

# A rare cause of recurrent nasal polyposis in adolescent age group: Samter's syndrome

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Dear Editor

Nasal polyposis is defined as classical tetrad Samter's syndrome in the form of hypersensitivity that develops due to chronic sinusitis, asthma and aspirin and other non-steroidal anti-inflammatory drug (NSAII) (1). Samter's syndrome, or Aspirin exacerbated respiratory disease as frequently used by adult cases, has been called NSAII exacerbated respiratory disease according to final report (2). This condition, which is well-known in adult asthma cases, is found in rates as high as 21% when provocation test (OPT) is conducted in this group (3). Rare reports only in the form of case reports exist in children (4). Reaction spectrum consisting of rhinitis, conjunctivitis, laryngospasm or bronchospasm occurs in the upper and/or lower respiratory tracts of the cases following aspirin or other NSAII intake (5). Due to recurrent nasal polyps and chronic sinusitis, these patients may need upper respiratory tract surgery frequently.

16-year-old male patient was consulted to otorhinolaryngology polyclinic for recurrent nasal polyp. It was learned that he had dyspnea on exertion for the past year, he experienced nasal blockage all the time and that nasal polypectomy was applied previously. He did not describe any comorbid allergic rhinitis or atopic eczema. It was learned that once following ibuprofen tablet intake and once following naproxen tablet intake, the patient's lips swelled with rashes in the body in two hours and the swellings recovered spontaneously in a day.

Respiratory function test (RFT) was normal. Skin test did not show aeroallergen sensitivity. In the whole blood count, 6% eosinophil was attention getting. The patient who had NSAII allergy, nasal polyp and dyspnea for the past year was thought to have Samter's syndrome. Since

he described multiple NSAII allergies and he was thought to have Samter's syndrome, oral provocation test with aspirin was planned. After oral provocation doses with aspirin were completed, he had a dyspnea complaint within the first hour. Auscultation revealed bilateral rhonchus at both lungs. Later, swelling developed in the eyelid. The patient who had 95% saturation was given short-acting bronchodilator and oral antihistaminic treatment. He was started inhaled steroid, oral montelukast and nasal steroid therapy for his asthma. Aspirin desensitization was planned to decrease polyp recurrence.

Samter's syndrome is generally characterized in adult women and it has a typical chronological course. Nasal congestion, anosmia, chronic pansinusitis, and polyp relapsing after surgery which occur in the third decade are seen first (5). While asthma symptoms develop a few years later, NSAII intolerance develops four years later (6). While asthma severity can be within intermittent/mild-severe interval, there may also not be any asthma complaints. However, most of the time asthma has a severe course and is poorly controlled and requires regular systematic corticosteroid use despite inhaler treatment (6,7).

Samter's syndrome is characterized with the chronic eosinophilic inflammation of upper and lower respiratory tracts. Although the pathogenesis of inflammation is not completely understood, it is due to symptoms that occur with the intake of NSAII chemically different from each other/aspirin intake and abnormal arachidonic acid metabolism that develops as a result of cyclooxygenase-1 (COX-1) enzyme inhibition. The reaction that occurs this way is not immunological and it is called cross-reactive NSAII hypersensitivity. It is decomposed in the lipoxygenase pathway due to arachidonic acid COX-1

Received: 05.09.2019 Accepted: 10.10.2019 Available online: 09.12.2019

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inhibition, leukotrienes increase with the activation of mast cells and eosinophil activation and it results in inflammation (8). Based on this, it is recommended to start preoperational leukotriene antagonist (montelukast) in order to prevent aspirin provocation and serious bronchospasm that will develop during desensitization that will be applied later (5).

History of asthma attack with aspirin or other NSAID strengthens the diagnosis for Samter's syndrome; however, as in our case, hypersensitivity with NSAID should be proven for actual diagnosis (8). In addition to isolated naso-ocular reactions such as itching, sneezing, obstruction and conjunctivitis following aspirin/NSAID intake during the test, bronchospasm occurring with FEV1 15%  $\geq$  decrease can be added to this. Rarely, gastric pain defined as systemic reaction, hypotension, urticaria or angioedema can accompany these, as in our case. Each of the four conditions that may occur during the test is diagnostic (5).

Aspirin desensitization may be necessary in these patients. The primary indication of Aspirin desensitization in Samter's syndrome is recurrent polyp formation which requires operation and which cannot be controlled with treatment, as in our patient (1). It has been shown in many studies that daily aspirin use with Aspirin desensitization provides obvious recovery in all nasal symptoms and quality of life and obvious increase in oral corticosteroid use and sinus surgery requirement with sinusitis and polyp formation (7,9). The need for re-operation every three years decreases to every ten years (1). Although there are studies about the effect mechanism of aspirin desensitization in molecular level, they are not clear yet (10). Studies about the optimal daily dose and the period of Aspirin desensitization also differ (7,9-11).

As a conclusion, every pediatric patient with asthma, chronic rhino sinusitis, nasal polyp or describing NSAID reaction can be a Samter's syndrome candidate. Suspected cases should be referred to allergy clinics for definitive diagnosis and follow-up and it should be known that Aspirin desensitization applied to cases with indication in experienced centers is also a treatment option.

*Competing interests: The authors declare that they have no competing interest.*

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