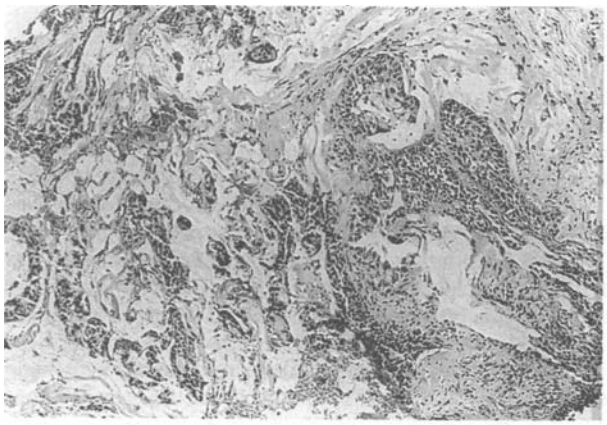

Fig. 1
Section of tumour shows an infiltrating carcinoma with a mixture of squamous and intermediate cell types, and foci of mucin-secreting cells. There are also cystic areas with mucin in the lumen (H & E; × 100).

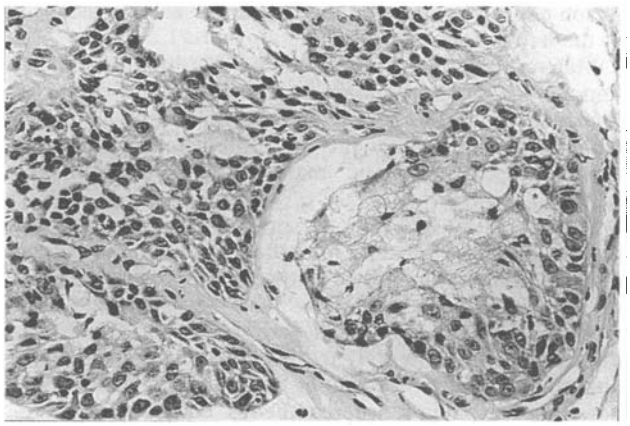

Fig. 2
Higher magnification of the tumour showing intermediate and epidermoid cells in sheets intimately admixed with mucous cells (H & E; × 350).

From the Departments of Otolaryngology* and Pathology†, National University of Singapore, Singapore.
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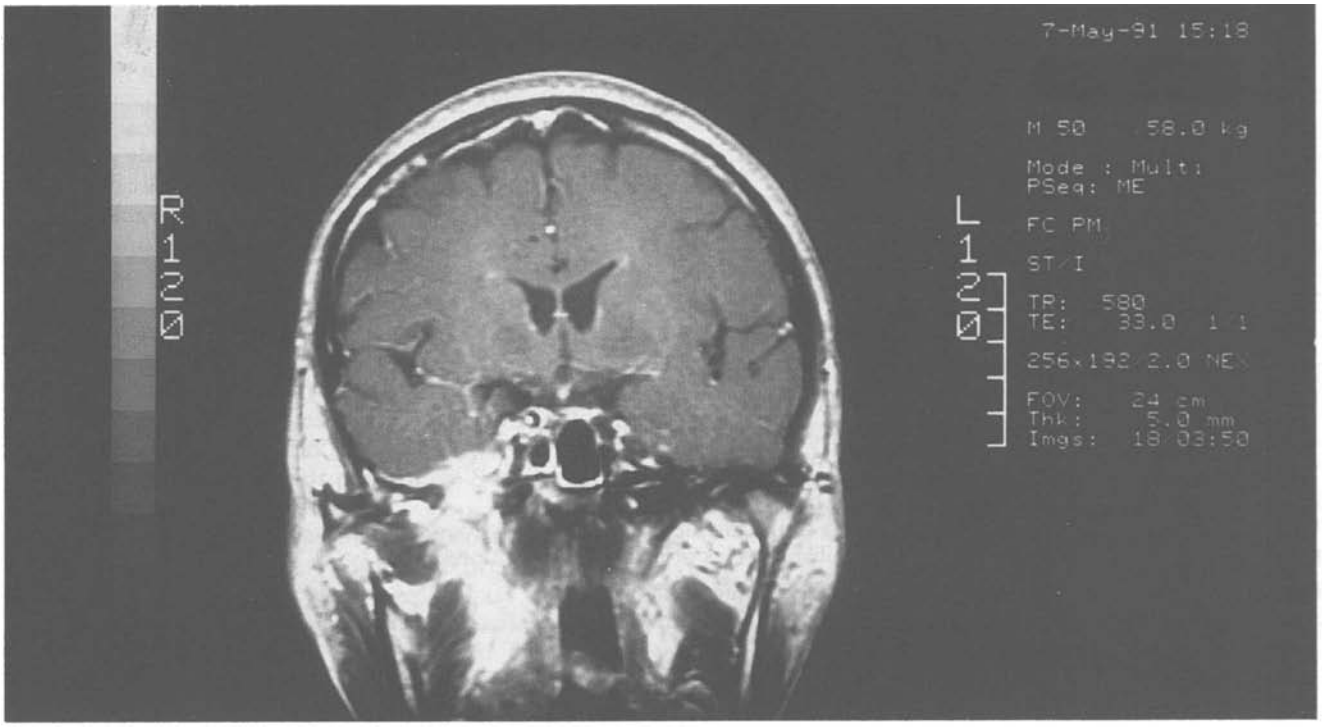

FIG. 3

MRI of the head showing a large tumour extending from the oropharynx to the right temporal lobe and along the lateral pharyngeal wall to the level of the second cervical vertebra.

bridges were prominent between the epidermoid cells. These cells also showed hyperchromatic nuclei with nuclear pleomorphism and were negative for mucin with Southgate's mucicarmine stain. The mucous cells were large and swollen, some having a signet-ring appearance. In some areas they formed multiple layers. They were mucin positive by the mucicarmine method (Figure 2). The histological features were those of an infiltrating muco-epidermoid carcinoma.

Plain radiographs of the chest were normal. Axial CT scanning of the temporal bone and postnasal space and magnetic resonance imaging (MRI) of the head (Figure 3) revealed a large tumour extending from the oropharynx to the right temporal lobe and along the lateral pharyngeal wall to the level of the second cervical vertebra. Enlarged lymph nodes were seen in the right upper neck at the level of the hyoid bone.

Management was discussed by a multidisciplinary tumour board. The tumour was considered too extensive for resection. Radiotherapy and chemotherapy were also withheld as little benefit was predicted from such treatments. Symptomatic management was therefore advised.

Subsequently, the tumour spread laterally along the external auditory meatus. In September 1991, the patient developed right vagal nerve involvement and sudden onset of right vocal cord paralysis. This was followed four months later by trismus and severe speech and swallowing difficulties. Examination at that juncture revealed an absent gag reflex indicating glossopharyngeal nerve paralysis. A palliative percutaneous endoscopic gastrostomy was performed for enteral feeding. The patient eventually died in March 1992.

Discussion

Mucoepidermoid carcinomas are malignant tumours normally encountered in the major (11 per cent) and minor (eight per cent) salivary glands (Foote and Frazell, 1960; Chaudhry *et al.*, 1961). They were first described by Stewart and Becker (1945). The minor salivary glands occur in a variety of sites and are usually scattered in the oral mucosa, palate, cheeks and lips. Lucas (1972) recognized eight groups – labial, buccal, retromolar, floor of mouth, glossopalatine, palatine, tonsillar and lingual. Minor salivary glands of the mucous type also occur in other heterotopic sites (Pasavento and Ferlito, 1976) e.g. hypophysis, ear, mandible (Bhaskar, 1963; Brown and Lucchesi, 1966), sternoclavicular joint and neck.

In addition mucoepidermoid carcinomas may arise from other glandular structures e.g. anal glands (Kay, 1954; Morson and Volkstadt, 1963), oesophageal mucous glands (Kay, 1968; Stanley and Albuquerque, 1970), tracheobronchial mucous glands (Alfred and Ivan, 1972), and biliary ducts (Luis and Ricardo, 1971). Our review of the literature, dating back to 1945 when mucoepidermoid carcinoma was first described, revealed no report of this neoplasm arising from the middle ear. This would, therefore, appear to be the first recorded case of such a lesion.

The lining of the middle ear has been described by Tos (1984) as a 'strongly modified respiratory type epithelium'. There is a gradual reduction in the mucosal thickness as the distance from the tubal orifice increases. The anterior third of the tympanic cavity is lined by a low pseudostratified epithelium, whereas the posterior aspect, together with the antrum and mastoid air cells, is covered by a single layer of flat epithelium. The majority of the cells are non-ciliated and almost no mucous glands are present (Lim, 1979). Tubuloalveolar seromucous glands, occurring in the lamina propria, are normally found in the cartilaginous (pharyngeal) part of the Eustachian tube. In air-filled middle ear clefts, little secretory activity is found. Batsakis (1989) states quite categorically that the normal tympanic cavity, antrum and mastoid air cells normally do not contain seromucous glands. The existence of any glands in

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children and adults are presumed to be due to pathological changes in the middle ear.

Glandular or mixed tumours, namely adenocarcinomas and salivary gland neoplasms found in the middle ear cleft may theoretically arise in any of the following ways:

(1) From seromucous glands that develop as a result of metastatic and functional alterations of the mucosal epithelium. The latter may be a consequence of middle ear disease which induces hyperplasia of the glandular elements in the middle ear.

(2) From chronic middle ear irritation causing the mucosa to undergo squamous metaplasia. Metaplastic changes have been postulated as giving rise to occasional cases of cholesteatoma of the middle ear behind an intact tympanic membrane. They have also been implicated as a source of squamous cell carcinoma of the middle ear. It is therefore reasonable to postulate that metastatic squamous elements could contribute to the 'epidermoid' component of mucoepidermoid carcinomas.

(3) From seromucous and minor salivary glands implanted in the middle ear during embryogenesis. Salivary gland rests are developmental anomalies arising in the proximal part of the second branchial arch and are called choristomas. They are commonly associated with other congenital abnormalities of the second arch. They were first described by Taylor and Martin (1961). The possibility of a salivary gland neoplasm rising from choristomatous middle ear tissue is however extremely remote. Moreover, we have been unable to find any record of a malignant lesion developing in a choristoma, all the cases reported being benign mixed tumours.

(4) From secondary invasion of the middle ear by tumour from adjacent primary sites, e.g. ceruminomas from the external auditory meatus, parapharyngeal space malignancies and cancers of the salivary glands. As the lateral cartilaginous part of the external meatus was free of tumour and the lesion presented initially in the depths of the external auditory meatus and middle ear, it is improbable that the lesion was an extension of a mucoepidermoid carcinoma of ceruminous or salivary gland origin.

It is interesting to note that although there have been approximately 13 cases of primary adenocarcinomas of the middle ear (Schuller *et al.*, 1983), there have been no reports of mucoepidermoid carcinoma. Kinney and Wood (1987) in their review of 30 malignancies of the external ear canal found only one case of mucoepidermoid carcinoma. Five of their patients had primary tumours arising in the parotid gland and extending to involve the external ear canal. Primary parotid tumours usually invade the external ear canal initially and later spread to the middle ear. It is therefore improbable that in our case, the lesion developed in the parotid gland. Furthermore, our patient did not have any obvious external neck or intra-oral swelling, while diagnostic imaging with CT scans and MRI showed the parotid gland to be normal and separated from the lesion by a plane of normal tissue.

In view of the prevalence of nasopharyngeal carcinoma in Chinese populations, we considered this as a diagnostic possibility. As already stated, the investigations proved negative.

Finally, we considered the possibility of a minor salivary gland neoplasm arising in the parapharyngeal space and spreading to the middle ear. However, the patient had no external neck swelling, nor intra-oral abnormality as one would expect in these circumstances. Careful examination of the CT and MR films revealed that the main tumour bulk lay in the middle ear. The bony erosion involved the lateral aspect of the petrous pyramid more than the petrous apex – the converse of what one would expect with a parapharyngeal tumour. Furthermore, spread of the tumour superiorly to the middle cranial fossa also favoured a middle ear origin as parapharyngeal tumours tend to extend along the lines of least resistance, usually inferiorly where there are no bony structures to impede progress.

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Address for correspondence:
Mr K. B. K. Soh, F.R.C.S.,
Department of Otolaryngology,
Leicester Royal Infirmary,
Leicester LE1 5WW.