CASE REPORT

Oral Tolerance Induction in Adults with Allergy to Gluten-Containing Cereals: A Case Report

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ABSTRAK

Bijirin ialah sumber utama bagi tenaga, protein, vitamin B dan mineral untuk populasi seluruh dunia. Disebabkan gandum senang dituai dan ketahanannya, ia ditanam dan dimakan di seluruh dunia. Alahan terhadap gandum adalah jarang, prevalens di kalangan kanak-kanak ialah 0.5-1.0%, sering berlaku di Jepun dan bahagian utara Europa dan di kalangan dewasa adalah lagi jarang, tetapi juga dibincangkan. Prognosis alahan terhadap gandum pada zaman kanak-kanak adalah positif, tetapi terdapat kes serious yang berterusan sehingga zaman remaja atau dewasa; dalam kes tersebut, induksi toleransi oral (OTI) dijadikan sebagai terapeutik alternatif kepada penghindaran diet. Kami membentangkan kes yang melibatkan seorang pesakit berumur 32 tahun yang mempunyai alahan gandum dengan serious anafilaksis. Sehingga sekarang, tiada laporan berkaitan dengan OTI dalam alahan gandum di pesakit dewasa yang seperti dibincangkan dalam laporan ini.

Kata kunci: Alahan gandum; alahan gluten; dewasa; penyahpekaan; protokol OTI

ABSTRACT

Cereals are the main source of energy, protein, B vitamins and minerals for the world population. Due to its ease of harvest and its resistance, wheat is the most cultivated and consumed cereal worldwide. Allergic reactions to wheat are rare, their prevalence in pediatric patients varies from 0.5-1.0%, being more frequent

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in areas such as Japan and northern Europe and in adults, although less frequent, they have also been described. The prognosis of allergy to wheat in childhood is favourable, but there are more severe cases that remain allergic until adolescence or adulthood; in these cases, oral tolerance induction (OTI) can be considered as a therapeutic alternative to the avoidance diet. We presented a case of 32-year-old patient with the onset of wheat allergy in adulthood with severe anaphylaxis. To date, there is no report of OTI with wheat in allergic adult patient, similar to the one described in this report.

Keywords: Adults; desensitisation; gluten allergy; OTI protocols; wheat allergy

INTRODUCTION

Cereals are the major source of energy, protein, B vitamins, and minerals for the world population. Due to its ease to be harvest and its resistance, wheat is the most grown and consumed cereal worldwide (Food and Agricultural Orginisation of the United Nations 2020).

Allergic reactions to wheat are rare. Prevalence of IgE mediated wheat allergy in paediatric patients varies from 0.5-1.0% (Nwaru et al. 2014), being more frequent in areas as Japan and northern Europe. In adults, although less frequent, it has also been described (Scibilia et al. 2006).

The prognosis of wheat allergy in childhood is favorable. More than half of paediatric patients develop wheat allergy, with a mean age of resolution of 79 months according to Keet et al. (2009). Children with high titers of wheat-specific IgE and more severe clinical manifestations are more likely to remain allergic to wheat until adolescence or adulthood (Koike et al. 2018).

In these cases, oral tolerance induction (OTI) may be considered as a therapeutic alternative to the avoidance diet (Nowak-Wegrzyn et al 2019; Rodríguez Del Río et al. 2014; Vila et al. 2015). So far, there is no report on OTI with wheat in adult allergic patients, as the one we described below.

CASE REPORT

We presented a 32-year-old woman referring recurrent outbreaks of generalised urticaria during the last 6 months. She had scattered wheezy lesions on the skin surface, but no lip, eyelid, or facial angioedema. Urticaria outbreaks lasted a few hours and were controlled with cetirizine as needed. She did not relate it to any trigger (cold, heat, pressure, medications such as nonsteroidal anti-inflammatory drugs (NSAIDs) or food).

RESULTS

Allergy study was performed with intraepidermal skin prick test (SPT) with the common aeroallergens in our area, latex, panallergens as profilin,

lipid transfer protein and polcalcin and milk, egg, white fish, blue fish, wheat flour, soy flour, gluten, legumes, shellfish, crustaceans, nuts and anisakis (Roxall Laboratory, Bilbao, Spain), yielded positive results to dust mites, wheat flour and gluten. The patient tolerated wheat-based foods and she did not relate the episodes of urticaria with wheat intake. She did not report respiratory symptoms that could be related to dust mites either.

Laboratory tests including complete blood count, biochemistry, acute phase reactants, thyroid hormones and IgA, IgG and IgM, were performed, all within normal ranges. Total IgE was 228 UI/ml. Serum specific IgE for gluten was 1.73 kUIa/L, Gliadina, 0.6 kUIa/L and Tri a 19 3.61 kUIa/L. At this moment, no dietary restrictions were made and oral antihistamines and steroids were prescribed in case of new episodes of urticaria.

Two months later she came back to the clinic. She had been avoiding cereals for four months due to a self-proposedly lose-weight diet. After finishing the cereal free diet, she ate a king size pizza for dinner and within few minutes she developed generalised urticaria and loss of consciousness. She received home medical assistance requiring intramuscular adrenaline, as well as parenteral antihistamines and steroids. She underwent hospital observation for 24 hours with complete resolution of symptoms.

Strict gluten avoidance was indicated. The patient failed to adhere to the proposed diet and continued ingesting small amounts of cereals developing mild episodes of urticaria. She reported bad quality of life. Therefore, we decided to perform oral tolerance induction with gluten-containing cereals.

Briefly, to establish the starting dose for the OTI, an open oral challenge test (OCT) was performed, with increasing doses of baby food flour with eight cereals with gluten (Nestle). Ten minutes after acumulative dose of 60 g of the baby food the patient presented an acute generalised urticaria and abdominal pain that required treatment with oral antihistamines and steroids. OTI was started with 30 g of baby food flour. The amount was increased on a weekly basis up to 120 g of flour. Then, baby food was replaced by Marie biscuits (Artiach) to ensure palatability. OTI was finished when a total amount of 180 g of wheat pasta (Gallo brand spaghetti) was reached (Table 1).

During the induction phase, the patient did not present any adverse effects. Since then, she had been consuming cereals with gluten on a daily basis for two years without presenting any allergic reactions. Her quality of life had improved significantly.

DISCUSSION

Wheat allergy is rare in adult population though it seems to have a prognosis at least as favorable as in the pediatric age, as reported by Scibilia et al (2019). In their study, 9 out of 10 adult patients allergic to wheat, achieved tolerance after a mean period of 4.2 years of avoidance diet. Even so, since cereals containing gluten are so ubiquitous, eliminating them from the diet may be

TABLE 1: Oral tolerance induction with wheat

Week	Food	Dose
1	Cereal baby food	30 g
2	Cereal baby food	60 g
3	Cereal baby food	90 g
4	Cereal baby food	120 g
…	Marie biscuit	150 g
…	Wheat pasta	180 g

*Cereal baby food contained 11.8 g of protein/100 g. **Marie biscuit contained 6.5 g of protein/100 g. Wheat pasta contained 12 g of protein/100 g.

difficult for allergic patients. In these cases, when quality of life is affected, OTI with wheat may be considered.

The few studies on OTI with wheat have been carried on in children with different outcomes. These results might be related to the different designs of the protocols as well as the clinical and serological characteristics of the patients included. The rate of desensitisation reported, ranged from the 52% (Nowak-Wegrzyn et al. 2019) to the 80% (Sato et al. 2015) and 83% (Rodríguez Del Río et al. 2014).

This was the first report on OTI with wheat in an adult patient. We followed the same protocol published previously by us (Vila et al. 2015). In that study, four paediatric patients achieved desensitisation tolerating 100 g of wheat bread at the end of the induction phase. One patient further developed symptoms after wheat ingestion and progressively eliminated it from his diet. Other three patients have a normal and non-restricted intake of gluten containing cereals. The patient presented here has completed this OTI protocol without relevant reactions, being desensitised two years after finishing the escalating phase.

CONCLUSION

To improve quality of life of adult patients who allergic to gluten-containing cereals but avoiding restrictive diets, OTI may be considered as a valid therapeutic option.

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