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Main Articles

'Ceruminoma' – a defunct diagnosis

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Abstract
The clinical and pathological features of glandular tumours of the external auditory meatus are presented. Their heterogenous clinical and pathological features make the collective term 'ceruminoma' ambiguous and misleading. The spectrum of pathological behaviour and histological features demonstrated by these interesting tumours necessitate a broader classification system. In our hospital 32 patients presented with tumours of the external auditory meatus over a 30-year period, of which seven were glandular in origin. A review of the histology of these glandular tumours enabled us to reclassify them as adenoma, cylindroma, adenoid cystic carcinoma or ceruminous adenocarcinoma. Together with the less common mucoepidermoid carcinoma and pleomorphic adenoma this subdivision forms a basis for a more meaningful classification system with prognostic and therapeutic implications specific to each tumour type. The term 'ceruminoma' should no longer be used unqualified.

Key words: Ear canal; Ear neoplasms

Introduction

Tumours of the external auditory meatus (EAM) are rare. The majority of malignant tumours of the EAM are squamous carcinomas (Lewis, 1981), and most of the remaining tumours are of glandular origin. Ceruminous glands appear in the cartilaginous outer third of the ear canal and lie deep in the dermis. The ceruminous glands of the EAM are modified apocrine sweat glands. The secretory portions of the ceruminous glands branch and drain into a duct which opens along with adjacent sebaceous gland ducts into the hair sacs of the fine hairs found in the EAM. The cells lining the secretory portion are cuboidal and contain pigment granules with lipid. The description 'apocrine' implies that a portion of the apical cytoplasm of the cell is shed into the lumen in the secretory process. Histologically this is seen as 'blebbing' of the apical surface of the cells (see Figure 3). The secretory epithelium is surrounded by well developed smooth muscle cells.

On light microscopy they are identical to apocrine sweat glands seen at other sites (axilla, groin, perianal, areolar) in the body (Bloom and Fawcett, 1986).

Over the last 30 years, 32 tumours of the EAM have presented to our ENT Department. Of these, 25 were squamous carcinomas and seven were of glandular origin. Such tumours are often referred to as 'ceruminoma', the assumption being that they arise from the ceruminous glands of the EAM. We believe that the term 'ceruminoma' is of little practical value to the surgeon, and is a misleading term, a view shared by many who have written on the topic (Johnstone *et al.*, 1957; O'Neill and Parker, 1957; Wetli *et al.*, 1972; Hageman and Becker, 1974; Pulec, 1977; Nissim *et al.*, 1981). From the surgeon's point of view these glandular tumours of the EAM present a number of difficulties. Firstly, because they are so rare few surgeons have experience in treating the condition. Secondly, confusion has arisen because of the variable nomenclature used to describe the glandular tumours of the EAM under the generic title of 'ceruminoma'. Thirdly, there is disagreement about their tissue of origin which need not necessarily be the ceruminous glands. Fourthly, the term 'ceruminoma' has been used to describe various entities, such as ceruminous adenoma, ceruminous adenocarcinoma, adenoid cystic carcinoma, cylindroma, hydradenoma, and many others, all of which have added to the confusion, and finally, and most importantly, these tumours exhibit a wide spectrum of malignant potential, so that treatment needs to be tailored to the individual histological type of tumour rather than to an all embracing so-called 'ceruminoma'.

Case reports

The Cardiff experience of glandular tumours of the EAM serves to illustrate the problems inherent in making a diagnosis of 'ceruminoma'. Since 1960 there have been 32 cases of tumours primarily involving the EAM.

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TABLE I

Case no.	Age (years)	Sex	Biopsy	Operative specimen	Treatment	Outcome
1	36	F	Adenoma	Meatal cyst	Local excision	No evidence of recurrence
2	70	M	Low grade adenocarcinoma	Adenoid cystic carcinoma	Local excision	Recurrence after one year
3	62	F	No biopsy performed	Adenoid cystic	Local excision and radiotherapy	No evidence of recurrence after seven years
4	52	F	Adenoma	Low grade adenocarcinoma	Local excision	Recurrence after four years
5	62	M	No biopsy performed	Cylindroma	Local excision	No evidence of recurrence
6	34		No biopsy performed	Adenoma Adenocarcinoma	Local excision Thoracotomy	No evidence of recurrence after six years
7	50	F	Adenoma Adenocarcinoma	Adenoma Adenocarcinoma	Local excision Local excision	Recurrence after one year

Twenty-five of these were squamous cell carcinoma and seven were of glandular origin – the 'ceruminoma'. At the time of the original diagnosis, these tumours could be seen to display differing histological features. These features conveniently provide for a classification which although perhaps implicit to that date, was first cogently expressed by Wetli *et al.*, (1972) who reviewed cases from the literature and proposed a classification describing four types of glandular tumour of the EAM: (i) adenoma, (ii) adenocarcinoma, (iii) adenoid cystic carcinoma, and (iv) pleomorphic adenoma. The seven cases of glandular origin can be classified as shown in Table I into ceruminous meatal cyst (one case), cylindroma (one case), adenoid cystic carcinoma (two cases), and adenocarcinoma (three cases). We have not seen mucoepidermoid tumours as described by Pulec (1977).

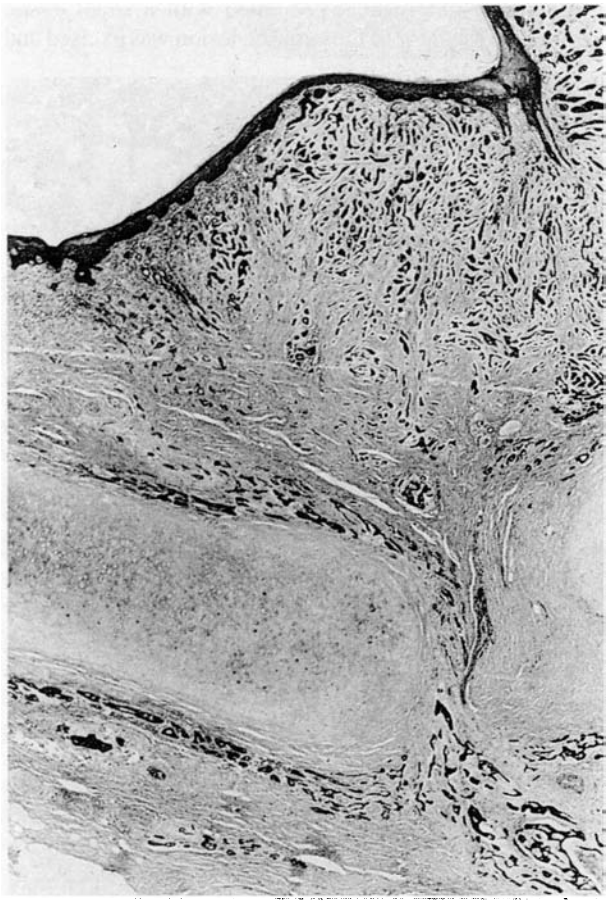

FIG. 1

Adenoid cystic carcinoma (*Case 2*) showing infiltration around islands of cartilage. (H&E; ×20).

The following case histories serve to illustrate both the characteristics and problems of the classification of tumours within our series. We have no experience of the pleomorphic adenoma forming the fourth category in the Wetli *et al.* (1972) classification.

Case 1

A 36-year-old woman presented with hearing loss and a swelling in the EAM which on examination appeared to be cystic in nature. Aspiration of the cyst revealed a thick milky content. An excision biopsy showed ceruminous glands some of which were dilated forming cystic structures lined by cuboidal cells showing apocrine features. Although this lesion was reported as a 'ceruminous ade-

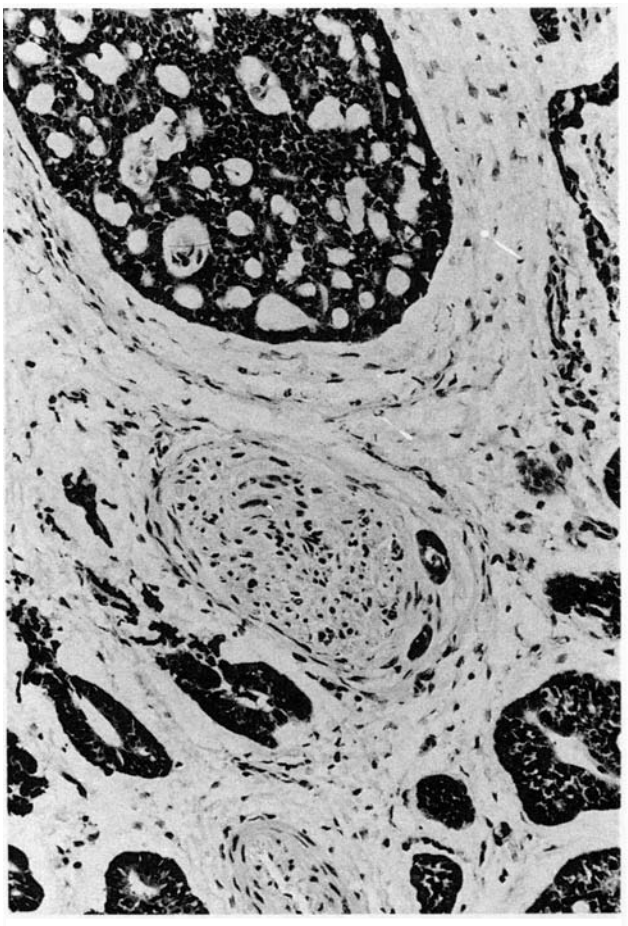

FIG. 2

Adenoid cystic carcinoma (*Case 2*) showing islands of tumour cells with characteristic cribriform pattern and perineural invasion. (H&E; ×200).

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noma' the histological appearance was of a microcystic dilation of glands due to duct obstruction rather than a true neoplasm.

There has been no recurrence on follow-up over 10 years.

Case 2

A 70-year-old man presented with pain, swelling, otorrhoea and deafness. On examination there was an obvious locally invasive tumour. A biopsy of the small (5 × 5 × 2 mm) tumour reported a low grade adenocarcinoma possibly arising from the ceruminous glands. The tumour was treated by extensive local resection of pinna, petrous temporal bone and adjacent dura mater.

Histology of the resection specimen showed skin with a focally ulcerated epidermis. The dermis contained tumour widely infiltrative around cartilage and involving fragments of bone (Figure 1). The tumour was composed of cords and islands of uniform, rounded, darkly-staining, cells showing pseudoglandular formation but no peripheral palisading and no evidence of true gland formation. Some islands showed a characteristic cribriform architecture and perineural invasion was seen (Figure 2). The tumour extended to the cut margins of the excision. Contrary to the original biopsy an adenoid cystic carcinoma was reported. No salivary gland tissue was present.

On follow-up there was local recurrence with intracranial extension and death within one year of the operative procedure.

Case 3

A 62-year-old woman presented complaining of blockage in the ear. On examination there was a soft swelling in the anterior wall of the EAM. The tumour was excised. The histology showed skin with an underlying tumour composed of islands of small, dark, cells with a cribriform pattern similar to those seen in *Case 2*. No perineural invasion was seen and no salivary gland tissue was present. The appearance was of an adenoid cystic carcinoma. The tumour was incompletely excised by local resection and the patient was given radiotherapy.

There has been no recurrence over a seven-year follow-up period.

Case 4

A 52-year-old woman presented with deafness of six months duration. On examination there was a cystic swelling arising from the roof of the external meatus. A wide local excision was performed with skin grafting. The histology showed irregular glandular structures lined by cuboidal epithelium showing apocrine features with lumenal 'blebbing' or 'snouting' (Figure 3). The original diagnosis was of a simple ceruminous adenoma but, on reviewing the histology areas, where tubules infiltrate into fat were seen (Figure 4) and although there was no perineural or blood vessel invasion, the tumour was regarded as a low grade ceruminous adenocarcinoma.

There has been no recurrence over a 20-year follow-up.

Case 5

A 62-year-old woman presented with a small cystic swelling on the floor of the ear. The lesion was excised and

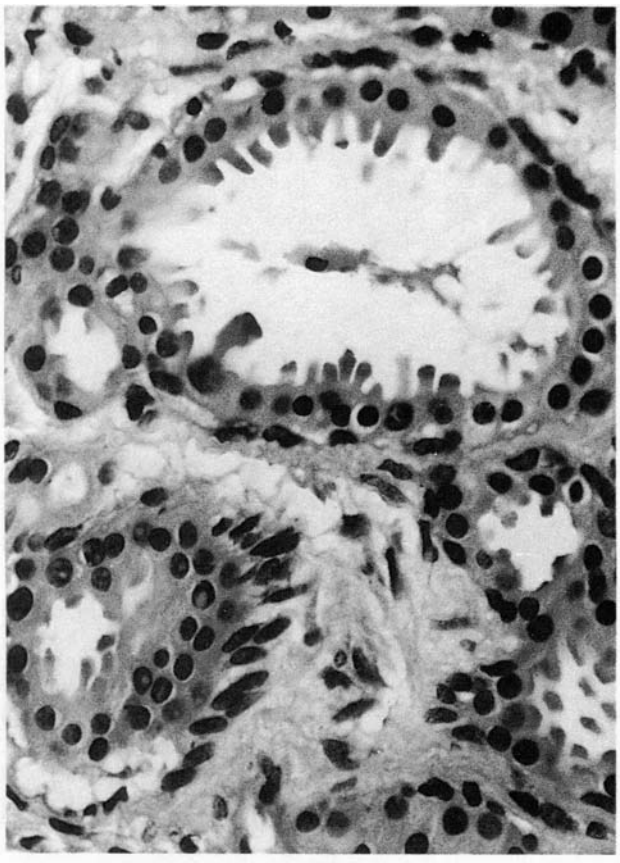

FIG. 3

Ceruminous adenocarcinoma (*Case 4*). Glands in which the epithelium shows prominent 'blebbing' or 'snouting'. (H&E; ×400).

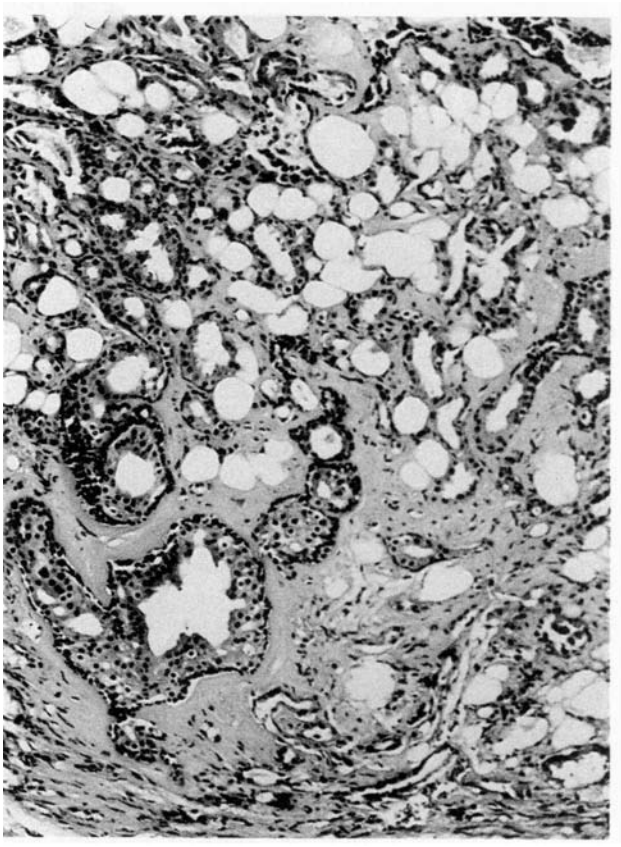

FIG. 4

Ceruminous adenocarcinoma (*Case 4*). Irregular glandular structures infiltrating adipose tissue and connective tissue. (H&E; ×100).

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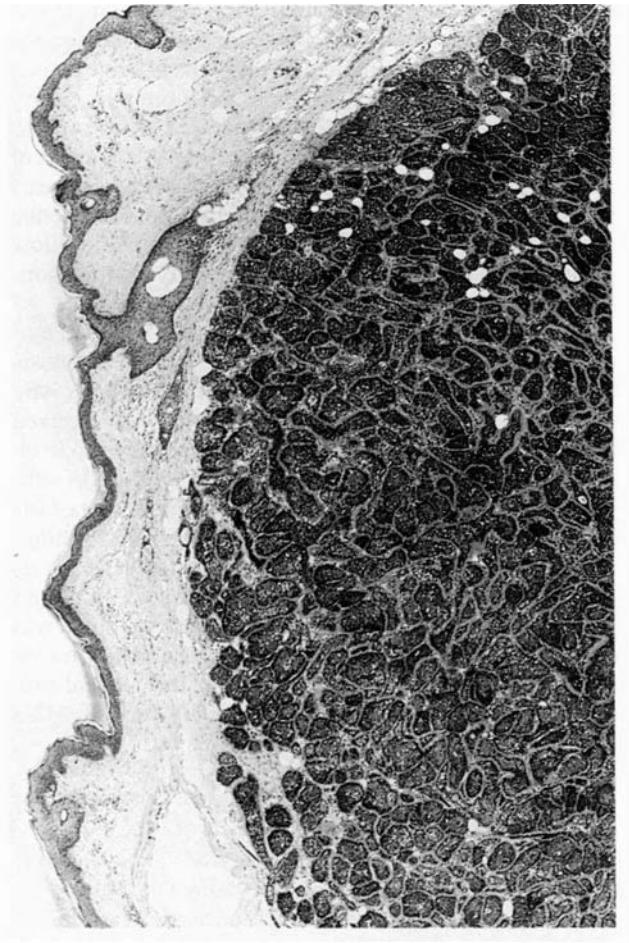

FIG. 5
Cylindroma (*Case 5*) with defined mass composed of islands of darkly-stained cells. (H&E; ×20).

histology showed intact skin containing hair shafts and sebaceous glands with a well-defined tumour in the dermis (Figure 5) composed of discrete islands of small, darkly-staining cells (Figure 6). Each island was surrounded by hyaline acellular material. Within the groups of cells there were rounded hyaline structures. The appearance was of a cylindroma which was incompletely excised. No salivary gland tissue was seen. The lesion recurred eight years later and was again excised locally.

There has been no recurrence over a 20-year follow-up.

Case 6

A 34-year-old man presented with a one-month history of pain in the ear. A 'cyst' was excised. The histology showed intact skin with a well-defined tumour in the dermis extending down to, but not invading, underlying cartilage (Figure 7). There were a variety of histological patterns (Figure 8A–D). There were islands of tumour composed of two cell types some forming glandular structures. In some areas the tumour appeared as solid sheets of basaloid cells. There was little cellular or nuclear pleomorphism and few mitoses were seen. The appearance was interpreted as a benign adnexal tumour (adenoma).

The patient developed haemoptysis six months later. A rounded mass was seen on chest X-ray and a left lower lobectomy was performed. The histology of the pulmonary lesion showed tubules, cylinders and ductules lined by two types of cell with areas similar to the tumour removed from the ear. The cytology remained bland and few mitoses were seen. The tumour was clearly a metastasis from the ear lesion and review of further sections of the original tumour showed evidence of perineural invasion (Figure 9). In the light of its behaviour and subsequent histology a diagnosis of low grade ceruminous adenocarcinoma was made.

There has been no recurrence on follow-up after six years.

Case 7

A 50-year-old woman presented with a polyp in the ear. Initial biopsies showed prominent ceruminous glands and a diagnosis of ceruminous adenoma was made. A year later she returned with pain and a recurrent mass which was excised. The histology showed skin with sebaceous glands and cartilage with an infiltrating adenoid cystic carcinoma. No perineural invasion was seen and the tumour was incompletely excised.

There was recurrence of the tumour within a year and five further resections were carried out over the next three years. The histology showed recurrent adenoid cystic carcinoma. The last resection included parotidectomy with excision of the lower half of pinna and the facial nerve was sacrificed. Histology showed the cavity wall from previous resections contained tumour. Complete excision could not be confirmed due to the fragmented nature of the specimen.

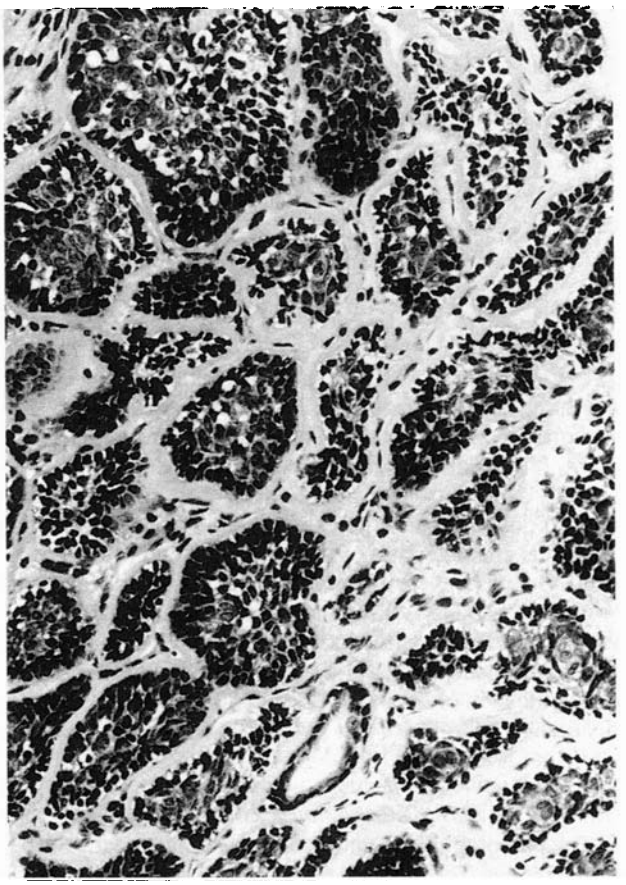

FIG. 6
Cylindroma (*Case 5*). Islands of darkly-staining cells surrounded by hyaline sheath in a jigsaw pattern. (H&E; ×100).

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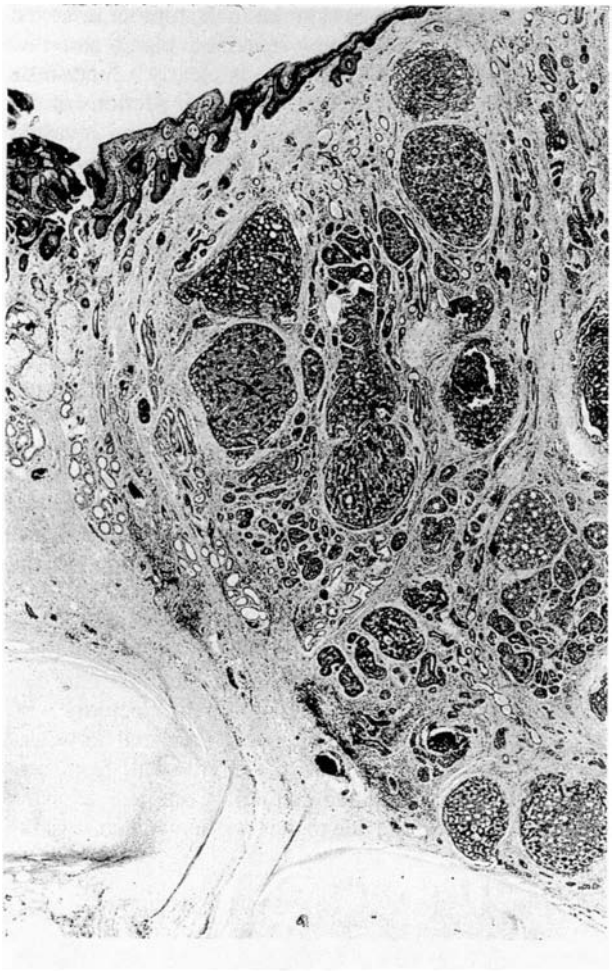


FIG. 7
Ceruminous adenocarcinoma (*Case 6*) showing a well-defined tumour infiltrating down to cartilage. (H&E; ×20).

The patient died six years after the first operative procedure from other causes.

Discussion

Presentation

The first symptoms of a simple adenoma is usually, if anything, hearing loss alone, caused by obstruction of the external auditory meatus. This was the presentation in *Case 1* (meatal cyst) and *Case 5* (cylindroma). Occasionally symptoms may arise from an otitis externa secondary to obstruction of the external auditory meatus.

Pain is characteristic of the malignant end of the spectrum. Our cases were unusual in that it was not a feature in two of the patients with adenoid cystic carcinoma. Nevertheless, pain is a useful characteristic which should make the surgeon suspicious of a malignant lesion if there are difficulties in histopathological diagnosis or classification.

Histological diagnosis

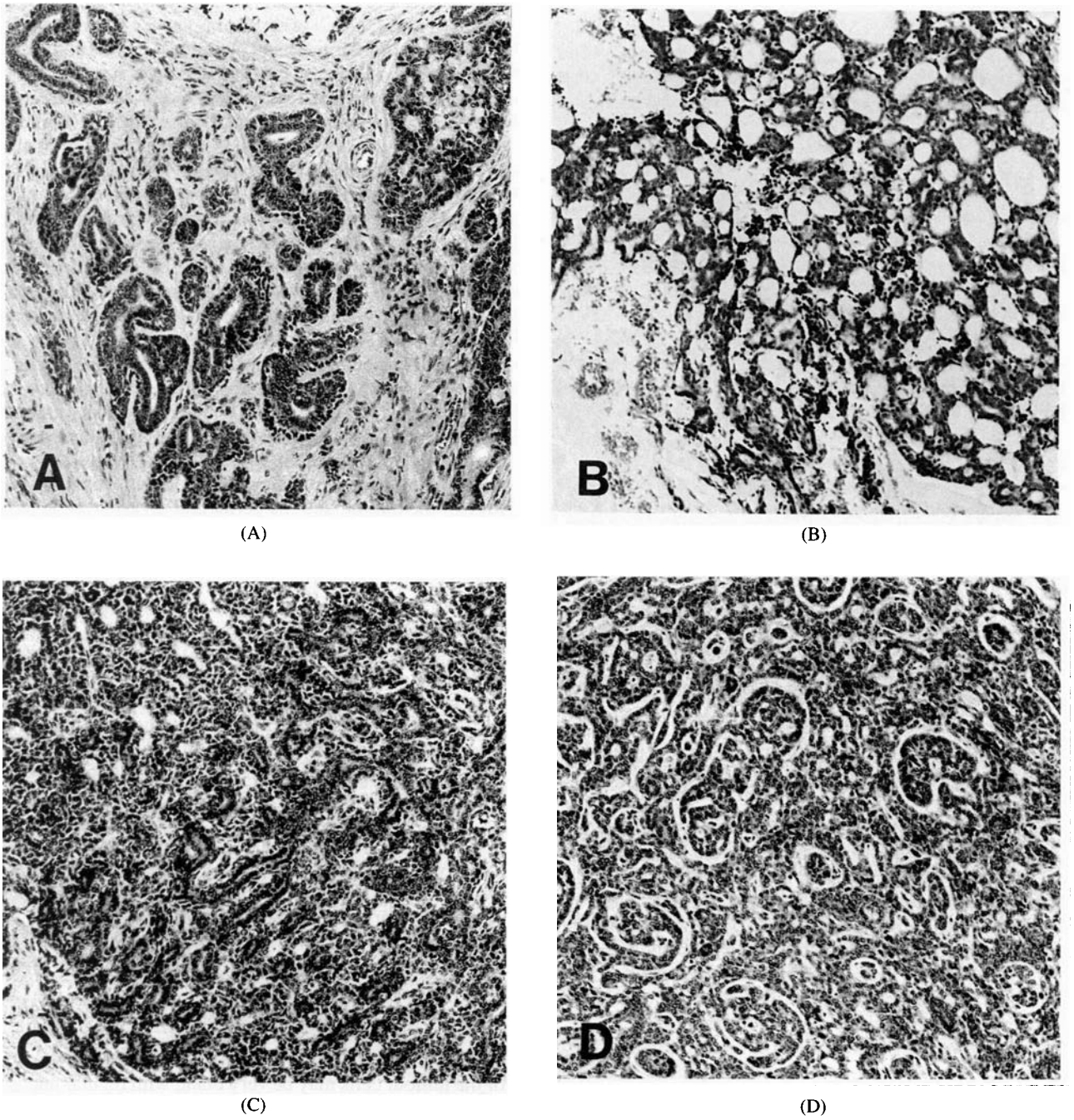
The pathological diagnosis rests on the adequacy of the initial biopsy. The material removed for biopsy may not be representative of the lesion as a whole as some tumours may show a variety of patterns in different parts of the same tumour. This has been mentioned in previous reviews and variation in structure was seen in one of our patients (Figure 8). Of particular concern is the diagnosis of low grade adenocarcinoma which may show a great variety of morphological patterns and more particularly may show very bland cytological features. Some authors have made the point that the only sure way of diagnosis of adenocarcinoma is to demonstrate invasion of bone, cartilage, vascular channels or perineural spaces by the tumour. The biopsy must be of sufficient size to allow detection of this feature which is usually best demonstrated at the periphery of the lesion.

Three patients illustrate this difficulty in histopathological interpretation. In one patient an initial diagnosis of adenocarcinoma was amended to that of adenoid cystic carcinoma once the operative specimen was received (*Case 2*). In another the diagnosis of adenoma made on excision biopsy had to be amended in the light of subsequent behaviour when a pulmonary metastasis appeared. Further sectioning showed perineural infiltration in the periphery of the specimen (Figure 9) and a diagnosis of adenocarcinoma was made (*Case 6*). In a third patient (*Case 7*) an excision biopsy of a meatal polyp was diagnosed as an adenoma and was thus designated to conservative management. One year later this patient presented with pain in the external auditory meatus and a biopsy showed an adenoid cystic carcinoma.

Treatment and prognosis

The adenoma and other benign lesions are adequately treated by local excision alone. The patient with a ceruminous meatal cyst in this series was successfully treated by wide excision of her lesion and skin graft to the defect. The patient with a cylindroma had incomplete excision of her lesion which consequently recurred eight years later. There has been no recurrence during 20 years following a second local excision with skin graft to the defect. In general terms, the malignant tumours require radical surgery with or without radiotherapy. In terms of prognosis, the adenoma are cured by suitable local excision. In our series the majority of the malignant tumours recurred locally or metastasized distally, but the clinical behaviour could not be predicted from the histological features. There is then a wide range of pathogenicity of tumours embraced by the term 'ceruminoma'.

We are obliged to turn to a review of the literature in order to get a better idea of the nature of these tumours. Most papers on 'ceruminoma' appearing in the literature over the past 30 years have been individual case reports (Smith and Duarte, 1962; Grossman *et al.*, 1964; Arora, 1964; Coyas and Adamopoulos, 1966; Ramadass, 1966; Neldner, 1968; Turner *et al.*, 1971; Koopot *et al.*, 1973; Hageman and Becker, 1974; Michel *et al.*, 1978; Habib, 1981; Nissim *et al.*, 1981), or a small series of up to seven cases (Cankar and Crowley, 1964; Batsakis *et al.*, 1967; Anagnostou *et al.*, 1974; Ramadass and Satuanarayana, 1973; Lynde *et al.*, 1984). Three major reviews of 'ceruminoma' have appeared in the literature in the past 20 years (Wetli *et al.*, 1972; Pahor and O'Hara, 1975; Dehner and Chen, 1980). The review and proposed classification by Wetli *et al.* (1972) has already been referred to by adding their own cases to those in the earlier literature they suggested a simplified classification for those tumours putatively of ceruminous gland origin. They



(A) (B)

(C) (D)

FIG. 8

Ceruminous adenocarcinoma (*Case 6*) showing variety of growth patterns within one tumour: (A) serpiginous widely spaced tubules; (B) adenoid cystic-like; (C) microtubules with minimal intervening cellular stroma; (D) 'glomeruloid' structures. (H&E; ×100).

hoped thereby to provide a more accurate basis for diagnosis, rational treatment and follow-up for these patients. Further cases were added or reassigned to different groups by Pahor and O'Hara (1975) and by Dehner and Chen (1980). Their figures serve to illustrate the rarity of these tumours. Most cases included are the subject of case reports or small series similar to ours. Notable exceptions are the clinicopathological study of 16 adenoid cystic carcinomas by Perzin *et al.* (1982) and the major contribution made by Pulec *et al.* (1963) devoted to adenoid cystic carcinoma together with his subsequent publication Pulec (1977) including his experience of other glandular tumours arising in the meatus. This latter publication included six adenoma, five adenocarcinoma, 24 adenoid cystic carcinoma and unusually two mucoepidermoid tumours. This series represents both personal and other experiences over a 70-year period at the Mayo Clinic and Otologic Medical groups from 1910. As such it represents a wealth of experience and practice, and is most useful in providing a principle of management. Drawing from this experience it is possible to characterize and classify the major groups of glandular tumours as follows (see Table II also).

Ceruminous (apocrine) adenoma. This is benign and usually presents with hearing loss. The lesions may be cystic and the presence of pain signifies complications

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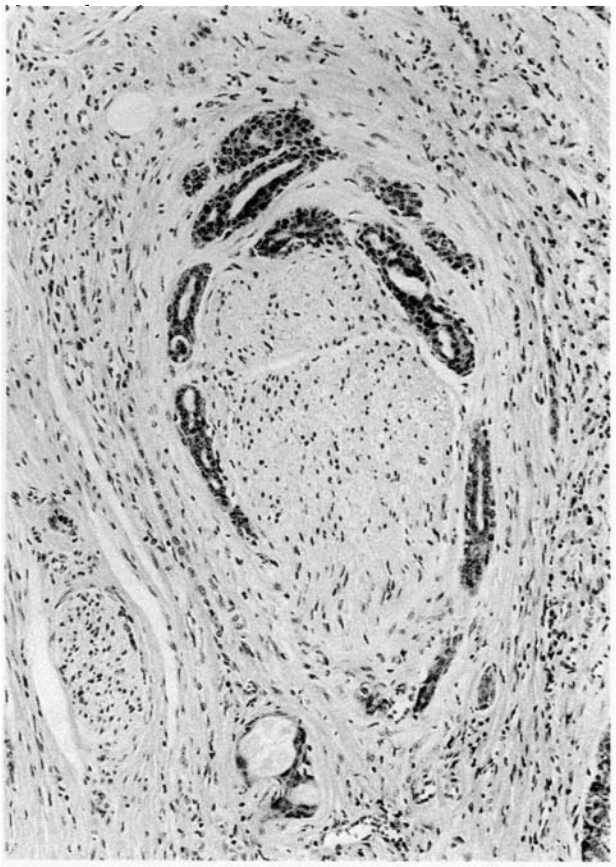

FIG. 9

Ceruminous adenocarcinoma (*Case 6*). Perineural invasion indicating invasive potential of this tumour. (H&E; ×100).

such as otitis externa. The presence of pain at presentation or persistent pain post-operatively should lead to a review of the histology and possibly a repeat biopsy.

Pleomorphic adenoma. Tumours identical histologically to lesions arising in the salivary glands may present as a mass in the ear. Although there is no convincing evidence either way, there is no reason to believe that these tumours arise in ceruminous glands and probably originate in heterotopic salivary gland tissue.

Cylindroma. Cylindroma in non-meatal skin is a slowly growing, single, well-circumscribed tumour occurring most often on the scalp where piliary complexes are common and apocrine glands occur. It may occur at multiple sites on the scalp forming the so called 'Turban tumour'. The origin of the tumour is uncertain but current opinion favours a tumour of the pilar apparatus of a low level maturity. The histological appearance is characteristic with solid irregular masses of basaloid cells showing some peripheral palisading of nuclei and islands of cells closely applied to each other in a jigsaw pattern. Each cell mass is surrounded by a hyaline sheath. The great majority of these tumours behave in a benign manner but may recur locally if incompletely excised. Rare cases of malignancy *de novo* or malignant transformation occur which can be recognized histologically by islands of cells showing cellular pleomorphism, nuclear anaplasia, atypical mitotic figures, loss of hyaline sheath, loss of peripheral palisading and local invasion.

As there are pilosebaceous units in the external ear canal, there is no reason why tumours histologically identical to cylindromas should not arise in the meatus. They should be diagnosed as cylindromas and regarded as benign like the dermatological lesions elsewhere unless they show evidence of malignant transformation. They should not be classified with the ceruminous adenocarcinomas. Confusion has arisen in the past because adenoid cystic carcinoma and ceruminous adenocarcinoma have been called cylindroma (Pulec *et al.*, 1963).

Adenoid cystic carcinoma. Adenoid cystic carcinomas arising in the external auditory meatus present with a history of pain which may have been present for some years, although this was not a feature of one of the cases presented here. The histological features and biological behaviour are identical to adenoid cystic carcinoma arising at other sites and the tumour may infiltrate perineurally. The problem with adenoid cystic carcinomas arising in the EAM is the local anatomy which makes it difficult to remove an adequate margin of uninvolved tissue around the tumour to ensure adequate excision at the first operation. This point has been stressed by other authors who recommend the widest practical resection. The invasive edge of the tumour and the perineural invasion mean that it is impossible to be sure of the extent of the tumour macroscopically. Intraoperative frozen sections of resection margins may be useful in order to confirm the absence of tumour although of course any bone or bony fragments in margin biopsies will cause difficulties.

Death may result from intracranial spread or pulmonary metastases, sometimes after multiple local recurrences. Unresectable tumours or death from the tumour has been associated with histological demonstration of tumour at resection margins, involvement of the parotid gland, extension into bone, perineural invasion and local recurrence of tumour (Perzin *et al.*, 1982).

Ceruminous adenocarcinoma. This term embraces a range of glandular carcinomas which have been divided into low grade adenocarcinoma and high grade adenocarcinoma on histological grounds. Patients present with pain in the ear and a mass in the meatus. There may be problems with the initial diagnosis as in two cases reported here (*Cases 4* and *6*), particularly with low grade tumours.

The low grade carcinomas show a great variety of architectural arrangement which has caused problems with diagnosis and classification in the past. The cytology of the tumour cells may be very bland with regular nuclei, no pleomorphism and few mitotic figures. The only criterion for the diagnosis of malignancy in these cases is that of infiltration into bone, cartilage, blood vessels or perineurally. Clearly an adequate specimen is required to demonstrate such infiltration at the margin of the tumour

TABLE II

CLASSIFICATION OF GLANDULAR EXTERNAL AUDITORY MEATUS (EAM) TUMOURS

Benign	Putative origin
Ceruminous (apocrine) adenoma	Ceruminous glands
Pleomorphic adenoma	Heterotopic salivary gland
Cylindroma	Pilosebaceous units
Malignant	
Adenoid cystic carcinoma	Heterotopic salivary gland
Ceruminous adenocarcinoma	Ceruminous glands
low grade	
high grade	
Mucoepidermoid carcinoma	Heterotopic salivary gland

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mass. It is not possible to predict the biological behaviour of a low grade adenocarcinoma on the basis of its histology and some of these low grade lesions may metastasize haematogenously to kidney or to lung as in one of our patients (*Case 6*) and as previously reported (Turner *et al.*, 1971). Further confusion has occurred in the past because these tumours were called 'cylindromas'.

High grade ceruminous adenocarcinoma is easily recognizable because of glandular irregularity and cellular pleomorphism. It is difficult to exclude a metastasis from some other site on histological grounds. When an enlarging mass presents in the ear with the histology of infiltrating adenocarcinoma, a metastatic deposit from another site should be excluded by further investigation.

Mixed patterns. Confusion has arisen in the past because of ceruminous adenocarcinomas which have areas within them resembling adenoid cystic carcinoma or other tumour patterns. The variety of histological appearances within ceruminous adenocarcinomas has already been stressed and mixed tumours should be reported as such.

Management

From these characteristics a suggested management plan is as follows:

Ceruminous (apocrine) adenoma. Conservative local excision with a skin graft to the operative site. No radiotherapy is necessary.

Adenoid cystic carcinoma. Adequate treatment requires wide local excision of tumour. Radical surgery initially offers the best chance of cure. The extent of the tumour may not be obvious on clinical examination, especially because of the tendency for perineural invasion by this tumour. Pre-operative assessment includes conventional CT scanning and possibly magnetic resonance imaging. Even with these additional investigations, it may be difficult to assess the extent of spread pre-operatively. Surgery should include excision of the external auditory meatus and possibly the pinna, with an extended radical mastoidectomy for tumour involving the posterior meatal wall and mastoid. Involved dura must be excised, and anterior spread necessitates a total parotidectomy and excision of the mandibular condyle. Because of the propensity to perineural infiltration the facial nerve needs to be sacrificed if there is any doubt about clearance and intraoperative frozen section to ensure clear margins should be considered. Irradiation is rarely curative in this type of tumour, but can play a part in palliation, especially of pain. Completeness of excision can only be assessed by the pathologist and for this reason orientation of the operative specimen to allow identification of true resection margins is particularly important. Piecemeal removal of tissue makes assessment of excision margins impossible.

Adenocarcinoma. The mainstay of treatment of this tumour is surgery, as in adenoid cystic carcinoma. Adenocarcinoma is much more responsive to radiotherapy which can be used in planned combined treatment and if necessary for palliation.

Poor general condition may preclude major surgery and multiple distant metastasis are a contraindication to major surgery, although conservative surgical intervention may have a place in a few patients. Solitary metastasis to kidney or lung may be a feature of these tumours, and as such they are amenable to resection as was the case with one of our patients (*Case 6*). Intracranial spread makes a cure unlikely, and paralysis of any of the lower cranial nerves is a poor prognostic indicator. In essence, the biggest operation that can be rationally developed gives the patient the best chance of a cure.

Conclusions

The purpose of this paper was to show that the term 'ceruminoma' is defunct and pathologically meaningless. This case presentation and literature review highlight the following key areas of difficulty.

(1) The rarity of these tumours means few otologists have a worthwhile experience of treating these tumours. Adopting a fairly rigorous review, and admitting for consideration only those tumours with adequate histology and documentation, we are discussing at the most 100 reported tumours.

(2) The term 'ceruminoma' has hitherto served only to cause confusion. It is too vague and has been used to describe a range of pathological entities which can now be more precisely defined. It is unsatisfactory because the construction of the term with the suffix 'oma' implies a benign lesion. (Melanoma is an equally misleading term but because it is sanctioned by universal and long usage and everyone is familiar with its implications, it is unlikely to be changed).

(3) It seems unlikely that all the lesions described under the heading 'ceruminoma' actually arise from the ceruminous glands. The origin of cylindroma of the skin is uncertain but the consensus of opinion is that it arises in the pilosebaceous unit. Adenoid cystic carcinoma is the commonest tumour arising in the EAM under the title 'ceruminoma'. In the EAM it is morphologically indistinguishable from tumours arising in the salivary glands and other tissues e.g. breast and skin. Adenoid cystic carcinoma is the commonest malignant tumour arising in minor salivary glands and ectopic salivary tissue but has been recorded in the ear and mastoid region. In addition, when neoplasms arise in apocrine glands of the skin, malignant behaviour is very rare. Primary adenoid cystic carcinoma in the skin is very rare and the likelihood is that adenoid cystic carcinomas in the EAM in fact arise in heterotopic salivary gland tissue.

In our view the term 'ceruminoma' should be abandoned in favour of a classification using the more specific and defined entities outlined here and by other authors.

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