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Case Series

Etiological diversity of otogenic facial nerve palsy in children – a case series and literature review

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ABSTRACT

Facial nerve palsy in children causes a significant impact on the quality of life. Infectious cause such as otitis media is the second most common etiology for facial nerve palsy in children. We describe the presentation, management and outcomes of four such children who presented to our institution with facial nerve palsy secondary to acute or chronic otitis media. Case one was unique due to the underlying immunodeficiency and severe grade of facial nerve palsy. Case three was managed conservatively, hence emphasising that early treatment can completely curb the need for a surgical intervention. Cases two and four, with grade two to three facial nerve palsy showed good clinical improvement after surgery. Timely reference to otolaryngology by pediatric peers can prevent the onset or worsening of complications secondary to otitis media such as facial nerve palsy. Recurrent episodes of otitis media should raise a high degree of suspicion of immunodeficient states, which will require a multi-disciplinary treatment.

Keywords: Facial nerve palsy, Chronic otitis media, Acute otitis media, Facial canal dehiscence, Immunodeficiency

INTRODUCTION

The facial nerve is responsible for facial expression, lacrimation, and salivation.[1] The incidence of facial nerve palsy (FNP) in children over the age of ten is 10.1 per 100,000 annually and in those under the age of ten is 2.7 per 100,000 annually.[2] Though the incidence in children is two to four times lesser than that in adults, it can cause a significant impact on the quality of life in young children. The long-term consequences on vision, eating, drinking, psychological issues and self-esteem are inevitable. Infectious etiologies account for 12–36% of pediatric cases, with otitis media (OM) being the most common infectious cause of FNP in children.[2] An early referral and onset of treatment, along with a high degree of suspicion to rule out associated immunodeficiencies, can provide the best clinical improvement as well as an enhanced quality of life. In this case series, we describe the presentation, management and clinical outcomes of four children who came to our institution with FNP secondary to otitis media.

CASE SERIES

Case 1

In case one, a three-year-old male child presented with recurrent episodes of left ear discharge, one to two episodes per month, treated conservatively with multiple oral antibiotics, elsewhere. In the present episode, child had purulent left ear discharge for fifteen days and fever for three days, following which the child developed difficulty in closing the left eye and deviation of angle of mouth to the right side for two days. The child also had a background history of septic arthritis of the right knee at 18 months of age. Otoscopic examination revealed congested external auditory canal and congested tympanic

membrane (TM). Child also had House-Brackmann (HB) grade five FNP.

High resolution computed tomography (HRCT) temporal bone was suggestive of left otomastoiditis with cholesteatoma formation and partial erosion of the incus, malleus and tympanic part of the left temporal bone. Child underwent left canal wall up mastoidectomy. Intraoperatively, necrotic tissue was found in the middle ear which was removed. No cholesteatoma was found. Intraoperative pus culture that was sent, showed heavy growth of MSSA and *Pseudomonas aeruginosa*. He received intravenous antibiotics for five days and was continued on oral antibiotics for two weeks. The clinical outcome did not improve postoperatively. Meanwhile, primary immunodeficiency workup was done by the pediatric infectious diseases team. Clinical exome sequencing confirmed the diagnosis of Bruton's X-linked agammaglobulinemia and the child is currently on regular treatment with intravenous immunoglobulins every once in three weeks.

Case 2

Case two was a six-year-old female child who presented with recurrent episodes of right ear pain and discharge. In the current episode, child had right ear discharge and pain for three days, deviation of angle of mouth to the left side and inability to close the right eye for one day and coryza for eight days. On examination, right TM was congested and HB grade 3 FNP was noted. HRCT temporal bone was suggestive of right otomastoiditis with soft tissue filling the epitympanum and mastoid, with erosion of tegmen tympani and facial nerve canal as seen in Figure 1.

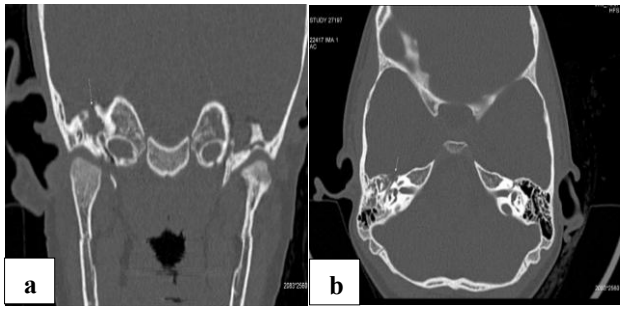

Figure 1: HRCT temporal bone in case two showing (a) erosion of right tegmen tympani (white arrow), and (b) erosion of right facial nerve canal (white arrow).

A diagnosis of right COM, mucosal type, active with mastoiditis and right FNP was made and child underwent right canal wall up mastoidectomy. Intraoperatively, ossicles were intact. Atticotomy was done and head of malleus was removed (Figure 2a). Granulation tissue in the epitympanum (Figure 2b), which was causing erosion of the tympanic segment of facial nerve, was removed. Child was started on facial physiotherapy and oral antibiotics. On the follow-up visit postoperatively, child showed remarkable clinical improvement compared to the preoperative status. She had normal symmetry of the mouth, complete eye closure without effort and normal movement of the forehead i.e. frowning.

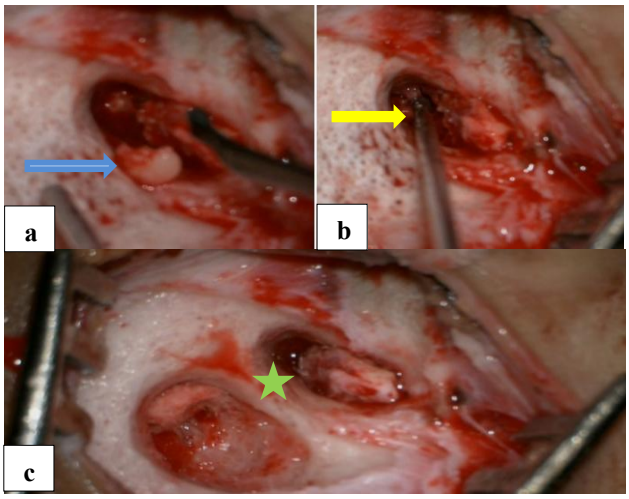

Figure 2: Intraoperative image showing (a) head of malleus (blue arrow); (b) granulation tissue in the epitympanum (yellow arrow); and (c) completed canal wall up mastoidectomy with intact posterior canal wall (green asterisk).

Case 3

Case three was a two-year-old male child who presented with right ear discharge and pain for ten days, fever for five days, deviation of angle of mouth to the left side and inability to close the right eye for three days. He had no previous history of ear discharge. He was treated elsewhere with oral and intravenous antibiotics before presenting here. On examination, child had grade 3 HB FNP. Diagnosis was made as right acute otitis media (AOM) with right FNP. There was minimal ear discharge in the right ear and considering the acute episode, the child was treated conservatively with aural suctioning and antibiotic ear drops. He received systemic antibiotics as per pediatric advice. HRCT temporal bone was suggestive of bilateral otomastoiditis. Post management, he attained complete clinical recovery with normal facial symmetry and no ear discharge.

Case 4

Case four was a two-year-old male child who presented with recurrent episodes of right ear discharge. The last episode was for two months, with intermittent foul-smelling ear discharge. The child also developed deviation of angle of mouth to the left side and inability to close the right eye for five days. Otoscopic examination revealed attic perforation and cholesteatoma flakes. Child also had grade 2 HB FNP. HRCT temporal bone was suggestive of right otomastoiditis and presence of soft tissue in the middle ear and mastoid as seen in Figure 3.

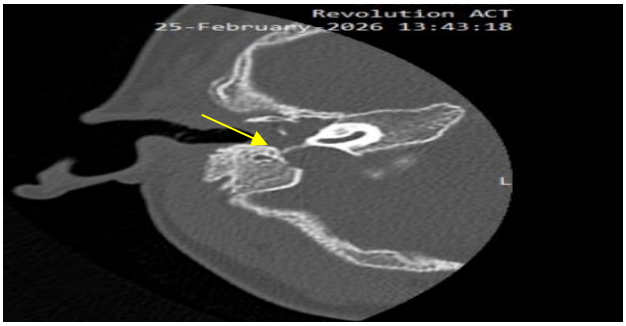

Figure 3: HRCT temporal bone in case four showing the presence of soft tissue in the right middle ear and mastoid (yellow arrow).

Diagnosis for this child was right COM, squamous type, active with right FNP. He underwent right canal wall down mastoidectomy and ossiculoplasty with total ossicular reconstruction prosthesis (TORP) over the oval window. Intraoperatively, cholesteatoma was noted in the middle ear and mastoid as shown in Figure 4. The cholesteatoma sac was attached to the dehiscent bony canal of tympanic segment of facial nerve. Malleus was intact while incus and stapes were absent.

On postoperative follow-up visit, child had complete clinical improvement with normal facial symmetry. Table 1 gives a summary of the diagnosis, intraoperative findings and management of the four children in our case series.

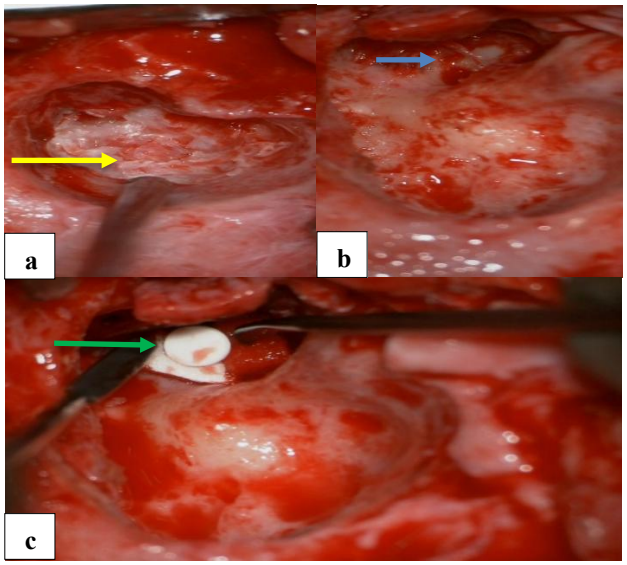

Figure 4: Intraoperative findings in case four showing (a) cholesteatoma in the mastoid cavity (yellow arrow); (b) dehiscent tympanic segment of facial canal (blue arrow); and (c) superior view of the TORP (green arrow) which was placed over the oval window.

Table 1: Summary of diagnosis, intraoperative findings and management of the four cases.

S. no.	Case no.	Age (years)/sex	Diagnosis	Intraoperative findings	Treatment
1	Case 1	3/male	Left COM with left FNP with primary immunodeficiency (Bruton's X-linked agammaglobulinemia)	Necrotic tissue in middle ear	Left canal wall up mastoidectomy
2	Case 2	6/ female	Right COM, mucosal type, active with mastoiditis and right FNP	Granulation tissue in the epitympanum causing erosion of tympanic part of facial canal; ossicles intact; atticotomy done, head of malleus removed	Right canal wall up mastoidectomy
3	Case 3	2/male	Right AOM with right FNP	Conservative management	Aural suctioning, antibiotic ear drops and systemic antibiotics
4	Case 4	2/male	Right COM, squamous type with right FNP	Cholesteatoma seen in the middle ear and mastoid; cholesteatoma sac was attached to the dehiscent bony canal of tympanic segment of facial nerve; malleus intact, Incus and stapes absent	Right canal wall down mastoidectomy + ossiculoplasty with TORP over the oval window

DISCUSSION

The most common aetiology of FNP in children is idiopathic (Bell's palsy), followed by infective causes, such as acute otitis media.[3] Various hypotheses have been postulated for the etiology of FNP in children with otitis media. Firstly, the early stage of AOM can cause retrograde infection within the facial nerve canal. Secondly, inflammatory bacterial toxins may cause peripheral demyelination of the facial nerve.[1] The facial nerve, when it travels in the fallopian canal to exit the

stylomastoid foramen, lies in close proximity to infections of the temporal bone. This causes inflammation and edema of the nerve, thereby compromising the nerve's vasculature because of the rigid fallopian canal, leading to paralysis.[4] Poor host resistance in infants can also play an important role in the development of FNP with otomastoiditis.[5] Among our four cases, only case three had FNP secondary to acute otitis media while the other three cases were secondary to chronic otitis media. Once a chronic infection occurs, granulation tissue or cholesteatoma can also directly compress the facial nerve.[1] This was clearly noted according to the intraoperative findings described in cases two and four.

Among the three cases treated surgically, all had different intraoperative findings namely necrotic tissue, granulation tissue and cholesteatoma, highlighting the varied presentation of facial nerve palsy secondary to otitis media. The surgical management was also tailored according to the intraoperative findings. In case one, though HRCT temporal bone was suggestive of cholesteatoma, intraoperative findings and histopathology reports were suggestive of necrotic tissue. This emphasises the fact that radiological investigations have to be corroborated with clinical and surgical findings to arrive at the accurate diagnosis.

Other than case one, the remaining three children presented with grade two to three HB FNP and had a good recovery. Case two, where the child only had congested TM on otoscopic examination, highlights the fact that the absence of a classic perforation or cholesteatoma flakes does not necessarily rule out the presence of COM. Case three reemphasises the importance of an early referral which enabled us to curb the disease progression with just a conservative management, instead of going up till surgical management. This is in line with the findings of various studies which concluded that a conservative management is the first-line treatment for FNP with OM.[6,7]

The incidence of facial canal dehiscence is seven percent in the pediatric population, with the tympanic segment of facial nerve being the most common site to be involved.[8] Case two and four also had pathology involving the tympanic segment of facial nerve. In a review by Piergabrielle et al it was noted that during surgery, some areas of bone dehiscence and edema of the tympanic portion of the facial nerve were present, therefore supporting the theory of infectious involvement of the facial nerve occurring through the bony dehiscence.[6] Congenital facial canal dehiscence occurs due to failure of ossification centres and it is generally a micro dehiscence, measuring less than 1 mm, detected in histopathological studies.[9]

An interesting point to note in case one is that the child presented to us with a grade 5 HB FNP and after canal wall up mastoidectomy, the clinical outcome did not improve considerably in the follow-up visits. This can be attributed to both the underlying immunodeficiency which had to be tackled as well as the severe grade with which the child presented. This emphasises the need for early referral and surgical management. Case one also highlights the importance of a multi-disciplinary approach. A high degree of suspicion due to recurrent purulent ear discharge in a young child, enabled us to effectively refer the child to the pediatric infectious team to rule out immunodeficiencies.

Similar to our case, Garg et al described a 4-month-old female baby with combined immunodeficiency syndrome complicated by bilateral otomastoiditis, right-sided parotitis, and FNP. The case study underscores the importance of considering combined immunodeficiency syndromes in the differential diagnosis of paediatric patients presenting with recurrent infections and COM.[10] The child in case one also had a past history of septic arthritis, which was in favour of an underlying immunodeficiency.

In a case series by Cynthia et al a 16-year-old male child with OM and FNP was also diagnosed to have lymphomatoid granulomatosis through a lung wedge biopsy. Ten months after his initial presentation, he was noted to have some improvement of his FNP from a HB grade six to a HB grade three with treatment of his underlying disease.[2] We may perhaps be able to observe a similar clinical improvement in our case one, with continued treatment for the underlying Bruton's agammaglobulinemia.

FNP and OM in children can often lead to the diagnosis of other pathologies. This need not be restricted to immunodeficiencies as seen in case one but even present as tumours. In a retrospective review by Tal et al a 7-year-old boy with FNP and OM, not responding to usual management, was later found to have a parapharyngeal rhabdomyosarcoma involving the petrous bone.[4]

In this review, they also found that children with OM and FNP were of a younger age group compared to those with Bell's palsy.[4] This is similar to our case series where all our cases were less than 10 years, with the maximum age being 6 years.

CONCLUSION

Timely reference of children with otitis media to the otolaryngology team can ensure adequate evaluation and early medical and surgical management. This will in turn prevent the onset or worsening of complications secondary to otitis media such as facial nerve palsy.

Persistent or recurrent episodes of otitis media, especially in a very young child and when associated with a purulent discharge, should always raise a high degree of suspicion of immunodeficient states. A thorough evaluation involving a multi-disciplinary team can help clinch the right diagnosis and provide better management outcomes.

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