Primary tumours of the vestibule and inner ear

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Abstract
Seven primary tumours of the vestibule and inner ear are described, six schwannomas and one traumatic fibroma. Schwannomas in this situation may occur as sporadic tumours, or may be a feature of neurofibromatosis type 2 (NF-2). In the latter condition they may occur in isolation or in association with, but separate from, schwannomas arising in the internal meatus. Direct extension into the vestibule of an intrameatal vestibular schwannoma is well reported, but extension of an intravestibular tumour into the internal meatus is not described. Traumatic fibromas of the vestibule are rare and the trigger could be an attack of labyrinthitis. Intravestibular tumours, although rare, are likely to be diagnosed with increasing frequency with the widespread use of MR imaging.
Key words: Vestibule; Labyrinth; Schwannoma, vestibular; NF-2; Fibroma

Introduction

Primary tumours of the vestibule are rare. Until recently they were found by chance at post-mortem or during the course of surgery, either osseous labyrinthectomy or during a translabyrinthine approach to the cerebellopontine angle. Clinical diagnosis is very rare and the majority of those subsequently confirmed at surgery had been labelled as 'atypical Menière's disease'. It is likely, however, that with the widespread use of MR imaging more will be detected, and operated upon. Most neoplasms of the vestibule are vestibular schwannomas. They may occur sporadically or more frequently as part of neurofibromatosis type 2 (NF-2) in which they may co-exist with separate intracranial tumours; indeed, the earliest cases in the literature were in NF-2, erroneously referred to as Von Recklinghausen's disease (Gray, 1933; Scott, 1938; Nager, 1969). Most intravestibular schwannomata remain confined to the inner ear but extension to the middle ear has been recorded (Storrs, 1974; Mafee *et al.*, 1990). Direct extension of an intrameatal tumour into the vestibule is a possible but unusual occurrence, the lateral wall of the meatus usually proving remarkably resistant to erosion. In this report seven primary intravestibular tumours are described; six schwannomata and one post-traumatic fibroma. Of the six schwannomata, four occurred in patients with NF-2 and two sporadically.

Case records

Case 1

This 44-year-old male presented in 1977 with a history of positional dizziness when lying on the left side, but no hearing loss. Examination revealed typical benign paroxysmal positional vertigo and nystagmus when carrying out the Dix Hallpike manoeuvre with the left ear under. Pure tone audiometry and petrous tomography were normal. Caloric testing revealed a left-sided canal paresis. No further investigations were carried out at this stage. He suffered constant unsteadiness for two further years before it finally resolved. Some 10 years after this initial presentation he returned complaining of progressive left-sided hearing impairment, left-sided tinnitus and disabling vertigo. A pure tone audiogram indicated that his hearing had deteriorated to a 60 dB level with poor speech discrimination. Computed tomography (CT) was normal but magnetic resonance (MR) was not available at that time. Although a definite diagnosis had not been reached it was suggested that he should have a bony labyrinthectomy. At surgery, a soft pink mass was found to occupy the vestibule and to extend into the semicircular canal system (Figure 1). The medial wall of the vestibule was intact and there was no suggestion of tumour extension into, or from, the internal meatus. The cochlea was not obviously involved. The nerve of origin could not be confidently stated, but it was thought to be one of the

FIG. 1

Case 1. Surgical specimen removed from the labyrinthine vestibule during transmastoid labyrinthectomy.

vestibular nerves. Histological studies confirmed that this tumour was a schwannoma with spindle-shaped cells arranged in whorled pattern with nuclear pallisading (Antoni type A). The patient has been free of vertigo for nearly 10 years since his surgery, and a recent MR scan (10 years after surgery) was clear of tumour. This case was previously reported by Birzgalis *et al.* (1991).

Case 2

This male was 22 years of age when he presented with a five-year history of left-sided deafness and intermittent dizziness. There were no other stigmata of NF-2 but two of his second degree relatives had had intracranial tumours (a pituitary adenoma and an unspecified tumour). Pure tone audiometry revealed a subtotal hearing loss on the affected side. The right side was normal. MRI revealed a lesion in the left vestibule which enhanced with intravenous gadolinium (GTPA) and a presumptive
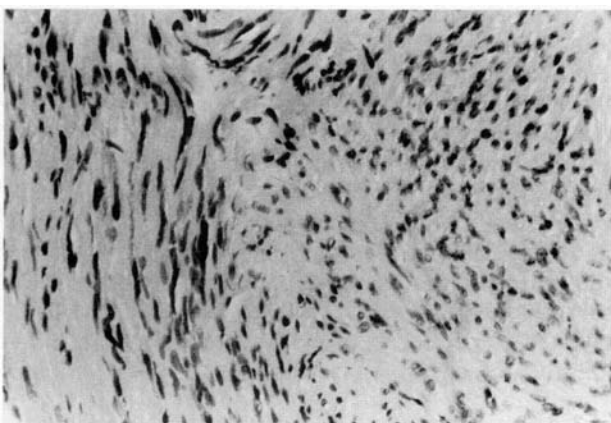
diagnosis of intravestibular schwannoma was made. An osseous labyrinthectomy was carried out and the tumour was found to occupy the anterior vestibule and to extend to the ampulla of the lateral semicircular canal. It was presumed to arise on the superior vestibular nerve. Histological examination confirmed that the tumour was a schwannoma with Antoni A and B patterns. A one-year post-operative MR scan showed no sign of tumour either at the site of surgery or elsewhere in the skull, but because of the high suspicion of NF-2 in a young person with a schwannoma and a family history of intracranial tumours he remains under regular surveillance.

Case 3

This 20-year-old male presented with a severe left-sided sensorineural deafness of two years duration and with signs of rising intracranial pressure. Audiometry revealed a total left-sided sensorineural deafness and a mild low frequency loss on the right. MR scanning demonstrated the presence of large bilateral vestibular schwannomata. A translabyrinthine removal of the left-sided tumour was advised. Tumour was found to occupy the vestibule, the superior semicircular canal and the anterior crus of the lateral canal. This tumour was removed in its entirety and the medial wall of the vestibule was seen to be intact. The internal meatus and posterior fossa were then opened and total removal of a separate 4 cm vestibular schwannoma was then carried out. The internal meatus was filled with this tumour, but there was no continuity with the tumour in the vestibule. Both tumours were reported as vestibular schwannomata: that from the vestibule exhibited mainly an Antoni type A appearance (Figure 2)
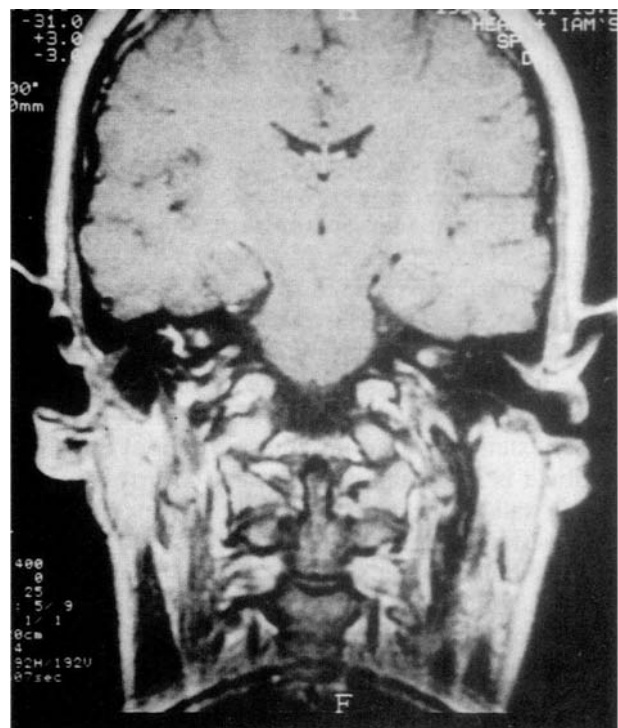

FIG. 2

Case 3. A schwannoma composed of Antoni A-celled areas, with nuclear palisading.

FIG. 3

Case 4. MR scan showing bilateral small vestibular schwannomas with a separate intravestibular tumour on the right side.
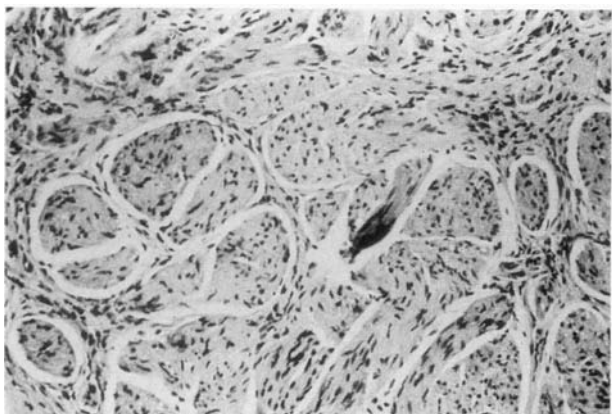
FIG. 4

Case 5. Traumatic neuroma demonstrating fibrovascular tissue with enlargement of some nerve twigs but no schwannoma.

whereas the intracranial tumour showed both Antoni A and B features. This patient has gone on to have his opposite vestibular schwannoma removed and an auditory brainstem implant inserted.

Case 4

This female was first seen when she was 35 years of age and gave an eight-year history of progressive right-sided deafness. The left side seemed to be normal. She had a striking family history of NF-2, with bilateral vestibular schwannomas in three other members in three generations. Audiometrically, there was no measurable hearing on the right. On the left side hearing was normal. MRI revealed the presence of bilateral intrameatal tumours with a separate intravestibular schwannoma on the right (Figure 3). As yet, no surgery has been performed.

Case 5

This 65-year-old female had presented with a sudden onset of profound left-sided deafness, otorrhoea and dizziness 12 months previously. The dizziness and discharge disappeared but the deafness did not. Pure tone audiometry confirmed that the hearing loss on the left side was total. MRI suggested the presence of an intravestibular lesion which enhanced with intravenous gadolinium DTPA, and which appeared to extend into the lateral end of the internal auditory canal. At osseous labyrinthectomy the lateral and posterior canals were found to be obliterated by fibrous tissue and by new bone, and a soft grey tumour was found which extended into the vestibule and appeared to be arising from the superior vestibular nerve. The medial wall of the vestibule was intact and there was no suggestion of extension into the internal meatus. On histological examination the appearance was of fibrovascular tissue with enlargement of some fine nerve twigs but with no evidence of schwannoma. The histological diagnosis was of a traumatic neuroma possibly related to a previous episode of labyrinthitis (Figure 4).

Case 6

This 23-year-old male presented with bilateral vestibular schwannomas and a family history suggestive of NF-2, although the affected family members were not available for assessment. In this case an unenhanced MR scan, performed at an outside hospital, demonstrated bilateral vestibular schwannomata but there was no pre-operative indication of a co-existing intravestibular schwannoma (Figure 5). His 2.5 cm left-sided tumour was approached through a translabyrinthine operation and as the labyrinthectomy was completed a separate pinkish grey tumour was found filling the vestibule completely and extending a short distance into the superior and lateral canals. The medial wall of the vestibule was intact and there was no continuity with the intracranial tumour, the intrameatal portion of which extended to the fundus of the meatus. Although the nerve of origin of the intravestibular tumour could not be confidently identified, the relationship to the superior and lateral canals made the superior vestibular nerve seem the most likely origin. The histopathological report confirmed that the intravestibular tumour was a schwannoma with both Antoni A and B patterns present. The intracranial tumour showed Antoni A features only.

Case 7

This 36-year-old female with a strong family history of NF-2 presented with a history of right-sided hearing loss which was initially fluctuant, and a tendency to stagger to the right side. MR revealed the presence of bilateral vestibular schwannomas filling the internal auditory canals and extending into the CP angles by approximately 0.5 cm. T2-weighted images suggested that the right vestibule did not contain fluid and on gadolinium-enhanced T1-
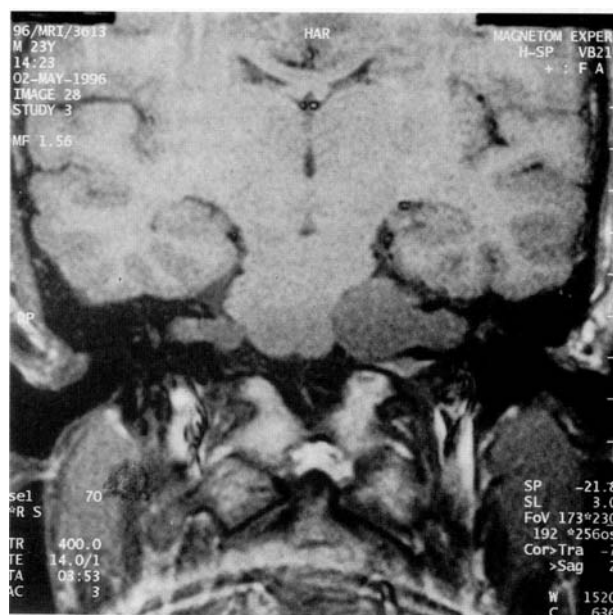

FIG. 5

Case 6. Unenhanced T1-weighted MR image demonstrating bilateral vestibular schwannomata with no evidence of intravestibular tumour.

FIG. 6a

Case 7. T2-weighted MR image. Bilateral vestibular schwannomata extending into the cerebellopontine angles. On the right side there is a low signal from the vestibule suggesting a solid mass compared with the high signal from the left side which is typical of fluid.

FIG. 6b

Case 7. Gd-enhanced T1 image. Note the high signal from the right vestibule.

weighted imaging there was a small area of intense enhancement in the vestibule, suggestive of an intravestibular schwannoma (Figures 6a and b). The hearing threshold was elevated by an average of 30 dB across the frequency range in the right ear. In the left ear the hearing was close to normal. But for the presence of the intravestibular lesion on the right side an attempt might have been made to perform a hearing preservation operation via a middle fossa approach.

Discussion

Seven cases are described of primary intravestibular tumours. Six were schwannomas and one was a traumatic neuroma. Of the six schwannomas, four occurred in patients suffering from NF-2 and two were apparently sporadic. Vestibular schwannomas usually arise within the internal auditory meatus from the Schwann cell covering of one or other branch of the vestibular nerve. It has been stated (Skinner, 1929) that the point at which the vestibular nerve acquires its Schwann cell covering is the site of origin of these tumours (glial neurilemmal junction; Obersteiner Reddlich zone). Schuknecht (1974) on the other hand stated that the origin can be at any point lateral to this junction rather than at the junction itself. Because the glial neurilemmal junction usually lies within the internal auditory meatus, the majority of vestibular schwannomas originate within the internal canal. The usual growth pattern of tumours arising within the internal meatus is of expansion in a medial direction, being the line of least resistance with extension into the cerebellopontine angle and widening of the porus of the internal meatus.

Much less commonly these tumours are found within the vestibule or in the cochlea. They may spread by direct extension of an intrameatal lesion, or may arise in the inner ear *de novo*. Of the latter, the majority are frequently, but not invariably, seen as part of neurofibromatosis type 2 (NF-2). Spoendlin (1966) has pointed out that the sheath of the eighth

nerve passes through the canal of Rosenthal to the habenula perforata and that is the presumed site of origin of primary intravestibular tumours. Although the majority of intracranial vestibular schwannomas arise on one or other branch of the vestibular nerve, it has been suggested that there is a greater tendency for primary intravestibular tumours to originate from the cochlear division (Huang, 1986), although the number of cases in which there is unequivocal evidence of the site of origin is too small to allow one to draw any definite conclusions as to site of origin. The cases described by Gussen (1971), Hoshino and Ishii (1972), Jorgensen (1962), Johnsson and Kingsley (1981) all arose on the cochlear nerve. The cases of Weymuller (1975), Stewart *et al.* (1975) and Vernick *et al.* (1984) arose on the superior vestibular nerve. That of Miyamoto *et al.* (1980) arose on the inferior vestibular nerve. All of the cases reported here were primarily intravestibular in origin with no indication of a cochlear nerve origin.

The first post-mortem description was that of Mayer in 1917 and since then there have been a number of case reports of temporal bone findings in such cases (Wittmack, 1930; Gray, 1933; Scott, 1938; Jorgensen, 1962). The first cases of audiologically evaluated, radiographically investigated and surgically removed tumours were those of Karlan *et al.* (1972) and of Wannamaker (1972). Gray (1933) described two autopsy cases (three temporal bones) of NF-2 with tumour in the inner ears. In one ear there was definite erosion of the tumour from the internal meatus into the inner ear. In the other two ears the impression conveyed is of primary lesions within the scala tympani and at the lower end of the modiolus respectively although the documentation is somewhat sketchy. Scott (1938) described two autopsy cases of NF-2. In one there was direct extension of tumour from the internal meatus into the vestibule with obliteration of the vestibule and extension into the superior and lateral semicircular canals and the basal turns of the cochlea. In the other case there was a primary intravestibular tumour involving the vestibule, the lateral canal and cochlea. Jorgensen (1962) described the post-mortem appear-

ance of a patient not suffering from NF-2 with a 3 × 2 × 1.5 mm schwannoma occupying the scala tympani of the basal turn of the cochlea and filling the canal of Rosenthal with loss of ganglion cells. No abnormalities were to be seen in the Organ of Corti, however, and Scarpa's ganglion and the auditory and vestibular nerves were normal. The patient had suffered from a progressive bilateral hearing loss for 10 years but no vertigo. Hallpike (1963) described a case of conductive deafness in a patient with NF-2 and at post-mortem a 'seedling' tumour was found in the vestibule impeding the movement of the stapes. A similar appearance was described by Johnsson and Kingsley (1981) of a 1.5 mm tumour arising in the scala tympani and seeming to originate in the distal cochlear neurone.

Gussen (1971) recorded an interesting post-mortem study of a 57-year-old man, not suffering from NF-2, who had a 24-year history of deafness in his left ear, with tinnitus and dizzy spells which sound typical of Menière's disease, lasting for three to seven hours, accompanied by vomiting and occurring about once per week. A diagnosis of 'pseudo Menière's syndrome' was made. At post-mortem (he died of tropical sprue) the temporal bone revealed an elongated tumour with typical appearance of a schwannoma arising just peripheral to the spiral ganglion and extending through the modiolus to all turns of the cochlea and into the spiral bony lamina. The interesting feature of the histological examination was that, despite the history of vertigo, all the vestibular sensory epithelia were normal. Adhesions and scarring were present in the vestibule, around the utricle and the endolymphatic duct. Gussen speculates that the dizziness could have resulted from altered fluid dynamics amounting to hydrops as a result of the scarring in the vestibule. Gussen's speculation is reinforced by the case of Benitez et al. (1967) of a 35-year-old female with NF-2 who died after removal of a large schwannoma from the left ear. Post-mortem examination of the right ear which had had a 55–70 dB pure tone threshold, revealed a tumour in the internal meatus, invading the basal turn of the cochlea and vestibule. There was good hair cell survival in the cochlea and in the maculae and cristae, but there was a granular precipitate in the endolymphatic and perilymphatic spaces. One of the two cases described by De Lozier et al. (1979) had a fluctuant low frequency loss and at surgery viscous perilymph was found in the vestibule in association with the intravestibular tumour. Hoshino and Ishii (1972) found a protein deposit in the scala tympani and moderate dilatation of the saccule and the common crus. It may well be that elevation in hearing threshold in the face of good hair cell survival might be due to a chemical alteration in the inner ear fluids. Vernick et al. (1984) make the point that the majority of patients with intravestibular schwannomas have a pre-operative diagnosis of Menière's disease. Their patient presented with fluctuant hearing, tinnitus and fullness in one ear followed by attacks of vertigo with nausea lasting for several hours. The audiogram

showed a predominantly low frequency 70 dB sensorineural hearing loss, with a normal ABR. Caloric testing confirmed the total absence of function on the affected side. At labyrinthectomy a 4 × 3 × 2 mm schwannoma was seen to arise from the ampullated end of the superior semicircular canal and extend into the vestibule. Mafee et al. (1990) in reviewing the 16 cases of proven intra-labyrinthine schwannomata in the English literature point out that all nine who were operated upon had a clinical diagnosis of Menière's disease. Casselman et al. (1994) report that nine (17 per cent) of 52 patients with clinical diagnoses of Menière's disease displayed other pathology on MR scanning which included intracanalicular vestibular schwannoma (three cases), cerebellar and brainstem infarcts (two cases), post-operative fibrous obliteration of the vestibular system (two cases), a venous loop compressing the cochleovestibular nerve (one case) and a congenital malformation of the lateral semi-circular canal (one case).

Intravestibular tumours can erode out of the inner ear to involve the middle ear. Storrs (1974) described two cases of vestibular schwannoma in the middle ear. In one case the tumour had clearly arisen in the inner ear. In the other case there was a 2 cm intracranial component, so it was not clear whether the tumour had originated in the vestibule and grown both medially and laterally or whether it was a primary intracranial tumour which had eroded laterally into the vestibule and the middle ear. Mafee et al. (1990) described two cases of extension of a primary intravestibular tumour into the middle ear, and in one of them were able to demonstrate on MR imaging the continuity of the inner and middle ear components of the tumour through the round window. The possibility of further lateral extension is suggested by the cases of Tran Ba Huy et al. (1987) and of Woolford et al. (1994) in which tumour eroded through the whole temporal bone and middle ear to present as an aural polyp.

Before the advent of MR imaging it was almost impossible to make a pre-operative diagnosis of intravestibular schwannoma. Plain radiology and CT were generally not sensitive enough, although it is possible that high definition CT might demonstrate expansion or erosion of the cochlea or vestibule. The use of MR imaging in the routine evaluation of all cases of unilateral cochlear or vestibular failure has led to an increase in the pre-operative diagnosis of intravestibular tumours (Brogan and Chakeres, 1990; Mafee et al., 1990; Saeed et al., 1994). They appear as lesions of intermediate signal on T1-weighted images and demonstrate intense homogenous enhancement on gadolinium enhanced T1 sequences (Mafee, 1995). Refinements in T2-imaging of the inner ear may be relied on to pick up the absence of a fluid signal when there is tumour present.

It is important to recognize that there are non-neoplastic enhancing lesions of the vestibular system visible on MR scanning. Casselman et al. (1993) have demonstrated that unenhanced and enhanced T1-weighted images in conjunction with CISS images

are extremely useful to show inflammatory changes in cases of labyrinthitis and haemorrhage within the labyrinth. Uncalcified soft tissue obliteration of the labyrinth can also be seen in Cogan's syndrome, otosclerosis and in post-meningitic cochlear implant candidates.

NF-2 is characterized by the occurrence of bilateral vestibular schwannomas. These tumours may be multifocal. They are usually confined within the internal auditory canal or posterior cranial fossa, but they may be found in the inner ear. Thus, an apparently solitary vestibular schwannoma may be associated with an ipsilateral intravestibular lesion, or with intravestibular schwannoma on the opposite side. Accurate imaging of the inner ears is therefore important in the planning of surgery for such individuals. The coexistence of an intravestibular tumour on the same side as an intracanalicular lesion might change the surgeon's view as to the appropriateness of any hearing preservation operation. Furthermore, any management strategy that included the possibility of a multichannel cochlear implant would be altered by the knowledge that tumour was present in the cochlea. Intralabyrinthine masses can be detected on high resolution, moderate to heavily T2-weighted images by carefully looking for absence of the normal bright signal from labyrinthine fluid. A small area of signal reduction or absence, however, can blend into the signal void of the surrounding compact bone and so be easily overlooked. For this reason we believe that there is still a place for intravenous gadolinium (DTPA) enhancement when imaging cases of suspected vestibular schwannoma, despite the advances in MR imaging techniques, particularly in NF-2 patients. The patient reported here as *Case 6* perfectly demonstrates this pitfall for the unwary. It is equally important to image the inner ear as well as the cerebellopontine angle and the internal meatus on the side opposite a solitary vestibular schwannoma especially if there is a family history of NF-2 or if there are other stigmata to suggest such a diagnosis, especially childhood cataracts or if the apparently sporadic first tumour presents under the age of 30.

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