CASE REPORT



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Sneezing – a symptom of respiratory or psychogenic superposition of illness in a teenager?

Kijanje – simptom respiratorne ili psihogene superpozicije bolesti u tinejdžera?

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Abstract

Introduction. The coincidental combination of allergic respiratory diseases and psychogenic or psychiatric illness is possible but rarely associated in a female teenager. Case report. A girl aged 12.5 years was admitted to the Pediatrics Clinic in the Clinical Centre Kragujevac with the main difficulty of sneezing as 10 sneezes in each of the 500-600 series a day, only in the waking state. Working diagnoses were tic disorders associated with Allergic rhinitis, asthma and biochemically determined rickets. The patient was treated with chlorpromazine, desloratadine, montelukast, fluticasone propionate, vitamin D, azelastine hydrochloride along with the elimination diet. After a year and a half, the patient rarely sneezed, but periods without sneezing were not longer than 3 months, and she 'cleared her throat' occasionally. Although her clinical condition is less severe now and without additional motor tics or symptoms of Tourette's disorder, because of its length we suspect the tic turned into a chronic vocal tic disorder. Conclusion. We reported a case of rare association between allergic rhinitis and condition of tic-related conversion in a female teenager and emphasized the necessity of revising the systematisation for the tic disorders and protocol for the allergic rhinitis in terms of their association.

Key words:

adolescent; rhinitis, allergic; tic disorders.

Apstrakt

Uvod. Udruženost alergijskih respiratornih oboljenja i psihogene ili psihijatrijske bolesti je moguać, premda se javlja retko kod devojčica u tinejdžerskom uzrastu. Prikaz bolesnika. Devojčica uzrasta 12,5 godina primljena je u Pedijatrijsku kliniku Kliničkog centra Kragujevac sa glavnom tegobom kijanje, po 10 uzastopnih kijanja u serijama od 500-600 dnevno i to samo u budnom stanju. Radne dijagnoze bile su tik poremećaj udružen sa alergijskim rinitisom, astmom i biohemijski utvrđenim rahitisom. Tinejdžerka je lečena hlorpromazinom, desloratadinom, montelukastom, flutikazon propionatom, vitaminom D, azelastin-hidrohloridom, sa pratećom eliminacionom ishranom. Posle perioda od godinu i po dana, bolesnica je ređe kijala, periodi bez kijanja nisu bili duži od 3 meseca i povremeno je "čistila grlo". Mada je sada klinička slika bila blaža i bez dodatnog motornog tika ili simptoma Tourette poremećaja, zbog dužine trajanja tegoba postoji sumnja da se tik preobratio u hronični glasovni tik poremećaj. Zaključak. U radu je prikazana retka udruženost alergijskog rinitisa i stanja tik-srodne konverzije kod tinejdžerke i istaknuta je potreba za reviziju sistematizacije tik poremećaja i protokola za alergijski rinitis u smislu njihove udruženosti.

Ključne reči: adolescenti; rinitis, alergijski; tik poremećaji.

Introduction

Sneezing in children is the most common symptom of allergic and infective rhinitis. Sneezing (or blowing) provides a high-velocity ejection from the mucosal surface ¹. Sneezing is an involuntary reaction, and a protective reflex that starts in the nasal mucosal lining travels to the medulla oblongata, from which many muscles are stimulated to take a deep breath ². After that the breathing stops for a short time,

the soft palate rises and closes the inner nasal opening. At the same time, the respiratory muscles, participating in the exhalation, are strained, and the pressure in the lungs rises. The soft palate lowers, and the air penetrates into the nose under high pressure, goes through the nasal passages removing all foreign and harmful substances from the nose.

As a sudden, strong and uncontrollable expulsion of the air through the nose and mouth, sneezing is rarely a serious health issue in children. The infective rhinitis and allergic

Correspondence to: Andjelka Stojković, Clinical Centre "Kragujevac", Pediatric Clinic, Zmaj Jovina 30, 34 000 Kragujevac, Serbia. Phone: +381 34 505 175; E-mail: andja410@mts.rs rhinitis are, logically, first suspected in primary health care and the treatment begins without testing protocols ³, which turns out in this case as a reason for potentiating the health problem of a patient. Sneezing is, also, a common symptom of nonallergic-noninfective rhinitis, acute and chronic rhinosinusitis and the like.

It is rare that, among the multiple common triggers of sneezing in children, a strong emotion is associated with allergic rhinitis, resulting in frequent and exhausting sneezing prolonged over several days, weeks and even months. As it is presented in this case report, such hardships were put forward for the consideration as a particular disorder, among various psychiatric and psychological disorders in a child or teenager⁴.

Tic, in medicine, does not imply any voluntary action of a body part when a person is excited, stressed, anxious, and after an excessive intake of caffeine or alcohol⁵. In that case, the human body shows that it is under stress or tired, although it was not always so easy to discern, especially during the teenage period. At this age, the frequent mood swings are part of growing up. During puberty, teenagers begin to change physically - most girls get their first period and a beard begins to grow in boys. For teenagers, these changes are very important and not easy to accept. While maturing, teenagers have to deal with a lot of things in their mind, which can contribute to their anxiety, irrational fear or somatization of an organic disease such as the allergic rhinitis. In this case report, the treatment of the allergic rhinitis symptoms in a teenager was difficult, time-consuming and on the edge of being unsuccessful due to the lack of information, about somatization of allergic rhinitis or possible association with tic in the ARIA (the Allergy Rhinitis and its Impact on Asthma) approaches 6.

Case report

We considered the unusual tic related to sneezing in a 12.5-year old female teenager associated with the allergic rhinitis, mild asthma⁷ and rickets. The girl was hospitalized because of sneezing, headache, and constipation in the early winter period. She sneezed in approximately 500–600 daily series of ten sneezing each, and exclusively while being awake. The order of all clinical findings, related to this case, is shown in Table 1. After 3 days of the treatment with chlorpromazine, the teenager sneezed 42 series of 10 sneezing each. Chlorpromazine was taken during following 5 months with a gradual dose reduction. Currently, after a year and a half, the patient rarely sneezes, periods without sneezing have not been longer than 3 months, and she 'clears her throat' occasionally. She had no symptoms of conjunctivitis.

At the beginning, sneezing was accompanied with the fever up to 38.5°C during 10 days, and the therapy was applied in the school infirmary, including xylometazoline nosedrops, paracetamol and ascorbic acid, desloratadine and mometasone furoate nasal spray. Because of the protracted sneezing, pharyngeal and nasal swabs were taken, so that in accordance with the antibiogram, amoxycillin with clavulanic acid was administered over 7 days. Since the teenager

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kept on sneezing despite her good general health, she was sent to the General Hospital for examination.

On admission to the Department of Pediatrics, in the General Hospital in the countryside of Serbia, main reasons for the hospitalization were sneezing, headache and constipation, despite normal physical findings. Diagnostic findings are shown in Table 1. During a 7-day monitoring and examination, the girl sneezed in approximately 200 series of ten sneezing each, suffered from substernal chest pain and constipation. Electroencephalography (EEG) showed the following: irritant activity was noted bilaterally at well-expressed and regular OA 12Hz in the frontal-central-temporal region (FCT). Due to the EEG findings and performed analyses, the patient was empirically treated with diazepam 5 mg tablets for 5 days. However, sneezing continued with variable intensity and frequency, exclusively during the state of wakefulness. As a consequence of constipation, a fissure appeared in the anal region and it was treated with policresulen and cinchocaine ointment and benzydamine solution. She was discharged from the General Hospital with the recommended therapy: omeprazole 20 mg capsule once a day, metronidazole 750 mg daily, lactulosa syrup 30 mL daily and policresulen and cinchocaine ointment.

During 9 days at home, the girl continued to sneeze even more intensely, 10 sneezes in each of the 500–600 series a day, only while being awake, during the following month and a half.

Because of this, the girl was sent to the additional examination to the Pediatrics Clinic in the Clinical Centre Kragujevac. On admission, the girl was eutrophic, complained about fatigue and exhaustion; the pharynx was hyperemic; livid nasal mucosa was without edema; diffuse expiratory wheezing was noted, and other physical findings were normal. Diagnostic findings are shown in Table 1. Detected vitamin D deficiency with elevated alkaline phosphatase and a high total IgE level suggested the association of allergic disease and rickets. Skin prick test clearly showed characteristics, potential risk factors and triggers of allergic diseases. Pulmonary function tests showed moderately constricted small airways along with moderately elevated total and specific airway resistance, mild "air trapping", and a positive bronchodilator response. The female teenager did not have asthma symptoms (a cough, wheezing, shortness of breath, chest tightness) and asthma was diagnosed using functional assessment of airflow limitation and airflow reversibility '. The above mentioned pulmonary and other examinations confirmed the following diagnoses that were intertwined: asthma (organic), rhinitis (both allergic and, predominantly, psychogenic superpositioned), the condition of tic related conversion and biochemically confirmed rickets.

The female teenager was discharged from the Clinic with recommended therapy: chlorpromazine 25 mg at every 8 hours over a month and parents were requested to keep evidence of the number of tics (sneezing) during the day. At the same time, the patient received therapy for asthma, allergic rhinitis and rickets including vitamin D_3 2000 IJ daily, desloratadine 2.5 mg daily, montelukast 5 mg daily, fluticasone propionate 100 mcg inhaled daily, and she was given

Place of treatment	Daily series of 10 sneezing each	Omer discomforts, clinical signs, clinical findings	Diagnostic findings	Therapy
School infirmary /6 weeks before admission to the Pediatric Clinic/	500-600	Fatigue, exhaustion Fever up to 38 5oC over 10 days Hyperemic pharynx Livid nasal muccsa (without edema) RR 22/min, HR 92/min, SaO2 98%, Eutrophic	Pharyngeal swab: A beta-hemolytic <i>streptococcus</i> Nasal swab: <i>Staphylococcus aureus</i> 100%	Xylometazoline nosal-drops, paracetamol, ascorbic acid, amoxycillin with clavulanic acid (7 days)
General hospital /2 weeks before admission to the Pediatric Clinic/	200 500-600	Headache, substernal chest pain, constipation RR 20/min, HR 88/min, SaO2 98%	Physical findings were normal. CRP 0; Le 4.98 × 10/-91; neu.0.48; mo.0.08; Er 5.33 ×10/- 121, Hg 149 g/L, PLT 236 × 10/-9L) EEG: invitant activity was noted bilaterally at well-expressed and regular OA 12Hz over fronto-centro-temporal region. bMRI: normal Abdominal ultrasound: normal Coular fundus review were normal. Pharyngeal and nasal swab: normal Perianal swab at intestinal parasites: normal	Observation Diazepam 5 mg (5 days)
Admission to the Pediatric Clinic After 1 day of treatment After 2 days of After 3 days of After 3 days of treatment	560 280 160 42	Headache, substernal chest pain, constipation	Video-EEG monitoring: bilateral mild nonspecific changes in fronto-central region along with well-presented sleep phases that rated as being normal. Psychologist: suspected tics. Psychiatrist: tic disorder. Serum level of Vitamin D 18 ng/mL, Alkaline phosphatase 236U/L, Total IgE level 379IU/L. Skin Prick test: grass pollen-2 mm, freshwater fish-2 mm, cow's milk-2mm, house dust-2 mm, freshwater fish-2 mm, tests: FVC 81%, FEV1 82%, FEF 75%5 59%	Chlorpromazine 12.5 mg. every 6 hours Chlorpromazine 12.5 mg. every 6 hours Chlorpromazine 12.5 mg. every 6 hours Chlorpromazine 12.5 mg. every 6 hours
The 4th days: Discharge from the clinic to the home treatment			Bronchodilator response +15% FEV1 Specific airway resistance (Sreff 179%, sRtot 204%), RV/TLC 138%, RV 147%, Fres 16.8 TLCO normal ENT examination: allergic thintits, excluded sinusits. Normal results of: C-reactive protein, peripheral blood smear, ionogram with calcium, phosphorus, magnesium ion in seum, chlorine in sweat, the microbiological findings of aspirate samples and nose and throat swabs, radiological examinations of the lungs and left hand, perianal swab for intestinal parasites, abdominal ultrasound findings, and the ocular fundus.	 Chlorpromazine 12.5 mg, every 8 h over a 6 weeks Desloratadine 2.5 mg daily Montelukast 5 mg daily Fluticasone propionate 100 µg inhaled daily with chamber with mask Vitamin D3 2000IJ daily Vitamin D3 2000IJ daily Elimination diet (without freshwater fish, cow's milk, tomatoes, seafood).
After 6 weeks of treatment at the home	15		The lung function and total respiratory system resistance were normal; Serum level of vitamin D 26.64 ng/mL; Alkaline phosphatase 91 U/L	 Chlorpromazine 12.5 mg, every 8 h, still 4 months Desloratadine 2.5 mg daily, still 4 months Momethason furoat 100 ug daily, over a 4 months

advice on the elimination diet (without freshwater fish, cow's milk, tomatoes, seafood). After a month of the treatment, the girl still sneezed up to 15 series per day, the lung function was normal, with vitamin D insufficiency detected and normal alkaline phosphatase activity in the serum.

Chlorpromazine, as a phenothiazine neuroleptic, was taken during 5 months with a gradual reduction of the dose. In the same period allergic rhinitis and asthma were treated (desloratadine, montelukast, fluticasone propionate) and after that, in the following six months only prophylaxis of allergic rhinitis continued (desloratadine, mometasone furoate). After a year and a half period of time, the patient rarely sneezed, periods without sneezing were not longer than 3 months, and she 'cleared her throat' occasionally. Prophylaxis of allergic rhinitis continued using topical antihistamine azelastine hydrochloride along with the elimination diet. Given that the number of tics (sneezing and "throat clearing") was small, psychological treatment, in the form of a counseling and conversation, was also recommended before the next check up in 4 months. Then, we suspected that there was a "hidden anxiety" which with available psychological (projective techniques, Beck Hopelessness Scale, Children's Depression Inventory) and psychiatrist (katamnestic) tests was not established.

The data from the personal history should also be mentioned: 5 months prior to the hospitalization, the female teenager was admitted to the Pediatric Surgical Care because of the episodes of belching and pain in the stomach which were successfully cured with ranitidine. Personal medical history revealed that she was prone to sneezing during the spring and autumn since she was 9 years old, but she not been tested or treated for allergic rhinitis so far.

There was no information on the use of psychoactive substances and drugs or exposure to toxic and other substances or about the similar problems identified in family members.

Discussion

In this paper, we described the case of an unusual tic related to sneezing in a female teenager associated with the allergic rhinitis and asthma. We considered this as "the condition of tic related conversion", what was working diagnosis. Accordingly, we did a differential diagnostic consideration.

Tic disorder is, as suggested by the American Academy of Child and Adolescent Psychiatry, a neuropsychiatric disorder associated with other psychiatric disorders, including attention deficit disorder, hyperactivity and obsessivecompulsive disorder ^{5, 8, 9}. Simple vocal tics indicate personality development disorder, indeed; that usually disappears because of its transient nature and is more common in male children. It is most commonly manifested as a cough, grunting, throat clearing and sniffling. It is transient if it lasts between 4 weeks and 12 months in children under 18 years of age, but if not due to the effects of psychoactive substances or other drugs, and not meeting the criteria of Tourette's disorder or other chronic motor or vocal tic disorders ^{8, 9}. Tic disorder is a neuropsychiatric disorder associated with other psychiatric disorders, including attention deficit disorder, hyperactivity, and obsessive-compulsive disorder ^{5, 7, 10}.

From patient's medical history we were not informed about her or her family suffering from Gilles de la Tourette's syndrome⁹ and we excluded the influence of the various substances and drugs. However, tic-related sneezing in our teenager is not of transient nature (the simple vocal tics)^{5,8,9} because it has lasted for year and a half, and although its clinical picture is less severe now and without additional motor tics, or symptoms of Tourette's disorder, because of its duration, we reflect the tic turns into a chronic vocal tic disorder.

The psychologist and psychiatrist did not find the reason for the teenager's emotional instability or excessive dependence on her parents and the fear of her parental deprivation, especially in receiving love and any exposure to punishment. There is a disorder marked as a psychogenic superposition of allergic rhinitis. Then, the "hidden anxiety" was not established with the available psychological and psychiatrist tests. The symptoms as a headache, constipation, retrosternal pain (with normal physical findings) were perceived as somatic symptoms because they have manifested at the same time with intense sneezing. This was the reason to think about "hidden psychological factors" that affected other medical condition but was not recognized by the Diagnostic and Atatistical Manual of Mental Disorders, Fifth Edition (DSM-5).

After this, a neurologist, as a treating physician, however, prescribed chlorpromazine. The low-potency, typical antipsychotic effect of chlorpromazine ¹⁰ was achieved in this patient. The treating physicians have had in mind the favorable circumstance that chlorpromazine acts as an antagonist on different postsynaptic and presynaptic receptors (dopamine-D1,-D2,-D3,-D4, serotonin-5HT1,-5HT2, histamine H1, α 1- and α 2-adrenergic, M1- and M2-muscarinic acetylcholine)¹⁰, as used here. Chlorpromazine reduced the anxiety and annulled psychogenic superposition of allergic rhinitis. Here is implemented dual therapy of sneezing.

On the other hand, the prevalence rates of allergic rhinitis are highest in school-age children^{2,3,6,7}. Thus, allergic rhinitis and infective rhinitis are logically first suspected in primary health care where, usually, the treatment begins without testing protocols. We were thinking about other types of rhinitis but we did not fit any of the known clinical pictures and received additional diagnostic findings, including sneeze on a full stomach¹¹. It is known that the treatment of moderate/severe persistent allergic rhinitis according to the approach titled the ARIA⁶ guidelines, and the patient should be checked in 2-4 weeks in primary health care. And, in the end, if there is no improvement, a surgeon should be consulted and specific immunotherapy considered. The treatment of allergic rhinitis according to the ARIA protocol⁶ involves stepwise approach³ but without established order of application of decongestant, oral H1-antihistamine, intranasal H1-antihistamine, intranasal corticosteroids (INS), antileukotriene and cromolyn for any severity of intermittent rhinitis and mild persistent rhinitis lasting up to 4 weeks. If there is no improvement, INS dose should be gradually increased or ipratropium bromide or oral corticosteroids should be introduced^{3,10}. Conditions of controlled persistent allergic rhinitis can be achieved by the treatment of the ARIA approach ⁶.

However, the ARIA approach ⁶ implies neither the possibility that allergic rhinitis may be associated with psychogenic or psychological disorders or the attention deficit-hyperactivity disorder (ADHD), nor that the allergic rhinitis is less controlled in the teenagers with tic or ADHD or psychological factors that affect rhinitis. It does not say whether these diseases deteriorate each other, providing no guideline whether the patients should be treated by the ARIA approach ⁶ or by the DSM-5 protocol ⁸ first or simultaneously by both approaches.

It is known that low level of vitamin D is associated with the increased frequency of asthma exacerbations, increased allergy markers and asthma severity in children ¹², and that vitamin D₃ supplementation may modulate respiratory mucosa. Vitamin D has a role in immunological modulation, aging, gene regulation, brain homeostasis, and neurodevelopment. But, at this moment we do focus on just the condition of tic related conversion associated with allergic rhinitis.

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For the doctors in the primary health care, as well as for other doctors, it is important to have the correct initial guidelines for the treatment of a certain disorder in teenagers, which affects the cost of treatment.

Conclusion

This case report emphasizes the necessity of revising the systematization of tic disorders, the DSM-5 protocol and the ARIA approach in terms of their association, emphasizing each other, and clearer diagnostic/treatment/prognostic approaches of these associated disorders with the age of teenagers.

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