The Journal of Laryngology & Otology (2014), ***128***, *1114–1116.*
© JLO (1984) Limited, 2014
doi:10.1017/S0022215114002722

CLINICAL RECORD

Samter's triad with aural involvement: a novel approach to management

P SETHUKUMAR, R HEYWOOD, A NARULA

Department of Otolaryngology, St. Mary's Hospital Paddington, Imperial College Healthcare NHS Trust, London, UK

Abstract

Background: Samter's triad is a well described condition manifesting as chronic rhinosinusitis with nasal polyposis, asthma and aspirin intolerance in a non-atopic individual. The underlying mechanism is still to be fully elucidated. However, aural disease has not been widely reported in these patients. In the few reported cases, most patients underwent major surgery with varying degrees of success.

Case report: We report two Samter's triad patients with aural involvement. Both were successfully managed by conservative treatment, thus avoiding the need for major surgery. It appears that more Samter's triad patients may have aural disease than previously thought.

Conclusion: We report good outcomes with conservative treatment, which is relevant because aural disease tends to reoccur in these patients.

Key words: Aspirin; Ear Diseases; Nasal Polyps; Mastoid Bone; Postoperative Complications

Introduction

Samter's triad manifests as chronic rhinosinusitis with nasal polyposis, asthma and aspirin intolerance in a non-atopic patient.[1] It was first described by Widal *et al*. in 1922,[2] but became more widely recognised following further studies by the immunologist M Samter working with RF Beers in 1967.[3] The pathophysiology is not entirely clear: arachidonic metabolites and a heightened host tissue response are present in this patient cohort, and exposure to aspirin and other cyclooxygenase-1 enzyme inhibitors exacerbates upper and lower airway irritation.[4] Elevated inflammatory cysteinyl leukotriene levels and a reduction in the protective effects of prostaglandin E2 are also postulated to be involved.[5] Histological analysis of affected tissue has shown a combination of eosinophilic infiltration and mast cell degranulation.[4]

In Samter's triad, polyposis is generally limited to the nasal cavity and paranasal sinuses but has been reported in the extradural space via dehiscence of the skull base.[6] Two cases of aural involvement and polyposis have been reported; both were managed by tympanomastoid surgery. We report two further cases of aural involvement in Samter's triad, one of aural polyposis with otitis media with effusion (OME), and another of OME without polyposis. Both cases were satisfactorily managed by non-surgical means.

Case reports

Case 1

A 40-year-old woman with a history of Samter's triad presented with significant hearing loss. She had developed asthma and sinusitis four years earlier and had no atopic component, but was found to be intolerant to aspirin. Functional endoscopic sinus surgery several months previously had provided some symptomatic relief. She was diagnosed with right-sided otitis media with effusion, and a tympanostomy tube was inserted.

The tympanostomy tube was extruded one month later, leaving a perforation in the anteroinferior aspect of the right tympanic membrane. She returned to our clinic two months later with decreased hearing and right-sided otorrhoea. Examination showed a right-sided polyp protruding through the tympanic membrane perforation. Pure tone audiometry showed a mild-to-moderate mixed loss.

The patient was managed conservatively, but re-presented two years later with exacerbation of both the nasal blockage and aural symptoms. This time there was also a small perforation in the anteroinferior quadrant of the left tympanic membrane, with a small polyp protruding through it. The polyp was removed in clinic. The right tympanic membrane had a persistent anteroinferior perforation and thick otorrhoea was present. Histological analysis confirmed the presence of abundant mucin-containing degenerating eosinophils (Figure 1).

The patient was treated with ciprofloxacin and betamethasone ear drops and fluticasone nose drops. There was a short-term improvement in her nasal symptoms, which later deteriorated. She then underwent revision nasal polypectomy. Examination under anaesthesia of the right ear confirmed a pinhole perforation. A decision was made not to perform any major tympanomastoid surgery. This patient's main complaints are currently right-sided otorrhoea and a unilateral stable pinhole perforation.

Accepted for publication 2 April 2014

https://doi.org/10.1017/S0022215114002722 Published online by Cambridge University Press

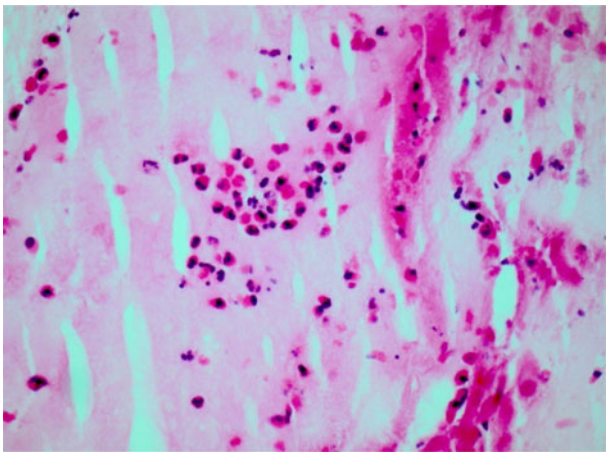


FIG. 1
Histological analysis of mucus extracted from polyp showing abundant degenerating eosinophils. (H&E; ×50)

Her symptoms flared up in May 2013, and she received a two-week reducing course of oral steroids. Review after two weeks confirmed a significant improvement in her nasal and aural symptoms, and she was advised to recommence taking the nasal steroid drops. At the last review in January 2014, her symptoms were still controlled following a recent course of high-dose oral steroids. Her current management includes short courses of high-dose steroids as 'rescue therapy' to use when her aural symptoms flare up. Thus, conservative measures have been considerably successful: both aural and nasal symptoms have responded extremely well to intermittent courses of high-dose oral steroids, thereby avoiding extensive surgery.

Case 2

A 59-year-old female Samter's triad patient had undergone left-sided mastoid surgery and tympanoplasty for cholesteatoma in 1985, followed by a revision procedure in 1998. She had left moderate-to-severe mixed hearing loss and normal hearing on the right.

She also underwent endoscopic sinus surgery and polypectomy for extensive nasal polyposis in 2006, and her nasal symptoms were controlled with nasal douching and topical steroids. Her asthma was controlled medically by using a steroid inhaler and avoiding precipitants such as aspirin. Sinus disease flare-ups coincided with deterioration in her respiratory function.

In December 2011, she developed hearing impairment to the right ear, with otalgia and otorrhoea. Examination revealed a markedly inflamed right tympanic membrane with a small central perforation and purulent otorrhoea. The left middle ear and mastoid cavity were healthy. She had bilateral nasal polyposis confined to the middle meatus. Audiometry demonstrated left-sided hearing loss and mild-to-moderate mixed hearing loss on the right side.

The infection responded well to a course of topical antibiotics and steroids, and the patient noted a marked hearing improvement. Six months later, the tympanic membrane was dry and intact. At the most recent review in January 2013, her aural symptoms had markedly improved despite recurrence of a discharging right pinhole perforation, which has since resolved. The need for major surgery was thus avoided.

Discussion

Although few reports have described the aural manifestations of Samter's triad, we found conservative management to be useful. Aural polyps alone are typically unilateral and usually the result of chronic inflammation or cholesteatoma of the mastoid and middle ear, although they may rarely indicate an underlying malignant process. The aural polyps associated with Samter's triad documented here and in the other two published cases were bilateral and presented with significant otorrhoea and conductive hearing deficit.[4,7]

Histological analysis of aural polyps in Samter's triad reveals polypoidal tissue covered in squamous and respiratory-like epithelium with dense eosinophilic inflammation.[7] Regions of mucin with degranulated eosinophils are also present, as demonstrated histologically in case 1. In contrast, aural polyps related to chronic inflammation tend not to have dense eosinophilia, and feature granulation tissue containing blood vessels and columnar cells (resembling those of respiratory tissue) that are susceptible to undergoing squamous metaplasia.[7] Mucosa of the middle ear and nasal cavity resemble pseudostratified ciliated columnar epithelium. It is interesting that, despite their anatomic proximity, aural polyps are not a common feature in patients with severe chronic rhinosinusitis with nasal polyps.

- **Samter's triad manifests as chronic rhinosinusitis with nasal polyposis, asthma and aspirin intolerance in a non-atopic patient**
- **Aural disease is not widely reported in these patients, and most have undergone major surgery**
- **Minimal surgical intervention is recommended**
- **We report two cases of patients with Samter's triad and aural involvement, in both of whom conservative management was successful**

All reported Samter's triad patients with aural involvement have been females with a mean age of 52 (range 40–59). Aural manifestations were conductive hearing loss and significant otorrhoea, with some otalgia. In the two previous reports, patients underwent aural polypectomy with tympanomastoidectomy and tympanostomy tube insertion. The first patient continues to be treated with serial tube placement (following extrusion) and topical steroid treatments.[4] Brobst and colleagues therefore advocated administration of intratympanic antibiotics, intratympanic steroids and oral steroids to provide the best improvement in hearing thresholds. Surgical management of their patient proved unsuccessful. In the second case report by Shen *et al*., a polyp was noted to be emerging through the tympanostomy tube seven weeks after surgery. This was removed and cauterised, and there was no evidence of aural polyp recurrence at the three-month follow up.[7] However, this is a short post-operative follow-up period for a chronic condition and it remains to be seen whether aural polyposis recurs.

Our first patient developed bilateral tympanic membrane perforations, one spontaneously and one following insertion of a tympanostomy tube which subsequently rapidly extruded. Her disease has been mainly managed with steroids (topical and oral), and topical antibiotics when required. She continues to experience intermittent bilateral otorrhoea. An extensive follow-up period demonstrated steroid administration to be of great benefit; upon

https://doi.org/10.1017/S0022215114002722 Published online by Cambridge University Press

examination in September 2013, her ear symptoms were stable with no otorrhoea. Conservative management with topical steroids and antibiotics has also been successful in our second patient.

Some studies have shown that aspirin desensitisation therapy, with the resultant induction of immunological tolerance, can form part of the management of Samter's triad patients.[8] However, the success rate is generally considered to be very poor; indeed, following discussion with an expert, neither of our patients formally underwent this therapy. The first patient also trialled an aspirin-free diet for six months, but with no effect on her symptoms.

Conclusion

Considering the well-documented natural history of nasal polyposis in Samter's triad, it was decided that further surgical management should be avoided unless absolutely necessary. Shen *et al*. state that the 'tendency to recur warrants complete surgical resection and concomitant medical management'.[7] However, the tendency to recur suggests that minimal operative intervention is warranted. In our patients, symptom amelioration following minimal surgical intervention was similar to that reported after more extensive surgery. Therefore, we question the need for major surgery and recommend conservative therapy as the optimal management strategy.

References

1 Samter M, Beers RF Jr. Intolerance to aspirin. Clinical studies and consideration of its pathogenesis. *Ann Intern Med* 1968;**68**:975–83
2 Widal F, Abrami P, Lermoyez J. Anaphylaxis and idiosyncrasy [in French]. *Allergy Proc* 1993;**14**:373–6
3 Samter M, Beers RF Jr. Concerning the nature of intolerance to aspirin. *J Allergy* 1967;**40**:281–93
4 Brobst R, Suss N, Joe S, Redleaf S. Bilateral inflammatory aural polyps: a manifestation of Samter's triad. *Int J Otolaryngol* 2009;**2009**:464958.
5 Ying S, Meng Q, Scadding G, Parikh A, Corrigan CJ, Lee TH. Aspirin-sensitive rhinosinusitis is associated with reduced E-prostanoid 2 expression on nasal mucosal inflammatory cells. *J Allergy Clin Immunol* 2006;**117**:312–18
6 Majithia A, Tatla T, Sandhu G, Saleh HA, Clarke PM. Intracranial polyps in patients with Samter's triad. *Am J Rhinol* 2007;**21**:59–63
7 Shen K, Peterson M, Mafee M, Nguyen QT. Aural polyps in Samter's triad: case report and literature review. *Otol Neurotol* 2012;**33**:774–8
8 Pfaar O, Klimek L. Aspirin desensitization in aspirin intolerance: update on current standards and recent improvements. *Curr Opin Allergy Clin Immunol* 2006;**6**:161–6

Address for correspondence:
Miss P Sethukumar
Department of Otolaryngology,
St. Mary's Hospital,
Paddington,
London W2 1NY, UK

E-mail: priya00sk@gmail.com

Miss P Sethukumar takes responsibility for the integrity of the content of the paper
Competing interests: None declared

https://doi.org/10.1017/S0022215114002722 Published online by Cambridge University Press