A Rare Case of Allergic Fungal Rhinosinusitis in Indonesian Elderly: A case Rport and Diagnostic Procedure

by Budi Sutikno

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Case Report

A rare case of allergic fungal rhinosinusitis in Indonesian elderly: A case report and diagnostic procedure

Drean Ferrys Widhiono, Budi Sutikno *

Department of Otorhinolaryngology Head and Neck Surgery, Faculty of Medicine, Universitas Airlangga – Dr. Soetomo General Academic Hospital, Surabaya, Indonesia

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ABSTRACT

Background: Allergic fungal rhinosinusitis (AFRS) is a rare case.

Case presentation: A 63-year-old-man presented right nasal congestion one year ago. There was a polyp in the right nasal cavity supported CTScan showed a solid mass with central hyperattenuating of ±8.4 × 2.4 × 4.4 cm. Total IgE value was 1,227 IU/ml, while Aspergillus specific IgE and Mucorous specific IgE using the micro-Elisa technique were negative or less than 0.35 IU/ml. The skin prick test was positive on exposure to house dust, cotton, chicken meat, and cow's milk. Mucosal polypoid and allergic mucin were found during functional endoscopic sinus surgery (FESS). Histopathology showed inflammatory cells of eosinophils.

Discussion: These results lead to a diagnosis of AFRS according to the Bent and Kuhn criteria. The highest incidence rate is in adolescents and young adults but it occurs in the elderly. So, some of the signs and symptoms of AFRS in adolescents and young adults do not appear.

Conclusion: AFRS can only be diagnosed during FESS when mucins are found, this case appear in the elderly to be very interesting.

1. Introduction

Allergic fungal rhinosinusitis (AFRS) is a non-invasive fungal disease of the paranasal sinuses, in which sinus inflammation occurs as an allergic reaction to fungal aerosols [1]. AFRS is found in 5–10% of CRS cases [2]. The number of AFRS cases is estimated to be 1–2% of the world population, with different incidence between regions [3]. The incidence of ARFS is influenced by geographic factors. Some literature reported that AFRS is found in temperate regions with high relative humidity [4]. The highest incidence rate is in adolescents and young adults with an average age of 21.9 years [5]. The diagnosis of AFRS can only be made during functional endoscopic sinus surgery (FESS) [6]. In addition, it is estimated that as many as 13% of AFRS were negative in fungal cultures [7]. Based on the Case Report Surgery (SCARE) 2020 guideline [8]. We were interested to report a case of a 63-year-old Indonesian man with a diagnosis of AFRS during FESS and negative in fungal culture.

2. Presentation of case

A 63-year-old Indonesian man presented a right nasal congestion one year ago. Right nasal congestion was persistent and worsened in the last three months. Nasal discharge was clear and watery. The patient's olfactory was impaired. Pain and nosebleed were not found. The patient had a history of allergy to dust and smoking for twenty years. Anterior rhinoscopy revealed mass filling the entire right nasal cavity, with smooth surface, reddish-white, and looked not easily to bleed (Fig. 1). Histopathology of the biopsy showed a piece of polypoid-shaped tissue, covered with squamous epithelium, and partly covered with respiratory epithelial cells. Fibrous connective tissue stroma with infiltration of inflammatory cells of eosinophils, neutrophils, lymphocytes, histiocytes. Dilated blood vessels were visible. There were no signs of malignancy. The conclusion was allergic polyp (Fig. 2).

Computerized tomography scan (CT Scan) showed a solid mass (35 HU) with central hyperattenuation (63–71 HU) indistinct borders, irregular edges, sized ±8.4 × 2.4 × 4.4 cm in the right nasal cavity with contrast enhancement (56 HU; Fig. 3). Immunology examination of total immunoglobulin E (IgE) using the Electro-chemiluminescence

* Corresponding author. Department of Otorhinolaryngology Head and Neck Surgery, Faculty of Medicine, Universitas Airlangga – Dr. Soetomo General Academic Hospital, Jalan Mayjend Dr. Moestopo No. 6-8, Airlangga, Gubeng, Surabaya, East Java, 60286, Indonesia.
E-mail address: budisutikno104@gmail.com (B. Sutikno).

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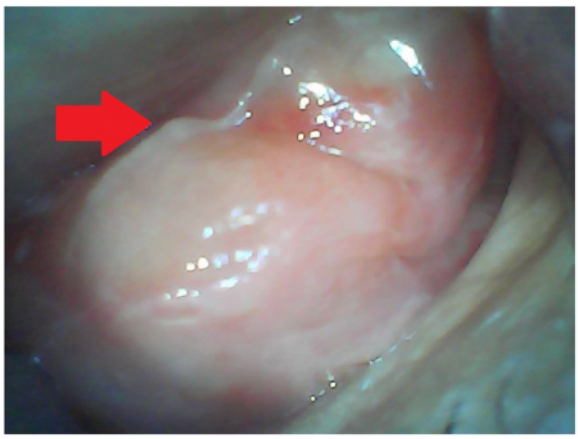

Fig. 1. The mass fills the right nasal cavity (arrows).

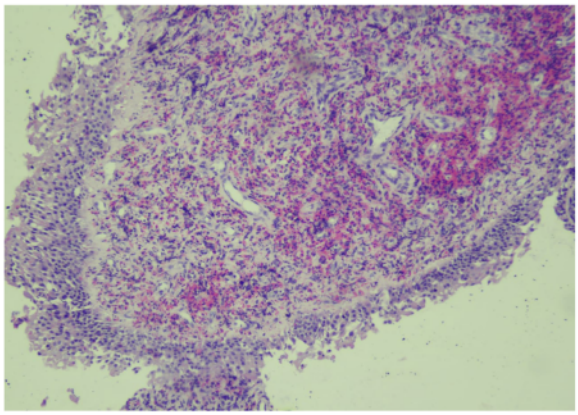

Fig. 2. Histopathology of biopsy shows infiltration of inflammatory cells of eosinophils, neutrophils, lymphocytes, and histiocytes.

Immunoassay (ECLIA) method obtained a value of 1,227 IU/ml (normal value < 100 IU/ml), while Aspergillus specific IgE and Mucorous specific IgE using the micro-Elisa technique were negative or less than 0.35 IU/ml. Skin prick test (SPT) obtained positive results on exposure to house dust, cotton, chicken meat, and cow's milk. Peripheral blood examination after administration of prednisone showed blood eosinophils of 2.9% with a normal reference value of 0.6–5.4%.

Before FESS, prednisone tablet 20 mg was given daily for one week, followed by 10 mg daily on following one week. FESS was performed by removing the intranasal polyp, followed by unsinectomy, middle meatal antrostomy, total etmoidectomy, and frontosinusotomy on the right nasal cavity. Mucosal polypoid and allergic mucin were found during FESS (Fig. 4).

Histopathology of specimen showed a piece of polyp-shaped tissue covered with respiratory epithelial cells, filled infiltration of inflammatory cells, lymphocytes, histiocytes, plasma cells, eosinophils, and a little spread of neutrophils. Some glands containing mucins were dilated. There was no sign of malignancy. The conclusion was allergic polyp (Fig. 5). Microbiology examination showed no fungal formation through KOH and there was no fungal growth in the culture.

3. Discussion

AFRS commonly occurs in young adults or adolescents with immunocompetent atopic, and rarely in children, but the disease has been found at any age. An initial diagnosis criteria that is still widely accepted was formulated by Bent and Kuhn in 1994. These criteria include type I hypersensitivity, nasal polyps, typical CT Scan findings, and eosinophilic mucin containing fungal without invading the mucous membrane. Then, minor criteria such as asthma, Charcot Leyden crystals, eosinophilia, unilateral, fungal culture, and bone erosion are added [9]. Patients must meet all major criteria for diagnosis, whereas minor criteria serve to support the diagnosis and describe the individual patient, but are not used to made the diagnosis [6].

The presentation of AFRS may range from subtle to dramatic. Indolent symptoms, such as painless and gradual nasal obstruction, anosmia, and the production of mucin, may progress for years. Nasal congestion, rhinorrhea, and post nasal drip are some of the most common symptoms and can present gradually. The expansile changes of the paranasal sinuses can result in either diplopia, due to proptosis, loss of visual acuity or visual field defects, due to optic canal encroachment [10]. Complaints of pain may indicate a concomitant bacterial infection. Facial dysmorphia in AFRS can be proptosis, telecanthus, and malar flattening, especially in younger patients [11].

AFRS patients have a wide sensitivity to a number of fungal and non-fungal antigens. Several studies showed positive SPT and in vitro response to fungal and non-fungal antigens in AFRS patients. Most of the AFRS patients have positive SPT for aeroallergens that are specific for IgE [5]. Total IgE levels are elevated, often reaching more than 1,000 IU/ml [6]. Nearly 90% of AFRS patients show evidence of type I hypersensitivity with elevated serum IgE levels and type III hypersensitivity with eosinophilia of the sinus mucosa with fungal antigens [12]. Fungal-specific IgE and IgG can be found in non-allergic and AFRS. The level of specific IgE for fungal is not significantly different in patients with AFRS [12]. Count blood cell show an increase in the number of eosinophils [13]. Several randomized clinical trials have showed that oral corticosteroids improve olfactory function, endoscopic scores, reduce polyps, and decrease blood eosinophilia, IgE, and IL-5 [14].

Imaging characteristics are an important component of the diagnosis of AFRS. CT Scan often show unilateral or asymmetrical involvement of the sinuses [6]. CT Scan show multiple opacities of the sinuses with central hyperattenuating. Allergic mucin causes a heterogeneous intensity characteristic of AFRS, although it is not specific for AFRS. This heterogeneity was initially thought to be related to the accumulation of hemosiderin in mucins, but a recent theory suggested that the heterogeneity is caused by deposition of heavy metals such as iron and manganese. Allergic mucin is a characteristic feature of AFRS [11]. This mucin consists of lamination of necrotic eosinophil skin with various stages of degeneration, sometimes small hexagonal crystals of Lysophospholipase (Charcot Leyden crystals) and a few fungal hyphae [15]. FESS is usually indicated to remove hypertrophic mucosa and allergic mucin. Allergic mucin looks thick, very sticky, and has a variety of colors, so that it is described as similar to peanut butter or axle grease [16].

Fungal are rare in AFRS, and even when present, they are often difficult to detect [17]. Fungal growth in culture media does not necessarily indicate AFRS, because fungi are present in a free environment, and can give false-positive results. Negative cultures do not rule out AFRS and positive cultures can represent environmental contamination. The culture results only act as supporting evidence for AFRS [9]. 13% of AFRS show negative fungal cultures despite histopathology confirming AFRS [7].

The histopathology findings in AFRS are very important for diagnosis. Histopathology of specimen on hematoxylin eosin staining will show a typical inflammatory infiltrate consisting of eosinophils, lymphocytes, and plasma cells. The mucosa will be hypertrophic and hyperplastic, but there are no signs of necrosis, giant cells, granulomas, or

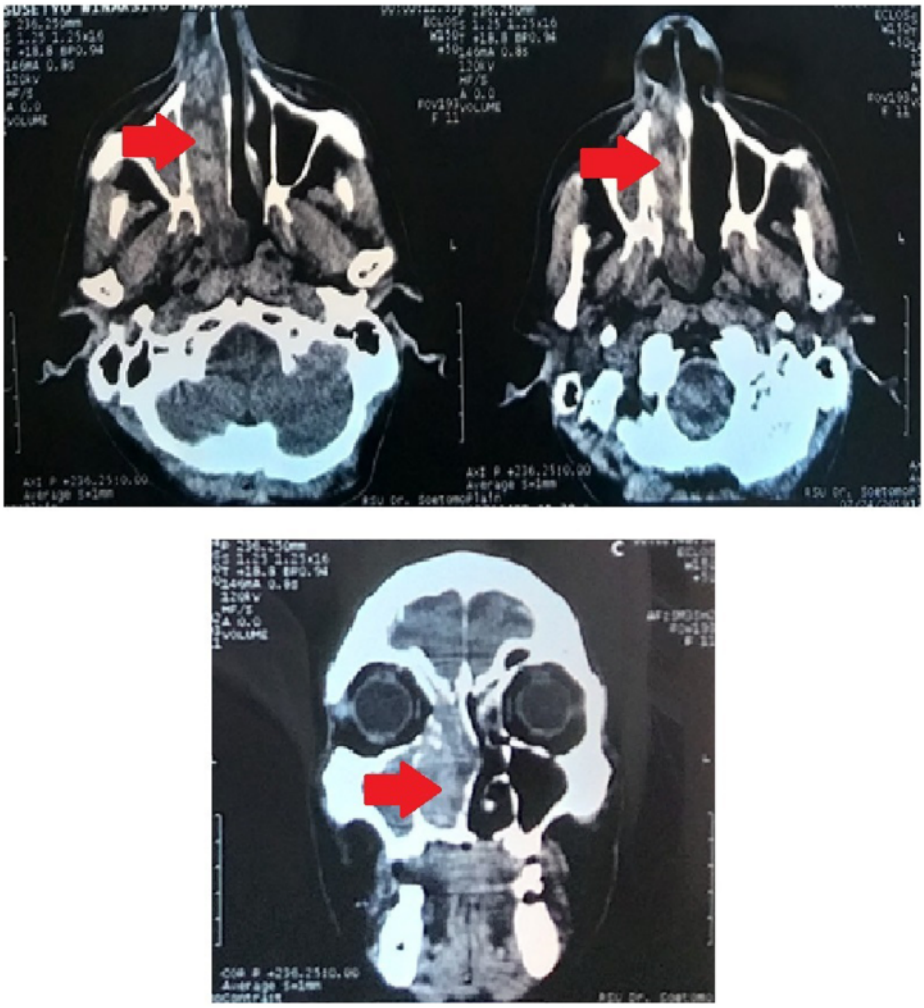

Fig. 3. CT Scan of the paranasal sinus axial view. Central hyperattenuating appears on the paranasal sinus and right nasal cavity (arrows).

invasion of surrounding structures. This latest finding will provide support for the diagnosis of fungal processes other than AFRS [6]. In this case, AFRS was found at stage III [9].

Base on the report found that the case of AFRS highest incidence rate is in adolescents and young adults but it occurs in the elderly. So, some of the signs and symptoms of AFRS in adolescents and young adults do not appear. The fungal culture results in mucins and tissue are negative, which is also a rare case.

4. Conclusion

Diagnosis of AFRS is based on major criteria proposed by Bent and Kuhn, whereas minor criteria serve to support the diagnosis and describe the individual patient, but they are not used to establish the diagnosis. Most cases of AFRS are diagnosed in young people and elderly people are rare.

Ethical approval

We have conducted an ethical approval base on the Declaration of Helsinki at Ethical Committee in Dr. Soetomo General Academic Hospital, Surabaya, Indonesia.

Sources of funding

None.

Author contribution

Drean Ferrys Widhiono: drafting, visualization, acquisition of data, revising; Budi Sutikno: drafting, revising, reviewing, conception and design.

Consent

Written informed consent was obtained from the patient.

Registration of research study

Not applicable.

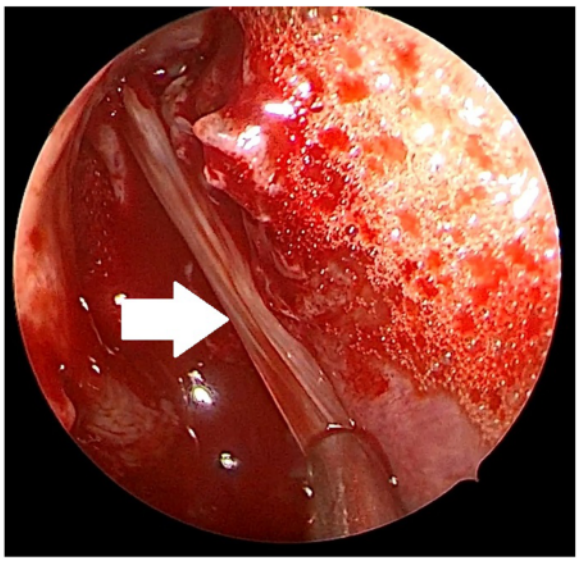

Fig. 4. Right nasal cavity. There are nasal polyps and allergic mucin (arrows) found at FESS.

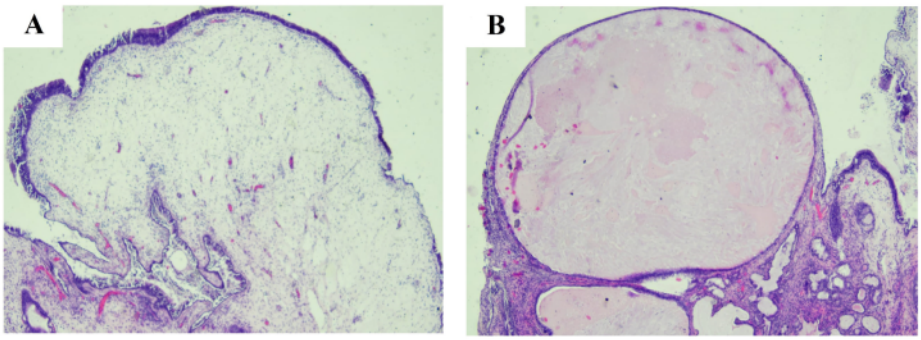

Fig. 5. Histopathology of the specimen shows infiltration of inflammatory cells of lymphocytes, histiocytes, plasma cells, eosinophils, neutrophils, and the glands containing mucins are dilated (A and B).

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Declaration of competing interest

The authors declare that they have no conflict of interest.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102400.

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