

A rare case in an adolescent patient with nasal polyps resistant to polypectomy: Samter's syndrome

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Abstract

The comorbidity of recurrent nasal polyps, non-steroidal anti-inflammatory drug hypersensitivity and chronic rhinosinusitis is called Samter's syndrome. It is less common in childhood than in adulthood. Patients first have nasal symptoms and then asthma and non-steroidal anti-inflammatory drug (NSAID) hypersensitivity begin to accompany. For definitive diagnosis, NSAID or aspirin hypersensitivity must be shown with provocation test. In this case report, oral provocation test with ibuprofen was performed on a 17-year-old female patient who had recurrent nasal polyps and chronic rhinosinusitis accompanied by asthma and NSAID hypersensitivity and the test was found to be positive. The patient was diagnosed with Samter's syndrome and aspirin desensitization was applied. Inhaled steroid, intranasal steroid and montelukast therapies were started. As a conclusion, although it is more common in adults, Samter's syndrome should also be considered in pediatric patients with recurrent nasal polyp, chronic rhinosinusitis, asthma, and NSAID hypersensitivity.

Keywords: Adolescent; aspirin; nasal polyps; samter's syndrome

INTRODUCTION

Samter's syndrome is characterized by recurrent nasal polyp, chronic rhinosinusitis, asthma, and hypersensitivity to aspirin and other non-steroidal anti-inflammatory drugs (NSAID) (1). Although the pathophysiology of the disease is not fully understood, the underlying mechanism is thought to be a disorder in arachidonic metabolism (2, 3). Although asthma is thought to be at the forefront in this disease, nasal symptoms develop first and asthma starts later in the patients. Patients frequently undergo surgical operation due to recurrent nasal polyposis and chronic sinusitis. Asthma attack and rhinitis that start after patients take acetylsalicylic acid (ASA) or other NSAIDs make us suspect Samter's syndrome. Definitive diagnosis of the disease is made with ASA provocation test (4).

Samter's syndrome is less common in children than adults because aspirin use is not common in childhood, except for rheumatologic diseases (5). This case report presents a 17-year-old adolescent with Samter's syndrome who was operated many times due to recurrent nasal polyp and frequent sinusitis, who had a history of hypersensitivity with multiple NSAIDs and who later underwent desensitization with ASA.

CASE REPORT

Seventeen-year-old female patient referred to otorhinolaryngology (ORL) clinic due to recurrent nasal polyp. It was learned from her anamnesis that she had been diagnosed with nasal polyp three years ago due to nasal obstruction, nasal discharge and anosmia. She had also had myringotomy operation due to frequent sinusitis and otitis. It was learned that she had undergone polypectomy operation twice due to nasal polyp, but nasal polyps formed again shortly after the operation. It was also learned that she used intranasal steroid therapy during this period, but she partially benefited from this therapy. The patient had shortness of breath, which was especially due to effort, for the last year. She did not describe allergic rhinitis and atopic eczema. It was learned that during this period diffuse urticaria developed in the body and angioedema developed on her lips after using ibuprofen twice and naproxen once for headache. The complaints of urticaria and angioedema had regressed spontaneously in a short time. Physical examination showed nasosplenomegaly and polyp in both nasal passages. The patient was consulted to the pediatric allergy clinic for advanced tests with a pre-diagnosis of Samter's syndrome. The patient's skin prick test performed in the allergy clinic did not show

Received: 06.04.2021 **Accepted:** 30.04.2021 **Available online:** 25.06.2021

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aeroallergen sensitivity. Respiratory functioning test (RFT) was found to be normal; however, reversibility test was found to be positive in RFT. Serum immunoglobulin levels and sweating test were normal. Eosinophil was 12.8% (850/mL) in the blood count. Oral provocation test (OPT) was performed on the patient with ibuprofen in the pediatric allergy clinic. Shortness of breath, diffuse urticaria and angioedema on the lips developed in the first dose of OPT, examination showed diffuse rhonchi in the lungs, 30% decrease in FEV1 occurred in SFT and the test was terminated. Oral provocation test was performed with meloxicam to determine the safe NSAID to use in case of need and no reaction was observed. Meloxicam was determined as safe NSAID for the patient. The patient who had NSAID allergy, nasal polyp, chronic sinusitis and asthma was diagnosed with Samter's syndrome. The patient was started inhaled steroid therapy for asthma and intranasal steroid and montelukast therapy for nasal polyps. Desensitization was performed with aspirin to decrease polyp recurrence (6). After aspirin desensitization, the patient was recommended to use prophylactic (regular) aspirin every day. No new polyp formation and sinusitis attack was observed in the patient who had been followed up in the ORL clinic for nine months. A significant improvement occurred in asthma complaints during this process.

DISCUSSION

Samter's syndrome is more common in the adult age group. It is rarely seen in childhood and adulthood. Nasal symptoms such as runny nose, nasal obstruction and anosmia are in the foreground in patients with Samter's syndrome. Patients have recurrent nasal polyps after polypectomy. These patients may require repeated surgical operations such as myringotomy due to chronic sinusitis. Type 1 hypersensitivity and asthma can also be seen with NSAID intake (2,4,6). Our patient was in the adolescent age group where Samter's syndrome is rare. Our patient had runny nose, nasal obstruction, anosmia, frequent sinusitis and nasal polyp resistant to surgical treatment. Multiple NSAID hypersensitivity and asthma had developed later.

Although the pathophysiology of the disease is not fully understood, the underlying mechanism is thought to be arachidonic acid metabolism disorder. Aspirin and other NSAIDs block the cyclooxygenase pathway of arachidonic acid metabolism and leads to an increase in leukotrienes, which are metabolites of the lipoxygenase pathway. With the activation of mast cells and eosinophils, leukotrienes increase and this in turn causes inflammation. For this reason, it is recommended to start leukotriene antagonist therapy to patients with Samter's syndrome (4-7). This case was also started leukotriene antagonist (montelukast) therapy in pediatric allergy clinic.

Avoiding NSAIDs that inhibit COX-1 plays an important role in the treatment of patients diagnosed with Samter's syndrome. However, the NSAID that patients will use without reaction in case of need should be determined.

Nimesulide or meloxicam that cause selective COX-2 inhibition can be preferred in these patients (8). Our patient developed type 1 hypersensitivity following ibuprofen and naproxen use. Nimesulide was determined as the NSAID that our patient could use in case of need.

In patients with Samter's syndrome, desensitization with aspirin is recommended in case of recurrent nasal polyp despite polypectomy (1,2). Although studies on the effect mechanism of aspirin desensitization are still continuing, there is no clear information yet (9,10). Daily use of aspirin reduces the need for surgery such as polypectomy and myringotomy, as well as nasal symptoms such as runny nose, nasal obstruction and anosmia (11,12). In our patient, nasal symptoms were found to decrease significantly when under aspirin desensitization.

CONCLUSION

As a conclusion, Samter's syndrome should be considered in patients who have recurrent nasal polyps despite surgical treatment, who have a history of reaction after NSAID intake, who describe asthma symptoms and who have chronic rhinosinusitis. These patients should be consulted to pediatric allergy clinics and should be followed with this clinic. Aspirin desensitization should be applied in addition to medical therapies in cases with definitive diagnosis.

Conflict of interest: The authors declare that they have no competing interest.

Financial Disclosure: There are no financial supports.

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