CASE REPORT

Recurrence of eosinophilic oesophagitis with subcutaneous grass pollen immunotherapy

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SUMMARY

Case reports have described an association between oral food/aeroallergen immunotherapy with the development of eosinophilic oesophagitis (EoE). The underlying mechanism of this is poorly understood, as is the role that both food/aeroallergen sensitisation plays in the pathogenesis of EoE. Specific immunotherapy has a long-standing history of use in the management of moderate/severe seasonal allergic rhinitis (AR), caused by tree/grass pollens. Subcutaneous immunotherapy (SCIT) to grass pollen is less commonly used in children than sublingual immunotherapy (SLIT) or oral immunotherapy for practical reasons. We describe a case of a child with severe grass-pollen related AR and known, but quiescent, EoE, who developed recurrence of oesophageal symptoms on two separate occasions, coincident with the commencement of SLIT to grass pollen. He was subsequently started on SCIT to grass pollen and developed recurrence of symptoms of EoE—a phenomenon that has yet to be reported in the medical literature.

BACKGROUND

A rise in the prevalence and recognition of eosinophilic oesophagitis (EoE) has been seen alongside the rise in IgE-mediated allergic disease in western countries, and concomitant allergic diseases such as asthma, allergic rhinitis and eczema are common in children with EoE.¹ The role of IgE food sensitisation in patients with EoE is unclear, but elemental diets are often used in its management with strong evidence of success.²

Aeroallergens have also been linked to the development of EoE. Mishra *et al* described an aetiological role for aeroallergen exposure in animal models.³ Fogg *et al*⁴ described severe eosinophilic inflammation of the oesophagus on biopsies taken in pollen season in a patient with EoE with seasonal symptoms. Other studies have observed a correlation between season and symptoms of EoE, supporting the link between aeroallergens and the development of EoE.^{5–7}

Specific immunotherapy has a long-standing history of effective use in the management of moderate to severe seasonal allergic rhinitis, caused by tree or grass pollens when maximal pharmacotherapy with intranasal steroids and antihistamines has provided inadequate symptom relief. In the UK, subcutaneous immunotherapy (SCIT) to grass pollen is less commonly used in children than sublingual immunotherapy (SLIT) for practical

reasons, but still offers an alternative treatment option for children who may not tolerate SLIT or for those in whom it is not tolerated, adhered to or contraindicated.

An association between initiation of oral food immunotherapy and the development of EoE has been reported. A systematic review by Lucendo *et al*⁸ found that new-onset EoE developed in up to 2.7% of patients undergoing oral immunotherapy for food allergy. Case reports have also described the association between pollen SLIT and the development of EoE.⁹ However, the authors could not find any case reports in the literature describing the development or recurrence of EoE following SCIT for an aeroallergen.

CASE PRESENTATION

A 10-year-old boy was reviewed in our allergy clinic for consideration of immunotherapy to grass pollen. He had a history of nut allergy, allergic rhinitis, atopic asthma and EoE.

Asthma was reported to be seasonal and was well controlled on inhaled steroid (fluticasone 200 µg inhaled, twice daily), with normal spirometry.

EoE was diagnosed histologically at age 8 with 45 eosinophils per high-power field in the lower oesophagus on endoscopy. Symptoms were reported to be seasonal, and our patient was treated with viscous budesonide and omeprazole for flares. Skin prick testing showed sensitisation to grass pollen (12 mm), house dust mite (7 mm) and alternaria (7 mm).

At the time of review (in the autumn), symptoms of EoE were under control.

Seasonal allergic rhinitis was suboptimally managed with a nasal fluticasone furoate spray (27.5 µg), montelukast 5 mg and an oral antihistamine. Our patient had previously been commenced on SLIT to grass pollen (five grass mix, Staloral; Stallergenes) on two occasions, but had been unable to tolerate treatment due to recurrence of symptoms of EoE.

The first trial of SLIT (begun during the late autumn), 10 months previously, had resulted in abdominal pain and symptoms of reflux 2 hours after the first dose. The dose was then tolerated for a week, but, on up-dosing, symptoms of abdominal pain and reflux recurred. The clinical impression was that of recurrence of EoE and therefore the SLIT was stopped after 1 week and he was commenced on budesonide slurry for 3 weeks with resolution of symptoms of EoE.

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The second trial of SLIT was commenced 2 months later (winter) on a slower up-dosing regimen with preventative protection with viscous budesonide and omeprazole. Our patient tolerated up-dosing to maintenance dose over 3 months, but he could not tolerate a decrease in the viscous budesonide on this dose.

The family were keen that the patient should receive desensitisation treatment, and thus he received a trial of SCIT to grass pollen, commencing in the winter months. The patient tolerated the first two injections of 0.1 and 0.2 mL (Allergovit, grass pollen) but developed reflux symptoms following the third dose (0.4 mL). After the fourth injection (0.8 mL), he developed significant vomiting, warranting an inpatient stay in hospital and a course of oral steroids. Despite no further SCIT, symptoms of EoE worsened over the following month, requiring two further inpatient hospital stays, further oral steroids and elemental diet for 2 weeks. A repeat endoscopy was not performed since symptoms were consistent with his initial presentation of EoE and improved with steroid treatment.

OUTCOME AND FOLLOW-UP

At review the following month, symptoms had settled and further immunotherapy has not been recommenced.

DISCUSSION

The role of aeroallergens in the pathogenesis of EoE has yet to be established. However, seasonal differences in disease prevalence and recurrence have been reported and case reports have described recurrence of symptoms of EoE associated with SLIT to aeroallergens.

We have described the first case in the literature of recurrence of EoE following SCIT. We suggest that the exposure to subcutaneous aeroallergen immunotherapy induced a similar immune

response to SLIT, resulting in recurrence of EoE. The exact mechanism underlying this is not fully understood but, in this case, cannot be as a result of the direct exposure of the allergen on the oesophageal mucosa. Patients with a history of EoE who are offered immunotherapy (both SCIT and SLIT) should be counselled about the risk of recurrence of disease.

Further studies are needed to help understand the role of aeroallergens in the development of EoE and may help to identify individuals at higher risk.

The possibility of using immunotherapy as a treatment option for EoE in children has been described¹⁰ but requires a better understanding of the disease, and further studies, before this could be a realistic option.

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REFERENCES

- 1 Miehke S. Clinical features of eosinophilic esophagitis in children and adults. *Best Pract Res Clin Gastroenterol* 2015;29:739–48.
- 2 Spergel JM. Eosinophilic esophagitis in adults and children: evidence for a food allergy component in many patients. *Curr Opin Allergy Clin Immunol* 2007;7:274.
- 3 Mishra A, Hogan SP, Brandt EB, et al. An etiological role for aeroallergens and eosinophils in experimental esophagitis. *J Clin Invest* 2001;107:83–90.
- 4 Fogg MI, Ruchelli E, Spergel JM. Pollen and eosinophilic esophagitis. *J Allergy Clin Immunol* 2003;112:796–7.
- 5 Ram G, Lee J, Ott M, et al. Seasonal exacerbation of esophageal eosinophilia in children with eosinophilic esophagitis and allergic rhinitis. *Ann Allergy Asthma Immunol* 2015;115:224–8.
- 6 Jensen ET, Shah ND, Hoffman K, et al. Seasonal variation in detection of oesophageal eosinophilia and eosinophilic oesophagitis. *Aliment Pharmacol Ther* 2015;42:461–9.
- 7 Green DJ, Cotton CC, Dellon ES. The role of environmental exposures in the etiology of eosinophilic esophagitis: a systematic review. *Mayo Clin Proc* 2015;90:1400–10.
- 8 Lucendo AJ, Arias A, Tenias JM. Relation between eosinophilic esophagitis and oral immunotherapy for food allergy: a systematic review with meta-analysis. *Ann Allergy Asthma Immunol* 2014;113:624–9.
- 9 Miehke S, Alpan O, Schröder S, et al. Induction of eosinophilic esophagitis by sublingual pollen immunotherapy. *Case Rep Gastroenterol* 2013;7:363–8.
- 10 Ramirez RM, Jacobs RL. Eosinophilic esophagitis treated with immunotherapy to dust mites. *J Allergy Clin Immunol* 2013;132:503–4.

Learning points

- There is a known association between the development/recurrence of eosinophilic oesophagitis with use of oral immunotherapy.
- This case report suggests the association may exist for the use of subcutaneous immunotherapy.
- The underlying mechanism for this is poorly understood, but this case suggests that the observation is not due to direct exposure of the aeroallergen onto the oesophageal mucosa.

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