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A Rare Human Cerebral Myiasis in HIV Patient: A Case Report

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Abstract— Human Immunodeficiency Virus (HIV) infection can be caused damage to the immune system and leading to immunocompromised conditions, making the person more likely to get other infections. Myiasis is a maggots infestation in the human body, in immunocompromised conditions, it can lead to severe conditions and cause death.Human cerebral myiasis in HIV patients is a rare condition. HIV Infection and type 2 Diabetes Mellitus (DM) play a major role in immune system dysregulation which leads to immunocompromise. Hereby, we report those case with its management.

Keywords: Cerebral Myiasis, HIV, Immunocompromise

Introduction

Human Immunodeficiency Virus (HIV) is a virus that attacks immune cells and leading to immunocompromised conditions by reduces the number of CD4+ cells, making the person more likely to get other infections. HIV consists of primary infections with or without the acute syndrome, asymptomatic stages, and advanced stages. [1] More than 70 million people have been infected by HIV worldwide and 35 million people have died from HIV. Globally, 36.9 million were living with HIV until the end of 2017. [2] In 2016, Indonesia had 48,000 new HIV infections and 38,000 deaths from AIDS. In 2016, 620,000 people were living with HIV and 13% of them had used anti-retroviral therapy (ART). [3]

Myiasis is a parasitic infestation of tissue caused by the *Diptera sp*. fly larvae. Myiasis is a common infection in the tropical area, which is a natural habitat for many species of flies. Poor hygiene and immunocompromised conditions are predisposing factors for myiasis in humans. Patients are often asymptomatic, but the infection may cause a serious problem if the larvae attack body organs. Cerebral myiasis is an interesting case because it is rare, literature study found eleven cases from 1939, one of them was HIV / Acquired Immunodeficiency Syndrome (AIDS). [4] The following article is a case report of cerebral myiasis in a patient with HIV from Indonesia.

Case Report

A 48 years-old Asian woman who has a history of vision loss and brain tumor surgery in 2011 came to the emergency ward with a chief complaint found maggots on the forehead wound. Initially, the wound formed from a pustule near the post-surgical site, it appeared 2 months ago. She has been getting wound treatment at the clinic before, she got simple debridement wound care and oral antibiotics but seems to no improvement. The wound is getting larger, and then the physician advised her to refer to the surgery outpatient clinic in the hospital. Because of the problem of national health insurance bills, her family refused to bring her to the hospital and do self-wound care at home. The wound is neglected, getting chronic and spread to the entire forehead until titanium mesh was exposed and found maggot manifestation. She felt painful and bloody sores. There are no complaints of fever, dizziness, decreased consciousness, seizures, or paralysis but she complained of weight loss of 7 kg in 6 months. Her husband died 4 months ago because of a pulmonary infection.

Figure 1. wound care process. a) during debridement; b) day 3. post debridement ; c) day 22. granulation of the wound

Physical examination showed the general condition was a weak, normal vital signs, normal Body Mass Index (BMI). A large wound was found in the frontal region 12x15 cm size, with erythema border and titanium mesh exposed, contaminated with maggots. There was no other neurological deficit. Laboratory studies found microcytic anemia with haemoglobin (10.8) , hypoalbuminemia (2,69), normal leucocyte 6.570, elevated C-reactive protein (104.91), and hyperglycemia with random blood sugar (RBS) 324 mg/dL. Anti-HIV positive found during work up pre-operative, with CD4+counts 131 cell/m3. The Chest x-ray is normal. Head Multi-Slice Computed Tomography (MSCT) with contrast showed the hyperdense area in dextra and sinistra of the frontal region, with hypodense area in the center of it, impressive gliosis, and tip ventricular shunt.

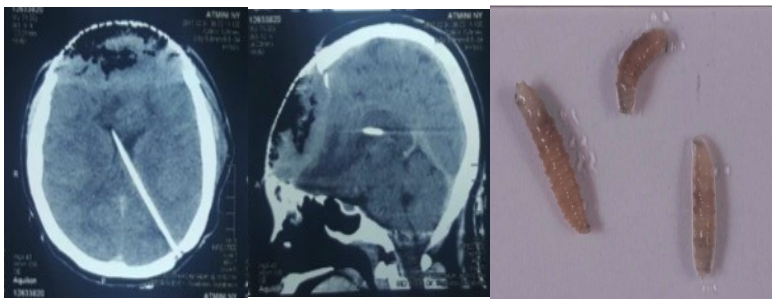

Figure 2. a) Head Multi-Slice Computed Tomography (MSCT) + contrast showed gliosis; b) parasitologic finding : three larva of *P. regina*

The patient was taken urgently to the operating room. Profilaxis antibiotic and rapid-acting insulin regulation is given until RBS ≤ 180 mg/dL. Surgical procedures performed include debridement and irrigation. Debridement was done manually, using irrigation and a gentle suction procedure. The patient was positioned in trendelenburg, wounds were cleaned using saline solution, then titanium mesh was removed. To disinfect the area, the operator used iodine while avoiding the brain cortex. Maggots were extracted manually and gently irrigated until it was ensured that all larvae had been lifted. Necrotomy was performed to remove the slough.

The wound was closed sterilely using absorbent gauze. To finish the procedure, the wound was treated aseptically and elective reconstruction (defect closure) was scheduled by a plastic surgeon. Anti-retroviral therapy was given on the day-2 of hospitalization. We used a fix dose combination qDay, contains Tenovofir 300 mg, Lamivudine 150 mg and Efavirenz 600 mg. Intraoperative cultures revealed environmental microbiotas (*Providencia stuartii*) and the parasitologic finding was *Phormia regina,* a common fly larva species causing wound myiasis.

The term myiasis was first described by Hope in 1840. Myiasis is defined as a parasitic infection by Diptera larvae that occurs in humans and animals. The larva lives inside the host by feeding on dead tissue, body fluids, and food that has been swallowed by the host. Clinically, myiasis is classified as primary and secondary based on host immunity, infestation, and anatomical location. [5]

Discussion

Primary, myiasis is a rare case in humans. Adult flies lay their eggs on the blood-feeding insect as vectors. While vectors damage host tissues and suck the blood, temperature around the vector stomach warms up, this causes the diptera eggs to hatch and fall to the host tissue surface, penetrating through vector puncture holes and developing into end-stage larvae. In secondary myiasis,

female flies lay eggs in open wounds, larvae penetrate tissues and feed on necrotic tissue. Other types of myiasis based on anatomical location include furuncular myiasis which is often caused by *Dermatobia hominis* (human botfly); Wound myiasis by *Cochliomyia hominivorax* larvae (America) and *Chrysomyia bezziana* (Africa, Australia, Asia); Cutaneous myiasis by *Hypoderma bovis* larvae (infested cattle) and *Gasterophilus intestinalis* (infested horses); posttraumatic myiasis; nasal myiasis and ophthalmomyiasis by *Oestrus ovis* (sheep botfly). The severity of myiasis depends on the location of infestation and tissue inflammatory response. [6]

This patient has neglected wound myiasis. It occurs because larvae have infested in open wounds**.** There are no signs of neurological deficits, sepsis, meningitis, or encephalitis, even though the patient has terrible open wounds with exposure to cerebral tissue for a period of time. Although the results of tissue culture show bacterial growth, the patient shows no symptoms of systemic infection. This result may closely related to the role of larvae in the wound healing process. It has been known since centuries ago, during the civil war in America until the discovery of antibiotics, that maggots are beneficial for wound healing. Currently, the Food and Drug Administration (FDA) has legalized 'medical maggots' as biotherapy in chronic wounds. The larvae only feed on dead tissue, this is the underlying mechanism for its function as biodebridement, while it also releases various kinds of enzymes that have antimicrobial potency, thus it may prevent tissue inflammation and bacterial invasion. [7]

Human cerebral myiasis is an exceedingly rare condition. The location of this parasitic infection appears to be more common in the frontal lobe as seen in eight of eleven cases. The first case was reported by Froomkin and Kasznelson in 1939, since then, few cases have been reported in the world. Reports on species found include *Hypoderma bovis* in three cases, *Phaeniera sericata* in one case, *Hypoderma lineatum* in one case, unidentified *Hypoderma sp. in one case, Dermatobia hominis* in one case, *Musca domestica* in one case, and unidentified species in two cases. [6] Most of the cases were found in Europe, two in the United States, and one in India. In this case report, we identify *Phormia Regina a*s the parasite.

The predilection of cerebral myiasis manifestations is in the frontal lobe, mainly the largest lobes and often involved in head injury. This patient's Computed Tomography (CT) scan shows larvae infestation in the frontal area. Most cases (85-90%) are caused by *Calliphoridae* larvae, which are classified as facultative obligate parasites. [7] Myiasis is usually found in developed countries and tropical or subtropical regions. Geriatric, poor hygiene, low socioeconomic status, immunocompromised conditions such as diabetes, cancer, maybe a predisposing factor to myiasis infection in humans. [8] This patient has several risk factors of having larvae parasitic infections, including immunodeficiency due to diabetes and HIV, low socio-economic status, and poor hygiene. Symptoms that most reported are pain, irritation, discomfort, alopecia, and pruritus. The surrounding tissue appears darker, wetter, and foul-smelling due to direct tissue damage, bleeding, and secondary infections. The diagnosis is made through the identification of larvae.

Since 2013, studies have recommended earlier management of HIV using antiretroviral due to their significant impact on decreasing mortality, morbidity, and transmission. Scientific evidence through cohort analysis and systematic review states that the difficulties of HIV management may be related to the development of comorbidities such as cardiovascular disease, kidney disorders, liver function disorders, malignancies, and neurocognitive disorders. Early administration of antiretroviral may reduce the risk of co-infection and increase patient safety. A recent study from randomized control trial revealed that ARV therapy reduces the number of infections in groups of homosexual couples with no infection of HIV.

The principle of myiasis management is larvae removal, either manually or surgically. [9] Other literature states that it is necessary to perform debridement and irrigation to remove larvae from the wound. The administration of chloroform, which may be the chloroform in vegetable oils or others, combined by removal of larvae under local anesthesia is also recommended as an option for wound myiasis treatment. These are some of myiasis management approach including occlusion/suffocation (petroleum jelly, liquid paraffin, beeswax, or heavy oil); surgical removal with local anesthesia (anesthetized with lidocaine and excised surgically); systemic/topical ivermectin (oral and orbital myiasis); wound myiasis (debridement with irrigation). [10]

In the perioperative period, administration of ART must also be continued. Recommendations for starting ART regardless of CD4+ count are based on a systematic review with Grading of

Recommendations, Assessment, Development, and Evaluation (GRADE), this systematic review assesses evidence in 17 observational studies that report immunological, virology, and HIV transmission outcomes in patients receiving ART treatment earlier. According to AIDS clinical guideline 2012, antiretroviral should be given during the perioperative period in doubt the clinical staging and CD4 + counts. In 2015, WHO issued ARV Guideline, that ARV started to be given to all adult HIV patients, regardless of clinical stage and level CD4+ and ARV should be given to all adult HIV patients at clinical stage 3 or 4 and CD4+ 350 cells / mm3. [2]

Basically, perioperative procedures in patients with HIV/AIDS are similar as other patients. However, a complete assessment of the patient's condition is needed. Variable to evaluate include renal and hepatic dysfunction, coronary heart disease, coagulopathy and homeostasis disorders, alcohol users status, drugs, history of Methicillin-resistent *Staphylococcus aureus* (MRSA) infections, and drug allergies.Postoperative evaluation in HIV patients is slightly different from non-HIV patients. HIV patients have a higher risk of hypercoagulation and may be at risk of postoperative thromboembolic complications. Early management of prophylaxis and mobilization is important in this group of patients. Assessment of pain scale also needs to be monitored closely about the history of narcotics, which may reduce pain assessment. [3]

Conclusion

The management of myiasis infection is based on its location, this case is classified as wound myiasis, debridement and irrigation is the most proper management in this patient. The severity of myiasis is also depended on its larva species. The larvae of *P. regina* is one of the species that can infest around orifices of the head and may burrow into brain tissue. Wound care and maintain the immunological state are the key management of this patient, by taking ART and blood glucose regulation.

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AMM: provided scientific and bibliographic features and participated in writing the manuscript, clinical work and patient monitoring and reviewing the manuscript. UH: Supervision of the work and participating in the literature review and writing of the manuscript.

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