CASE REPORT
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A rare nasal myiasis in a patient with diabetes mellitus

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ABSTRACT

Background: Nasal myiasis, a type of cavitary myiasis, is an infestation of parasitic insect larvae or eggs in the nasal cavity of the human body. The risk of nasal myiasis increases in individuals with immunocompromised such as diabetes mellitus (DM). The aim of this study was to report a rare case of nasal myiasis in a patient with DM.

Case Presentation: A 54-year-old female patient was referred to Dr. Soetomo General Hospital Surabaya with a chief complaint of spontaneous anterior epistaxis from the left nostril for three days. Anterior rhinoscopy revealed a narrowed left nasal cavity covered with excessive bloody discharge and>50 larvae coming out of the nasal passage. Nasoendoscopy showed ulcerated lesions on the left nasal cavity floor and nasal lateral wall, destructions of the osteomeatal complex and lateral wall, thick bloody discharge with excessive crust, as well as a larva around the osteomeatal complex entering the maxillary sinus. Paranasal sinus CT scan showed no further infection to surrounding organs. The patient had a history of type 2 DM for the past 2.5 years. The patient was diagnosed with nasal myiasis, unregulated type 2 DM, and diabetic neuropathy. Therapies included larval removal by endoscopy, wound irrigation using antiseptic solutions, maxillary sinus irrigation, administration of the topical antiparasitic ivermectin, and insulin therapy. After seven days of the treatment, the patient was discharged from the hospital with a resolved infection and controlled blood sugar levels.

Conclusion: Awareness of risk factors for myiasis along with proper therapy is prominent. Nasal endoscopy, together with nasal irrigation and antiparasitic drug administration was effective in the management of nasal myiasis.

Keywords: *Cochliomyia* sp., diabetes mellitus, diabetic neuropathy, nasal cavity, nasal myiasis.
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INTRODUCTION

Myiasis, first introduced by F.W. Hope in 1840, is the infestation of human or animal tissue or cavities by the eggs or larvae of parasitic flies.[1] The disease distribution occurs worldwide, with the greatest abundance in tropical and subtropical areas, where the climate is suitable for flies breeding. Myiasis is commonly reported in individuals >50 years of age.[2,3] Obligate myiasis denotes larvae necessarily feeding on living tissue, while facultative myiasis represents maggots living freely or parasitic.[4] Infections may occur either on the skin (cutaneous myiasis), body cavity (cavitary myiasis), or lesions (wound myiasis).[5] Open and traumatic wounds, scabs, suppurative lesions, and infected ulcers are among the risk factors for myiasis.[6]

Cavitary myiasis, including ears, nose, and throat (ENT), is basically the least common type of myiasis. However, among the ENT systems, nasal myiasis is the most common one, accounting for approximately 75% of all ENT myiasis cases. Several factors such as a wide space in the nasal cavity that provides easy access to flies and a less sensitive nasal mucosa have been associated with a higher incidence of nasal myiasis compared to other types of myiasis.[4]

The risk of nasal myiasis increases in individuals with immunocompromised diseases such as diabetes mellitus (DM), in particular.[4,7] DM is defined as a hyperglycemic syndrome resulting from defects in beta cell insulin secretion, tissue insulin resistance, or both. It has been considered a global health problem,[8] affecting approximately 387 million people in 2013 and will be doubled by 2035 according to the International Diabetes Federation (IDF) estimation. Among several types of DM (type 1 DM, type 2 DM, gestational DM, and specific DM), type 2 DM is the most dominant, accounting for approximately 90% of DM cases worldwide and 97.5% of DM cases in Indonesia.[9,10] DM also often lead to various serious health complications, resulting in an increased risk of morbidity and mortality.[10] Hence, multiple studies have been conducted to suggest some effective methods for early DM screening, possible risk factors for diabetes and factors affecting DM, as well as information regarding patients' management and cares to reduce DM cases or further diabetic complications.[8,11-15] However, since DM is an acquired immunodeficiency condition, individuals with DM are also susceptible to various types of infections,[16,17] including myiasis. In this article, we report a case of rare nasal myiasis in a patient with diabetes mellitus.

CASE PRESENTATION

A 54-year-old female patient, a farmer, was referred to Dr. Soetomo General Hospital Surabaya with a chief complaint

of spontaneous anterior nosebleed from the left nostril for 3 days. Additional complaints were intermittent dull pain in the nose, cheeks, upper gums, around the orbits, and forehead since the last week. At the same time, nasal congestion along with decreased sense of smell were also reported. Posterior nosebleed was not detected and there was no history of trauma to the nose. The bleeding volume was quite large but decreased after epinephrine tampons insertion. During the tampon change, larvae were found along with the blood. There was no bleeding in other parts of the body or history of previous recurrent bleeding. There was no history of foreign bodies or complaint of lump in the nose. Chronic colds, weight loss, and medicine consumption were denied. The patient's blood pressure was normal during the treatment at previous hospital. The patient did not have fever during nosebleed. The patient complained of bad breath for the last 3 months.

The patient had a history of DM for the past 2.5 years, for which she received metformin therapy 500 mg 3 times/day and glimepiride 2 mg once/day before breakfast. After a month of treatment, the patient felt better and obtained a normal blood glucose level, thus she stopped glycemic control and medication. However, the patient legs often felt heavy, especially during the night. There was no complaint of burning sensation or tingling pain, leg pain when walking, visual disturbances, or skin itchiness. History of hypertension and smoking were denied.

Physical examination revealed a fair general condition. Vital signs showed blood pressure of 120/70 mmHg, heart rate of 96 x/minute, respiratory rate of 20x/minute, temperature of 36.7°C, oxygen saturation of 98% (without oxygen supplementation), and Wong Baker scale of 5/10. The head and neck examination exhibited non-anemic conjunctiva, anicteric sclera, no cyanosis, and no dyspnea. The jugular venous pressure (JPV) and lymph node were normal. Chest examination indicated symmetrical movements. The right midclavicular line found the lung-hepatic border in the 6th intercostal space. A regular single S1 and S2 were detected. No murmur, gallops, or pericardial friction rub was heard.

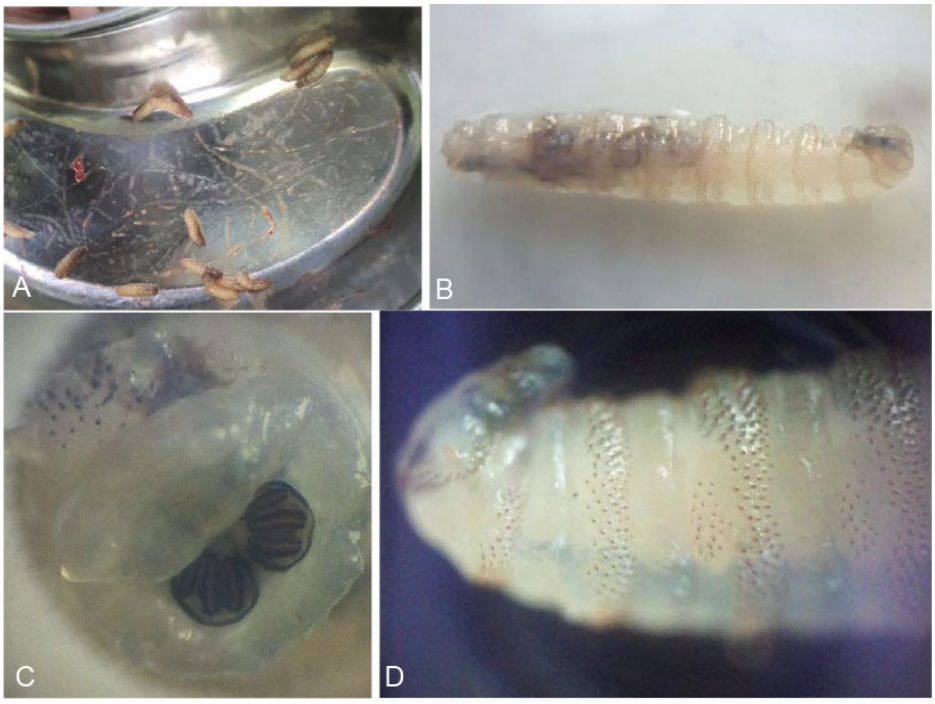

Figure 1. (A) The larvae were manually removed by forceps; (B-D) The larva was identified as *Cochliomyia sp.*

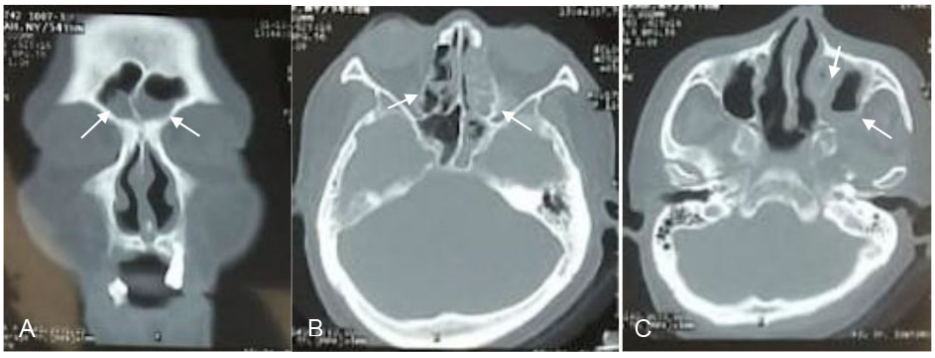

Figure 2. Mucosal thickening on the (A) right and left frontal sinuses; (B) ethmoidal and sphenoidal sinuses; and (C) right and left maxillary sinuses (dominant of the left side).

The results of lung examination showed vesicular breathing over both of the hemithorax. Rhonchi and wheezing were not detected. Abdominal examination indicated flat abdomen, normal bowel sounds, epigastric tenderness, and non-palpable liver and spleen.

Examination of the extremities revealed no deformity, muscle atrophy, or ulcers on both legs. Both legs had normal skin color and warm. The dorsalis pedis artery pulse was normal and symmetrical. No hair loss or anhidrosis was found on both legs. The results of ankle-brachial index examination were within normal limit. The Achilles tendon reflex was +1/+1. Examination of the compressive sensation with a 10 g Semmes-Weinstein monofilament indicated a loss of touch-pressure sensation at the 1st metatarsal head of the right big toe and the distal area of the plantar plate of the right and left big toes. Vibration test with a 128-Hz tuning fork revealed a loss of vibration sensation on the dorsal region of the great toes of both feet.

Otoscopy of both ears showed that the ears were covered with cerumen, and the tympanic membrane was difficult to evaluate. Rhinoscopy of the right nasal cavity was normal; however, left nasal rhinoscopy revealed narrowed nasal

cavity covered with excessive blood and discharge along with 50 larvae coming out of the nasal passage (Figure 1A). After nasal irrigation, nasal concha medius was found oedematous and there were numerous small ulcers on the nasal mucosa partially covered with crusts. The phenomenon of the soft palate (palatum molle) was negative. Non-erythematous tonsil T1-T1 in size (without detritus) and non-hyperemic pharynx were observed.

Based on all the anamnesis, medical history, physical evaluation, and adjuvant examination, the patient was diagnosed with nasal myiasis sinistra, epistaxis, unregulated type 2 DM (random blood glucose of 431 mg/dL), and diabetic neuropathy. The patient was treated with rapid-acting insulin aspart 3x4 units/hour intravenous, resulting in a decreased blood glucose level (260 mg/dL). Insulin aspart injection (4 units) was then repeated once and the blood glucose further declined to 198 mg/dL. Other therapies included nasal tampon insertion, NaCl 0.9% infusion 1000 cc every 24 h, B1 diet 1900 kcal/daily, subcutaneous insulin detemir injection 10 units every 24 h at bedtime, ranitidine 50 mg every 12 h and metamizole 1 g every 8 h intravenous, vitamin B12 1 tablet per oral every 8 h, and nasal irrigation with ivermectin 1% and povidone-iodine 1% 3 times/day.

On the second day of treatment, less nosebleed and nasal pain (Wong Baker scale of 3/10) were reported. A larva was obtained through nasal irrigation. The patient was in a fair general condition with stable vital signs. However, laboratory examination revealed high HbA1c (12.3%) and blood glucose levels (231 mg/dL before breakfast, 242 mg/dL before lunch, 228 mg/dL before dinner, and 230 mg/dL at bedtime). Thus, regimen insulin intensification therapy with insulin aspart (4 units) every pre-meal (breakfast, lunch, and dinner) and an increased dose of insulin detemir (14 units) was given.

Nosebleed and larva were no longer observed on the 3rd day of treatment. Wong Baker scale value stayed the same (3/10). Nasal endoscopy was performed on the left nasal cavity and revealed some ulcerated lesions on the nasal floor and lateral wall. There were also destructions of osteomeatal complex and lateral wall, thick bloody discharge, and a larva around the osteomeatal complex entering the maxillary sinus. The patient then underwent inferior concha meatotomy and left maxillary sinus irrigation, where multiple larvae came out along with irrigation solution. Blood sugar level decreased to the range of 171-180 mg/dL after correction and an increased dose of insulin detemir (16 units) and insulin aspart (6 units) were prescribed.

On the 4th of therapy, the patient's condition had improved. Wong Baker scale was 2/10. However, blood sugar levels were still high (ranged from 142-154 mg/dL), and thereby injections of a higher dose of insulin detemir (18 units) and insulin aspart (8 units) were carried out. On the next day, blood sugar values decreased by more than 20 points at each time of assessment (before breakfast, lunch, dinner, and bedtime) and the same insulin therapy was continued. On parasitological examination, the larva was identified as *Cochliomyia sp.* which belongs to the family of *Calliphoridae* (Figure 1B-D).

During the 6th day of treatment, thick discharge with excessive crusts, lateral wall and osteomeatal (ethmoid bulla) destructions, and mucosal re-epithelization were observed in the left nasal cavity during nasal endoscopy evaluation. No larva was found during extraction process. Blood glucose levels declined to the range of 120-127 mg/dL.

Paranasal sinus CT scan was carried out on the 7th day of treatment to evaluate any possible infection due to defects in the lateral wall of the nasal cavity. The result revealed mucosal thickening (20-41 HU) on the right and left frontal sinuses (dominant on the left side), right and left ethmoidal and sphenoidal sinuses, as well as right and left maxillary sinuses (dominant on the left side) (Figure 2A-C). Blood glucose value decreased to the range of 114-120 mg/dL and Wong Baker scale of 1/10.

On the following day, the patient was discharge and underwent outpatient treatment with insulin aspart 8 units every pre-meal (breakfast, lunch, dinner) subcutaneous, insulin detemir 18 units before bedtime subcutaneous, paracetamol 500 mg/oral whenever pain occurred, vitamin B12 1 tablet/oral every 8 h, and nasal irrigation with ivermectin 1% and povidone-iodine in the otolaryngology unit.

DISCUSSION

Nasal myiasis, the most common type of cavitary myiasis in the ENT system, is recognized as an infestation of parasitic insects' larvae or eggs in the nasal cavity of the human or animal body. This disease affects individuals of all ages, especially adults and geriatrics with no gender predisposition. Several predisposing factors such as low socioeconomic status, nutritional deficiency, poor personal hygiene, patients with disabilities, malignancy, basic sinonasal diseases such as atrophic rhinitis (ozaena), immunocompromised state such as diabetes mellitus, and conditions that reduce protective reflexes for sneezing and sensory sensations of the nasal mucosa such as leprosy and diabetic neuropathy have been associated with nasal myiasis.[4,6,7,18]

In the present case, the history of the patient's occupation as a farmer has been associated with an increased risk of myiasis. Farmers working in rural areas often have close contact with livestock, thereby having a higher chance of suffering from a larval infestation.[19] In addition, having a low socioeconomic status and poor personal hygiene might be other predisposing factors associated with larval infection in the patient. In addition, the patient also had a history of uncontrolled type 2 diabetes mellitus for more than two years leading to immunocompromised condition and diabetic neuropathy. Neuropathy has indeed been reported as the most common diabetic complication among type 2 DM patients in Indonesia.[10] Poor glycemic control may impair the function of the innate and adaptive immune system, while diabetic neuropathy, which was confirmed by abnormal results of the pressure sensation and vibration tests on both of the patient's legs, has caused reduced protective reflexes in the patient's nasal mucosa and thereby facilitates the infestation of flies into the nose.[20] Diabetic neuropathy also enables easier larval infestation and habitation in the nasal cavity since the patient

was not alert of the first larval attack. This combination of immune system and defects sensory neuropathy due to hyperglycemia might have been the factor causing myiasis in this patient. However, chronic complications of vasculopathy as well as basic sinonasal disease, which may attract parasitic infestation through the production of odor, were not found in the patient.

Infection in diabetic individuals may occur due to several factors including impaired immune response owing to hyperglycemia, vascular insufficiency (vasculopathy), peripheral neuropathy, and increased colonization of commensal microbes on the skin and mucosa.[10,21] Vasculopathy leads to a decreased blood flow, impairing oxygenation of peripheral tissue and reducing the transport of antibiotics to the peripheral areas, while neuropathy may facilitate lesions development from local trauma by providing entry to parasites.[22] The patient suffering from diabetic neuropathy is often unaware of lesion formation until clinical signs of infection become apparent. Diabetic neuropathy found in the current patient has also been observed in >50% of patients with type 2 DM.[16]

In nasal myiasis, the infection begins when female flies lay their eggs on the nasal mucosa or when a person touches the nasal mucosa area where the eggs are attached. The eggs hatch in a moist and warm mucosal environment, and turn to larvae within 7-9 hours in tropical areas. The larvae then grow in a region between the nasal mucosa and periosteum by obtaining nutrients from the surrounding tissue with the help of proteolytic enzymes found in their mouth and feces.[4,18] *Cochliomyia hominivorax*, *Chyrsomya bezziana*, *Wohlfahrtia magnifica*, *Oestrus ovis*, *Lucilia sericata*, and *Drosophila melanogaster* are among the species that commonly cause nasal myiasis.[4] In the current case, the larva was identified as *Cochliomyia* sp through parasitology examination (Figure 1B-D).

Patients with nasal myiasis commonly present with multiple symptoms, including epistaxis, nasal obstruction, nasal or facial pain, headache, foreign body sensation in the nose with or without crawling sensation, nasal cavity odor, and impaired sense of smell. These symptoms appeared as a result of larval invasion and movement in the mucosa. Invasion of larvae causes edema in the nasal mucosa, causing nasal congestion and the formation of thick and smelly nasal discharge. The dried discharge will form crusts on the mucosal surface. The larval invasion also causes bleeding, pain, and destruction of olfactory bulbs resulting in olfactory disturbances. About 20% of the patients also experience a larva spontaneously crawling out of the nose.[1,4,23] Almost all the aforementioned symptoms of nasal myiasis were observed in the patient in this case.

Several methods have been developed for the diagnosis and treatment of nasal myiasis. Anterior rhinoscopy has been considered a simple diagnosis method of nasal myiasis. During rhinoscopy, a number of larvae can be seen in the nasal cavity with edema, ulceration, and crusting on the nasal mucosa. Anterior rhinoscopy is a diagnostic and therapeutic tool since it helps manually remove the larvae using forceps. However, since larvae are generally photophobic, this method sometimes possesses limited capability of finding and removing the larvae hiding in the deepest areas of the dark nasal cavity. Nasal endoscopy, on the other hand, provides deeper access and a more detailed picture of the nasal cavity than rhinoscopy, enabling larvae removal occupying hard-to-reach locations in the nasal cavity. In addition, a CT scan is recommended once clinical signs or nasal endoscopy findings suggest a possible involvement of organs around the nose (paranasal sinuses, orbits, and brain).[5] In this case, paranasal sinus CT scan was performed as nasoendoscopy revealed destruction in the nasal cavity lateral wall and osteomeatal complex. Damage to these structures may raise suspicion of possible infection of the paranasal sinuses since these structures are directly adjacent to this part of the body.

Treatment of nasal myiasis includes larval removal, wound care, and therapy with antiparasitic drugs. Nasoendoscopy procedure to remove larvae is more preferable compared to rhinoscopy. Complete larval expulsion is prominent since their fragments can trigger allergic reaction in the patient. Wound care is carried out periodically by washing the nose using an antiseptic solution. Antiparasitic drugs can be administered orally or topically, and ivermectin is the most widely used parasiticide. In various case reports, administration of ivermectin facilitated larval extraction and accelerated wound healing. Ivermectin showed a good safety profile with minor adverse effects. Other alternatives include thiabendazole and turpentine solution. Antibiotics are given only if clinical signs are indicative of secondary bacterial infection.[4,5]

In this case, larval removal was carried out through nasoendoscopy procedure twice daily, maxillary sinus irrigation, and nasal irrigation three times daily. There were no clinical signs of secondary bacterial infection in the patient, thus antibiotics were not required. Ivermectin solution 1% was given as topical antiparasitic. Blood sugar regulation was initiated by rapid insulin regulation and basal insulin regimen from the first day of treatment, which was then intensified to a basal-bolus insulin regimen on the second day of treatment. According to the standard guidelines for diabetes medical services from the American Diabetes Association (ADA) 2019, the recommended pre-prandial blood sugar level for individuals with diabetes is 80-130 mg/dL, which in the patient was achieved on the fifth day of treatment. The patient was discharged on the 8th day of therapy and underwent outpatient treatment with insulin aspart, insulin detemir, paracetamol 500 mg/oral (whenever pain occurred), vitamin B12, and nasal irrigation with ivermectin 1% and povidone-iodine in the otolaryngology unit.

CONCLUSION

We report a case of a 54-year-old woman with a chief complaint of epistaxis. The patient had a history of uncontrolled type-2 DM since the past 2.5 years. Based on the clinical and adjuvant examinations, the patient was diagnosed with nasal myiasis, unregulated type-2 DM, and diabetic neuropathy. The patient underwent various treatments including larval removal by endoscopy procedure, wound care by nasal irrigation using antiseptic solutions, maxillary sinus irrigation, administration of the topical antiparasitic ivermectin, and

insulin therapy to control blood sugar. CT scan revealed no further infection to surrounding organs. Parasitological investigation identified *Cochliomyia* sp. as the causative pathogen. After seven days of treatment, the patient was discharged with the resolved infection and corrected blood sugar levels.

PATIENT CONSENT

The patient signed informed consent form and had agreed the case to be published in scientific journal.

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DISCLOSURE OF CONFLICTS OF INTEREST

The authors declare no conflict of interest regarding the manuscript.

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AUTHOR CONTRIBUTION

Authors contributed significantly to the case-report.

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