Case Report

A Severe Case of Brain Myiasis: Treatment Rationale and Review of Literature

Abstract

Cerebral myiasis is a rare condition caused by a parasitic infestation of fly larvae feeding on the host's necrotic or living tissue. Only 16 cases of cerebral myiasis have been published. We presented the case of a 72-year-old man with a neglected infestation of an extensive ulcerative cancer of the scalp. A large cranial lesion, with exposed brain and dura mater and severe Sarcophaga carnaria maggot infestation, was evident. We gently removed the maggots and covered the defect with thick gauze and sodium hypochlorite solution dressing. We additionally present a review of the literature to highlight shared features and suggestions for care management. In all cases, there was an absence of fatal meningitis and encephalitis, which is surprising given the open skull erosion with prolonged cortical exposure and points to the protective effects of larvae wound infestation.

Keywords: *Brain, cerebral infestation, larvae, myiasis, neglected*

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Introduction

Myiasis in humans is caused by a parasitic infestation of fly larvae feeding on the host's tissues.[1] Most cases occur in tropical and subtropical regions.[2] Myiasis in higher-income nations is typically associated with travel or immigration. Additional factors, such as hygiene, diabetes, immunocompromised status, and delay in seeking medical attention, may contribute.[3]

Cerebral localization of myiasis is exceedingly rare,[2-4] and the involvement of a large area of brain tissue can result in very severe manifestations. Here, we describe a rare case of cerebral myiasis in Northeast Italy and present a literature review to highlight the pathological features of and suggestions for managing such occurrences.

Procedure

In 2017, a 72-year-old male was diagnosed with right frontal basal cell cancer. The cancer was treated weekly with topical agents. Unfortunately, due to improper nursing care and missed clinical appointments, the patient developed a right frontal extensive ulcerative lesion.

In June 2019, the patient was found stuporous with a large cranial lesion with exposed brain and severe maggot infestation [Figure 1a].

The patient lived alone and was resistant to nursing care. He had been advised to seek medical attention repeatedly but had always refused. Maggot infestation went unrecognized by relatives. The patient was likely demented, which may explain his refusal of care and loss of awareness.

Vital signs were normal, but he was febrile (38.2°C/100.8°F). Complete blood cell count revealed a white blood cell count of 15.1 K/μL with 91% neutrophils [Supplementary Video 1]. Physical examination revealed bilateral frontal scalp and cranial erosion (13 cm × 11 cm) [Figure 1b]. The edges of the skin were hypertrophied and erythematous, and the dura mater was exposed. A cluster of approximately 100 maggots was found at the center of the defect, without apparent cerebrospinal fluid (CSF) leakage. A computed tomography (CT) scan [Figure 2] revealed an extensive frontal bony defect with brain exposure.

We urgently cleaned the exposed wound to debride the maggots, obtain tissue specimens, and protect brain integrity. An infectious disease specialist and a

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plastic surgeon were contacted, and a literature search was completed to plan the best approach. (Cerebral myiasis video).

The patient was positioned supine and tilted into a Trendelenburg position, using gravity to aid in removing the maggots and checking for spontaneous CSF leakage [Figure 1c]. After the removal of the larvae, the surface was moisturized with a 50:50 sodium hypochlorite sterile saline solution.

After maggot removal, the bone was debrided until it obtained a normal aspect. The dura mater was gently scratched to remove any debris. The sagittal sinus was not touched to avoid bleeding, and the infiltrate tumor was debulked where possible [Figure 3a]. In the absence of signs of CSF leakage, we avoid dura substitutes to lower the risk of infection. The exposed cortex was covered with patches of fibrin and collagen matrix. The defect was eventually covered with a sodium hypochlorite solution dressing and fat gauzes [Figure 3b].

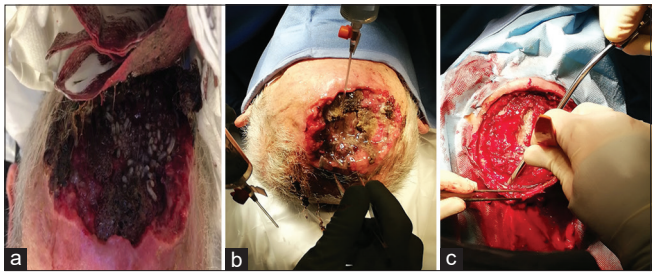

Figure 1: (a) Severe frontal maggot infestation. (b) Larvae debridement with saline-sodium hypochlorite solution. (c) Removing necrotic and pathologic bone and skin tissue from defects margins

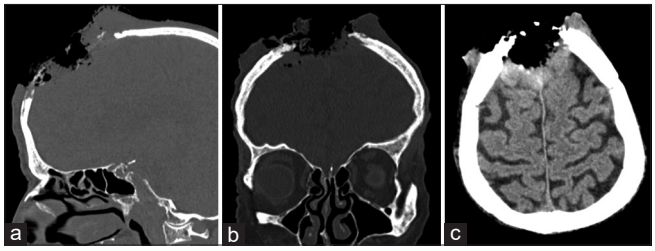

Figure 2: (a) Sagittal bone computed tomography scan. (b) Coronal bone computed tomography scan. (c) Axial computed tomography scan. It is evident the severe and profound destruction of the scalp layer done by the basal cell carcinoma

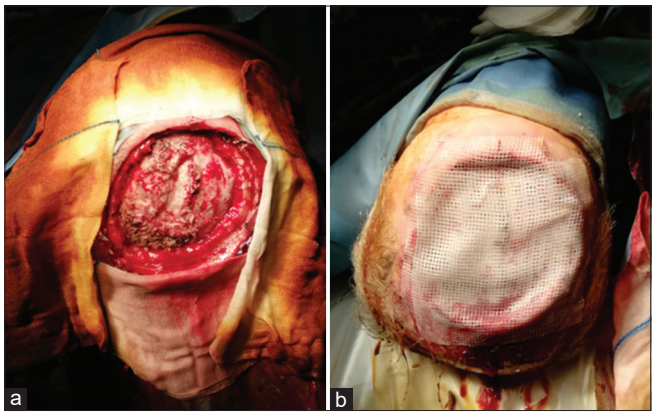

Figure 3: (a) Debrided defect. Remnants of the superior sagittal sinus are visible in the center. (b) Dressin g the defect with fatty gauze

We administered intravenous broad-spectrum antibiotics. Urine and blood cultures were negative, and the fever resolved rapidly. There were no signs of new larvae development. The maggots were classified as belonging to the *Sarcophaga carnaria* species. Microbiological and CFS cultures were normal. After defervescence, the patient was discharged to a medical ward, and plastic reconstruction was planned. Unfortunately, the patient refused further intervention and died two months later.

See proposed video for case presentation. The case is described and narrated extensively in the video provided with the paper (Cerebral myiasis video).

Discussion

Both primary and secondary cerebral myiasis are exceedingly rare conditions, especially in high-income countries.[1,2,5] In primary myiasis, the larvae penetrate the skin via a puncture wound, while in the secondary variety, fly eggs are laid into a skin ulcer.[4] Benign or malignant dermatological or traumatic conditions have been associated with secondary myiasis, but rarely as extensively as presented here[3-6] [Table 1].

Cerebral myiasis is rare, with only 16 published cases, and may lead to life-threatening complications.[2-4,7-15] The majority of known cerebral myiasis cases in higher-income countries were consequent to neglected skin tumors,[3,14,15] while in lower-income countries, they were mostly attributable to inadequate wound care.[4,9,16]

Almost all cerebral myiasis cases were found close to the scalp surface in frontal regions.[3,8,14-17] Rarely, intraparenchymal involvement with consequent hematoma formation or abscess development was found.[10,13,17] Symptom presentation may be delayed from a few days to years, particularly among patients with dementia. The larvae may penetrate the brain and spread via CSF to the subependymal space, where it can remain undetected for years.[10] Deep parenchymal involvement and older age [4,14,16,18,19] are associated with decreased survival.[10,16,17,18,19]

Proper management requires a multidisciplinary team (i.e., infectious specialists, radiologists, neurosurgeons, and plastic surgeons). Among patients with a confirmed calvarium erosion, a contrast-enhanced CT or magnetic resonance imaging (MRI) is recommended to rule out venous sinuses' integrity or for phlogistic processes.[6] MRI imaging may also help assess the integrity of arachnoidal layers or signs of infection. Furthermore, CSF sampling is recommended for cytological and microbiological evaluation.

Table 1: Cases of cerebral myiasis reported in the literature

Author	Age, sex	Race	Nationality	Predisposing factors	Symptoms/signs	Symptoms duration/previous events	Location	Imaging
Froomin 1939[12]	50 years, female							
Semenov 1969[19]I	4 years, male						Occipital	
Zucoloto and Ross, 1971[11]	Unknow, male 53 years, male							
Rossi and Zucoloto, 1973[16]	5 months, female	American Indian/ South American	Brazilian		Lump on her scalp	Unknow	Frontal	
Gilly *et al*., 1976[7]	7 years, male						Frontal	
Pouillaude *et al*., 1980[17]I	6.5 years, male				Intracerebral hematoma			
Arbit *et al*., 1986[15]	63 years, male	White	Canadian		Crawling in the head	Unknow	Frontal	
Kalelioğlu *et al*., 1989[13]	8 years, male	White	Turkish	None	Focal motor-type convulsions	10-days	Parieto-occipital	CT
Cheshier *et al*., 2007[3]	75 years, male	White	American	Angiosarcoma		unknow	Frontal	
Marco de Lucas *et al*., 2008[10]	11 years, male	American Indian/ South American	Colombian		Seizures + hydrocephalus	2 years history of nonspecific headaches	Frontal + ventricle	CT/MRI
Terterov *et al*., 2010[14]	42 years, male	White		HIV			Frontal	CT
Giri *et al*., 2016[9]	38 years, male	Asian	Indian	Alcoholic	Fever and limb weakness	Subdural hematoma + depressed comminuted fracture	Fronto-temporal	CT
Navarro and Alves, 2016[8]	36 years, male	American Indian/ South American	Brazilian	Melanoma			Frontal	
Piña-Tornés *et al*., 2016[2]	30 years, male	Am. Indian/ South American	Equadorian	Skizofrenia			Parieto-occipital	
Aggarwal and Maskara, 2018[4]	26 years, male	Asian	Indian		Crawling sensation	Neglected burn injury	Parietal	CT
Present case, 2020	72 years, male	White	Italian	Oncologic disease	Unknow	Unknow	Frontal	

Author	Entrance path	Brain involvement	Species	Treatment	Medical treatment	Outcome
Froomin 1939[12]		Superficial	*H. bovis*			Unknow
Semenov 1969[19]I			*H. lineatum*			Death
Zucoloto and Ross, 1971[11]		Deep	Callitroga American			Death Death
Rossi and Zucoloto, 1973[16]	Infested scalp lesion	Deep	*D. hominis*	Debridement		Death
Gilly *et al*., 1976[7]			*H. bovis*			Death
Pouillaude *et al*., 1980[17]I		Deep	*H. bovis*			Death
Arbit *et al*., 1986[15]	Neglected squamous cell carcinoma	Superficial	Diptera sarcophaga	Debridement + reconstructive surgery	IV broad-spectrum antibiotics	Discharged alive
Kalelioğlu *et al*., 1989[13]	Unknown	Deep	*H. bovis*	Debridement	Anticonvulsant therapy	No neurological deficit
Cheshier *et al*., 2007[3]	Neglected angiosarcoma of the scalp	Superficial	*P. sericata*	Debridement	IV broad-spectrum antibiotics +	Refused additional care/ discharged alive

Contd...

Table 1: Contd...

Author	Entrance path	Brain involvement	Species	Treatment	Medical treatment	Outcome
Marco de Lucas *et al*., 2008[10]	Frontal fracture	Deep	*D. hominis*	Refused surgery		Unknow
Terterov *et al*., 2010[14]	Frontal fracture	Superficial	Unknow	Debridement + reconstructive surgery	IV broad-spectrum antibiotics + antifungal drugs	Living independently
Giri *et al*., 2016[9]	Frontal craniectomy	Superficial	Musca domestica nebulo	Debridement + reconstructive surgery	IV broad spectrum antibiotics	Improved
Navarro and Alves, 2016[8]	Neglected melanoma		Unknow	Debridement	IV broad-spectrum antibiotics + antiparasitic drugs	Discharged alive
Piña-Tornés *et al*., 2016[2]			*D. hominis*	Debridement + reconstructive surgery	IV broad-spectrum antibiotics	Discharged alive
Aggarwal and Maskara, 2017[4]	Neglected frontal	Superficial	Unknow	Debridement + reconstructive surgery	IV broad-spectrum antibiotics	Discharged alive
Present case, 2020	Neglected basal cell carcinoma	Deep	*S. carnaria*	Debridement	IV broad-spectrum antibiotics	Discharged alive, refused additional care

1 – Only abstract; HIV – Human immunodeficiency virus; CT – Computer tomography; MRI – Magnetic resonance imaging; *H. bovis – Hypoderma bovis; H. lineatum – Hypoderma lineatum; D. hominis – Dermatobia Hominis; P. sericata – Phaenicia sericata; S. carnaria – Sarcophaga carnaria*

Treatment relies on larvae removal, debridement of necrotic/malignant tissue, and reconstruction of the defect. In the literature, several agents or drugs are proposed for larvae removal.[1,15] In the case of massive erosion and brain exposition, gentle irrigation with saline solution and mechanical removal is suggested.[2,9,14,15] A sodium hypochlorite solution, both intraoperatively and as a dressing, is also recommended. Intravenous broad-spectrum antibiotics (>6 weeks) are mandatory to control and prevent secondary infection. In addition, in cases with brain or dural sinus invasion, it is better to limit the debulking maneuvers to visible lesions to avoid additional damage.[4] Further interventions, such as reconstruction surgeries, should be considered only after myiasis resolution.

There was a surprising absence of meningitis or encephalitis in the setting of an open skull erosion with prolonged cortical exposure in all available reports. Maggot infestation may have reduced bacterial infection risk by protecting the tissue surface area.[3,9,15] Although the patient was febrile, there was no evidence of systemic infection. This may be attributable to the beneficial effects of wound infestation with larvae, which were first used medically during the American Civil War.[15] The larval activity might have prevented a secondary bacterial infection by eating dead tissues, leading to more prolonged survival despite delayed treatment.[9,14]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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