The background of the slide is a microscopic view of red blood cells. The left side features a light blue background with several large, semi-transparent white circles. The right side is a dark background filled with numerous red blood cells, some in sharp focus and others blurred, creating a sense of depth.

An Unusual case of OI in advance HIV

Dr. Kalpak Bhatt

Dr. Dhruv Patel

Dr. Harsh Toshniwal

IDTM Associates

Ahmedabad

- Mr BD 33-year-old male admitted in our hospital on 8th June 2024
 - Imparting past history :diagnosed HIV 1 in 2016, taken irregular ART for 3 years and stopped since 2019.
- Present complains since 20 days
 - Low grade intermittent fever
 - Difficulty in sitting, standing & walking : gradually increasing
 - Increasing pain in extremities, started from lower extremities, now affecting upper extremities.
 - Admitted for above complaint in tertiary care hospital Udaipur from 4th June to 7th June 2024. Did not improve.
- Discharge Diagnosis was:
 - AFI with acute onset progressive paraparesis with sensory involvement.
 - Inflammatory myopathy
 - PLHA.

Investigation : Udaipur

- SGPT 285 (10-50)
- SGOT : 329 (10-50)
- CPK Total : 2027
- LDH : 641 (135-214)
- Absolute CD4 : 33
- NCV : Sensory Axonal Neuropathy.

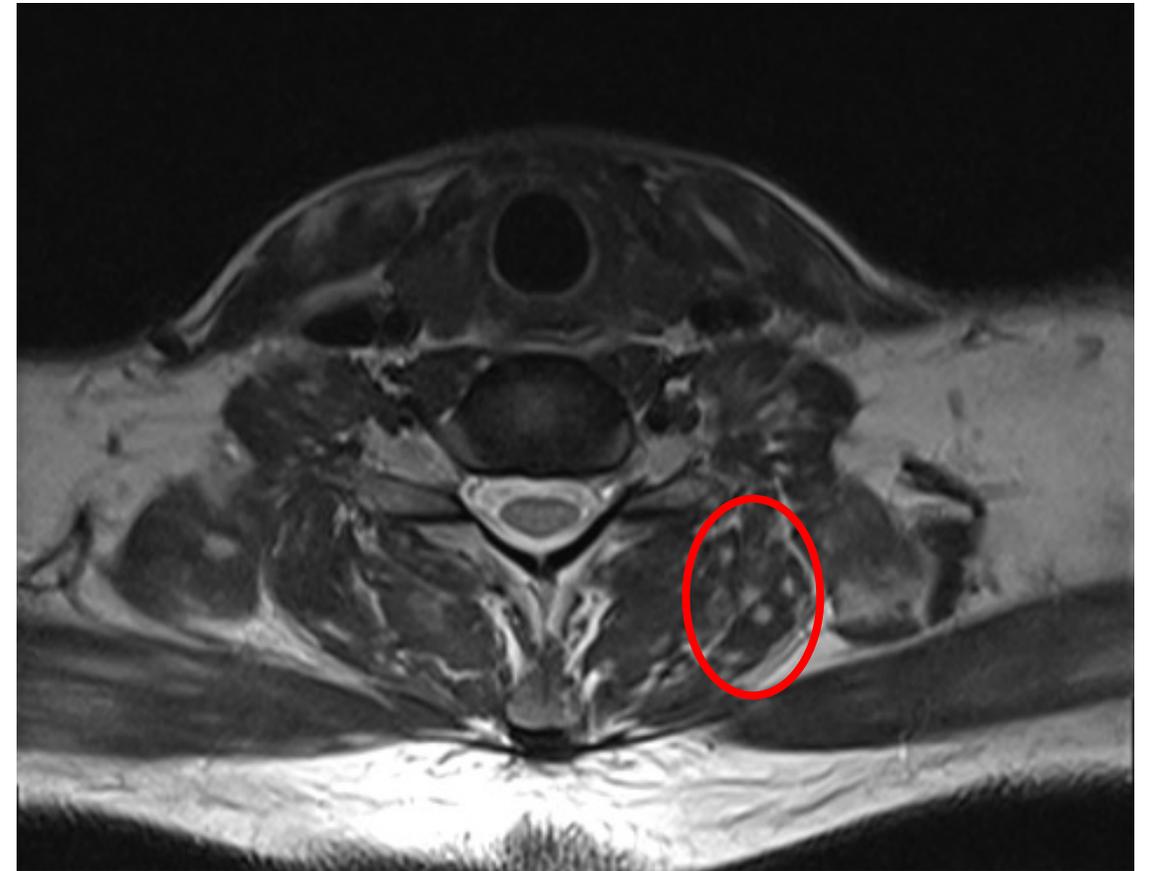
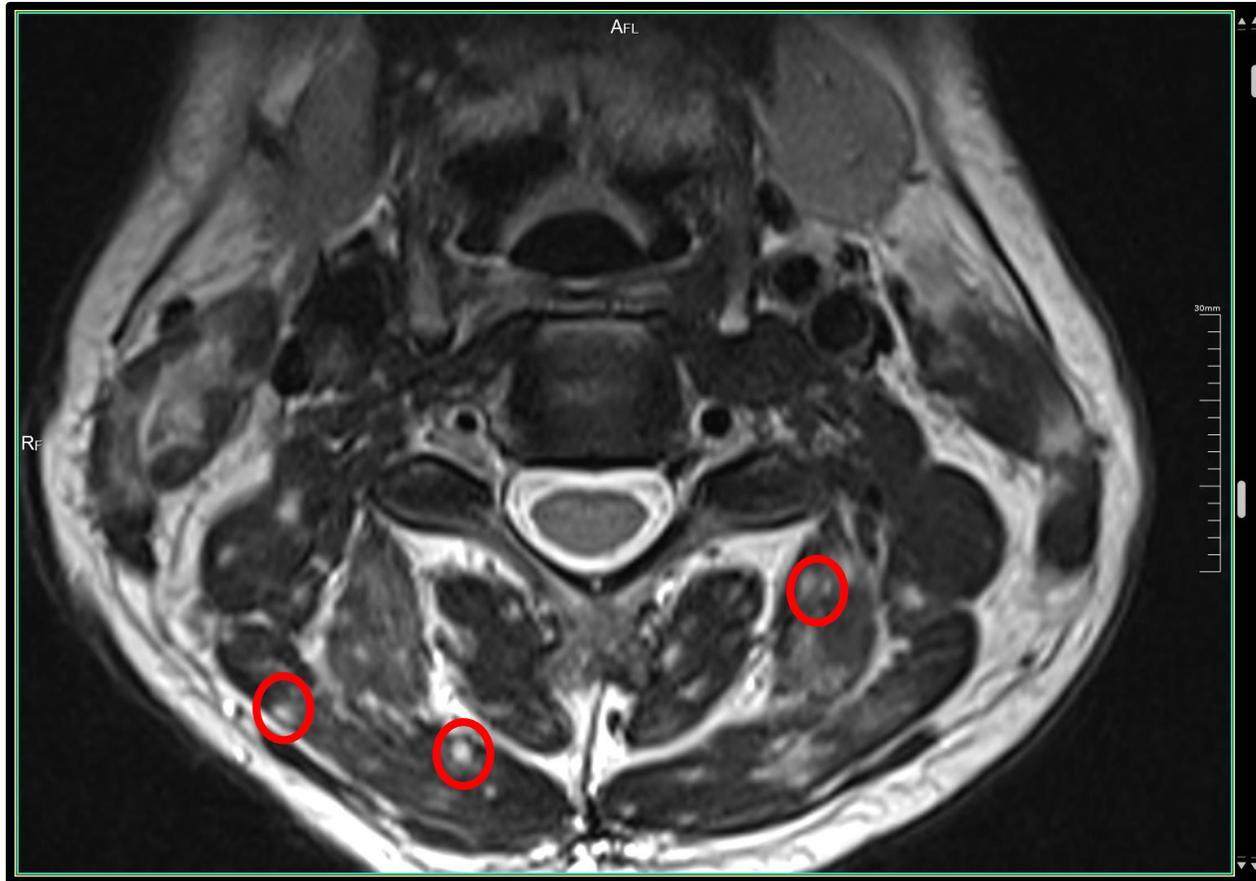
Date	05/06/2024
Hb	12.1
TC	7500
PC	263000
ESR	97
CRP	52
LDH	641
CPK TOTAL	2027
Creatinine	0.5
Na	134.6
K	4.6
Urine Myoglobin	
SGPT	285
SGOT	329
Alkpo4	117
Bilirubin	0.31
S. Protein	6.6/2.3/4.3
HBsAg	NR
HCV	NR
VDRL	NR
S. Typhus	NR
Dengue PCR/ NS1	NR
B12	2000

	Udaipur	Ahmedabad
Date	05/06/2024 :	08/06/2024
Hb	12.1	
TC	7500	
PC	263000	
CD4	33	
ESR	97	
CRP	52	
LDH	641	1033.57 (125-200)
CPK TOTAL	2027	3709.46 (30-200)
Creatinine	0.5	
Na	134.6	
K	4.6	
Urine Myoglobin		800 (<65)
SGPT	285	264
SGOT	329	
Alkpo4	117	
Bilirubin	0.31	
S. Protein	6.6/2.3/4.3	
HBsAg	NR	
HCV	NR	
VDRL	NR	
S. Typhus	NR	
Dengue PCR/ NS1	NR	
B12	2000	

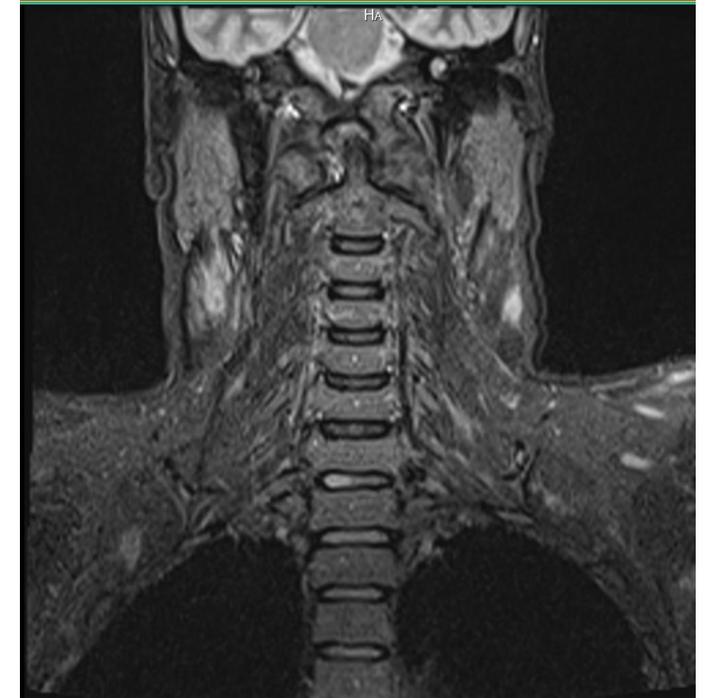
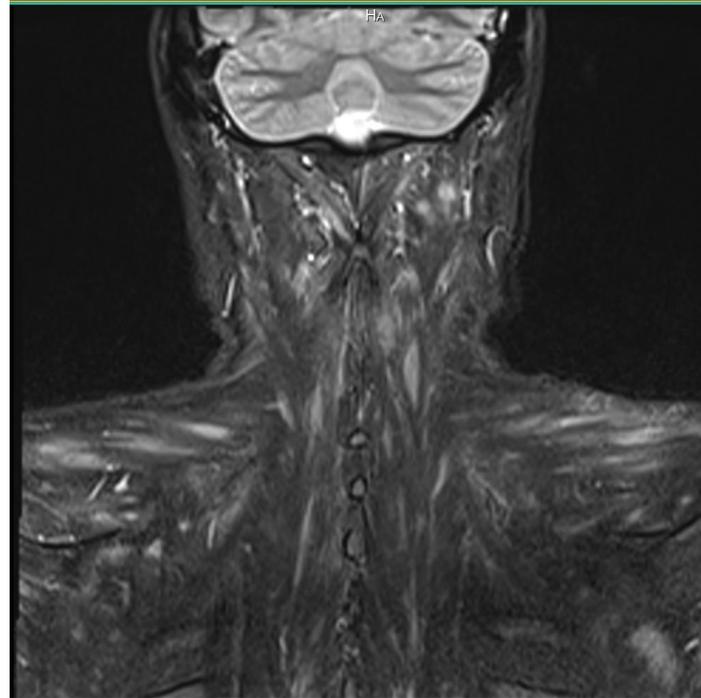
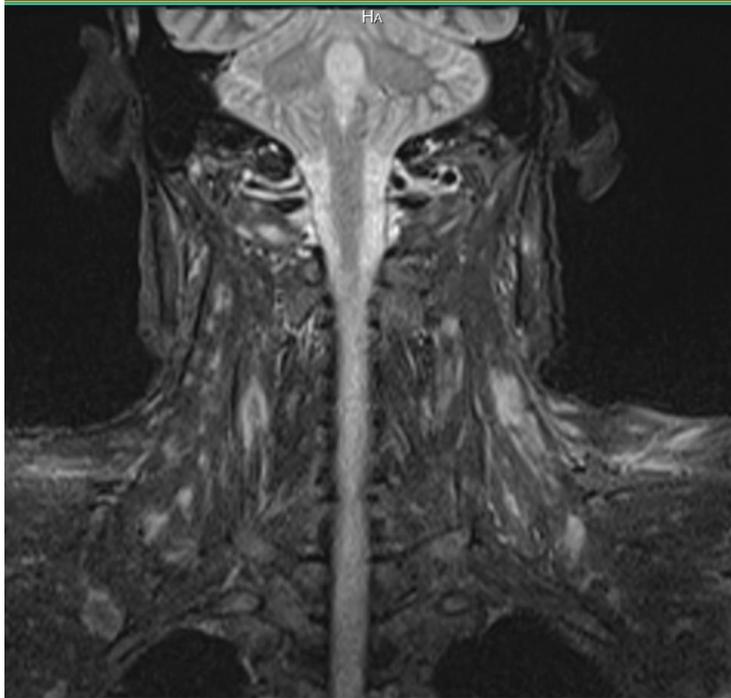
Ahmedabad : 08-06-2024

- **Opinion :**
 - Neurologist & Immunologist
 - ANA by If & Blot : Negative.
- **MRI Brain:** No evident acute infarct or intracranial SOL.
- **MRI spine:** patchy hyperintense cystic areas (T2 images) involving multiple muscle of cervical, thoracic, lumbar and pelvic regions- suggest changes of myositis. MR appearance could represent changes of **disseminated cysticercoses infection.**

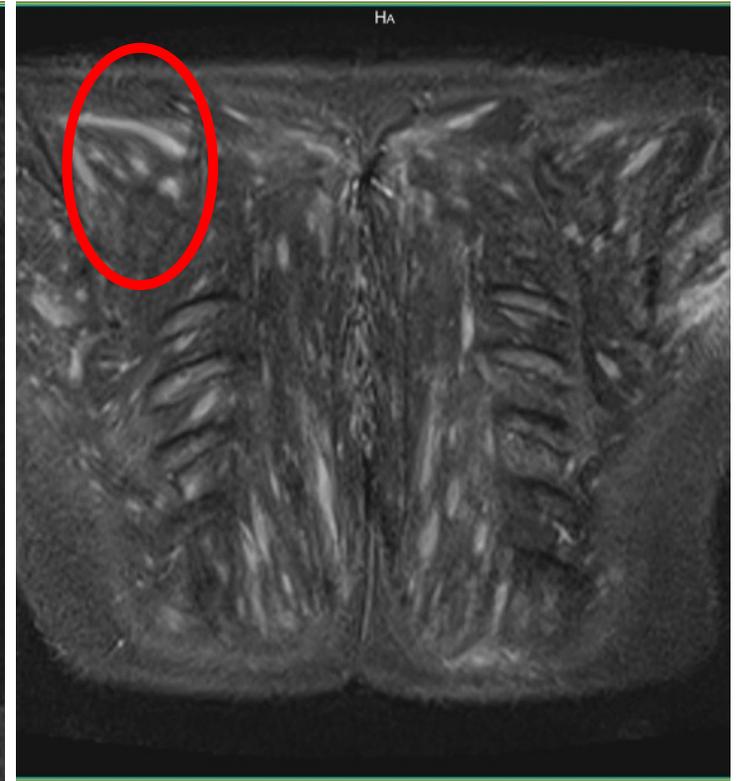
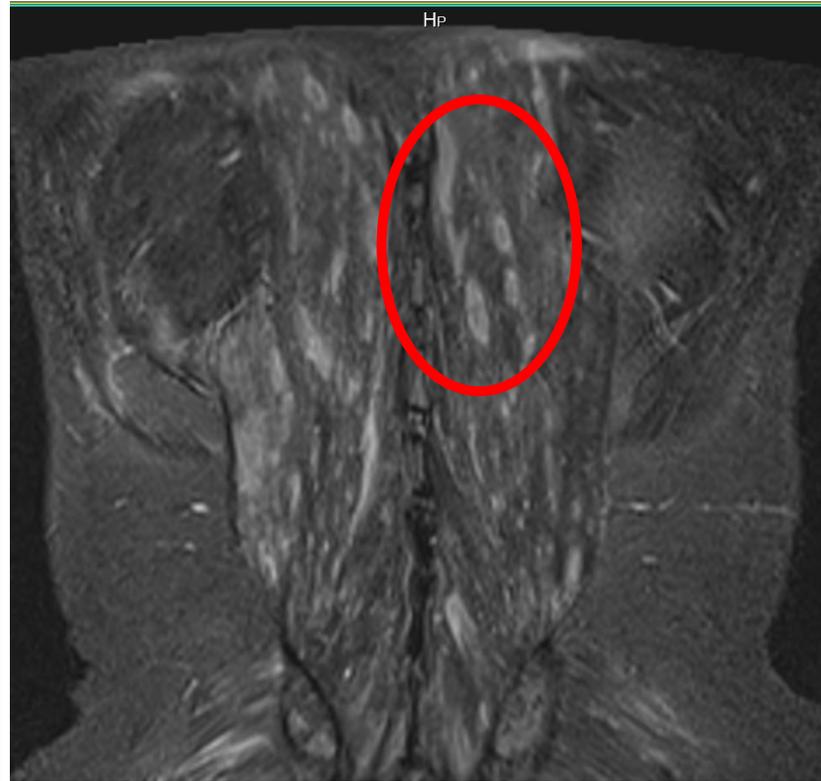
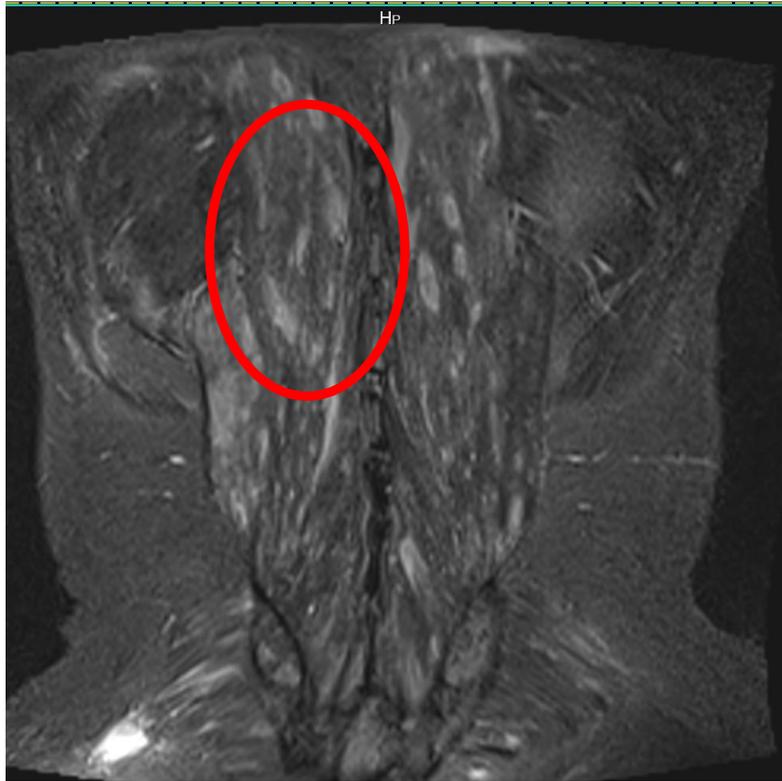
Multiple hyperintense foci in sternocleidomastoid & paraspinal muscles



Multiple discrete hyperintense foci in paraspinal muscles & chest wall in cervico-dorsal region.



Multiple discrete hyperintense foci cysts in posterior paraspinal muscles (Coronal Plain)



Case continued

09/06/24

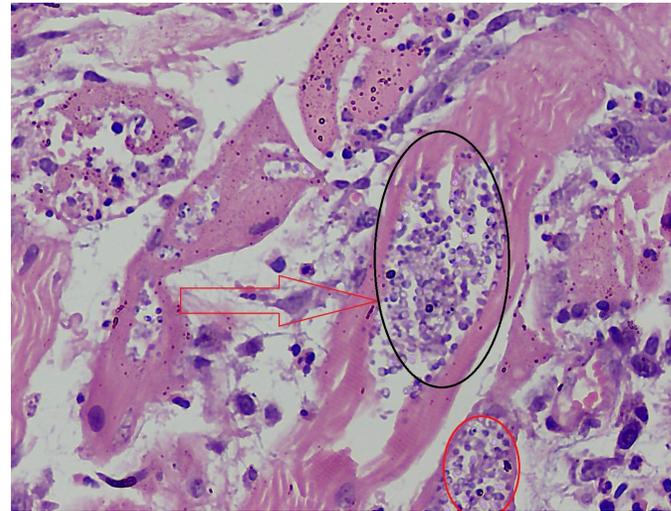
- Considering parasitic infection, we ordered stool for OI.
- Stool Microscopy: Few Rhabditiform larvae of ***Strongyloides Stercoralis*** are seen.
- Hyperinfection syndrome of ***S. stercoralis***



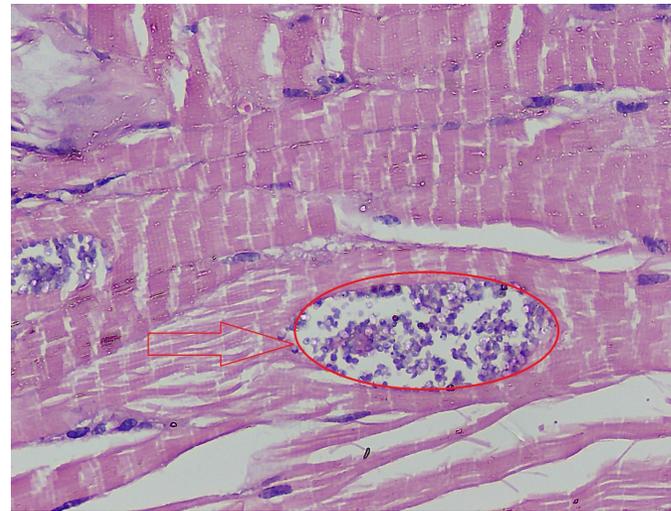
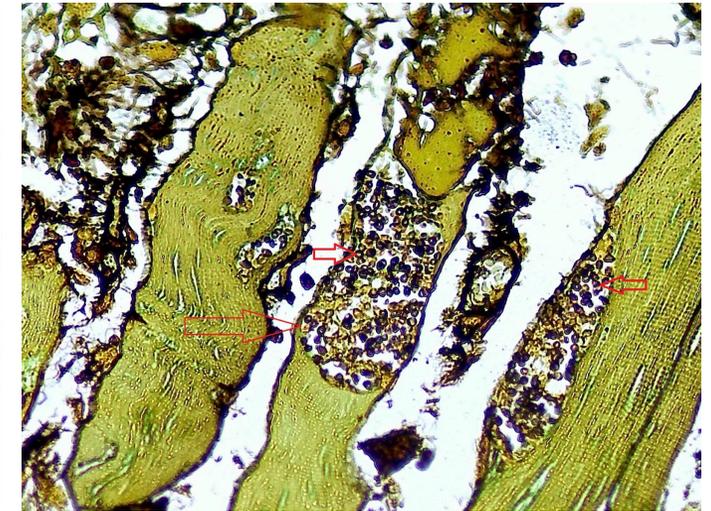
10/06/2024:

- Muscle biopsy was performed from most tender area of neck
- **HPE:** The myofibers show presence of clusters of unstained ovoid yeast like body
- Consistent with microsporidia.
MICROSPORIDIAL MYOSITIS

HE Stain



GMS Stain



Treatment

- Albendazole 400 mg 1 Bid
- Ivermectin 200 mcg/Kg
- Itraconazole 200 mg 1 Bid
- TMP SMX DS 1 Bid
- ART (DTG+3TC+DRV/r)
- Patient started improving clinically
- Was able to walk with support
- Biochemistry improved
- Patient was discharged on 9th day (16th June 2024)
- Follow up on 21th day

Date	05/06/2024	08/06/2024	11/06/24	16/06/24
LDH	641	1033.57		516
CPK TOTAL	2027	3709.46	2624.78	862
Creatinine	0.5		0.50	0.64
Urine Myoglobin		800 (<65)		615
SGPT	285		217	182
SGOT	329		478	233

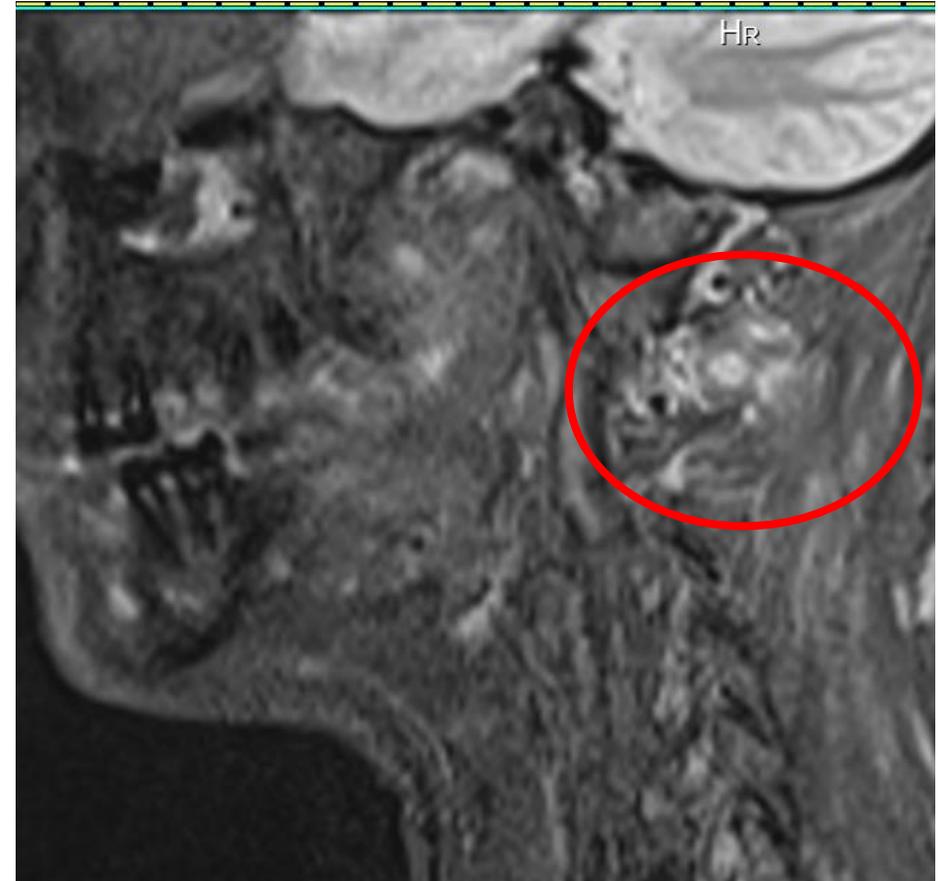
Case continued

- Patient returned on unscheduled visit after 8th day of discharge (24th June 2024)
- Severe pain in all extremities
- Dyspnoea
- Difficulty in deglutition
- Blurred vision
- High fever
- All muscle of body were tender on touch
- Blood Test & MRI was requested

Date	05/06/2024	08/06/2024	11/06/24	16/06/24	24/6/24
LDH	641	1033.57		516	
CPK TOTAL	2027	3709.46	2624.78	862	2678
Creatinine	0.5		0.50	0.64	0.46
K					4.6
Na					124
Urine Myoglobin		800 (<65)		615	
SGPT	285		217	182	227
SGOT	329		478	233	289

- Blood Culture : 2 Sets : Negative
- USG ABD & soft Part : WNL except accidental finding of gluteal abscess (4cm x 4cm)
- USG guided PUS Aspirated
 - Direct Microscopy :
 - *GPC, Few septate branching fungal hyphae, AFB Negative*
 - *Modified Trichrome Stain : Plenty of Microsporidia.*
 - Bacterial Culture grew : MRSA.

Tongue & Tonsillar fossa



Multiple discrete hyperintense foci are seen involving muscles.

Case continued

- Albendazole, TMP SMX, Itraconazole and ART were continued in same dose.
- Pyrimithamine 25 mg : 2 Bid day 1 F/B 1BID
- Nitazoxanide 500 mg : 2 Bid
- Inj. Doxycycline 100mg i/v Bid
- Inj. Methylprednisolone 40 mg Bid
- RT insertion, Foleys catheter & symptomatic management started

Date	05/06/2024	08/06/2024	11/06/24	16/06/24	24/6/24	30/6/24
LDH	641	1033.57		516		
CPK TOTAL	2027	3709.46	2624.78	862	2678	11324
Creatinine	0.5		0.50	0.64	0.46	0.69
K					4.6	5.6
Na					128	132
Urine Myoglobin		800 (<65)		615		583
SGPT	285		217	182	227	547
SGOT	329		478	233	289	983

Case continued

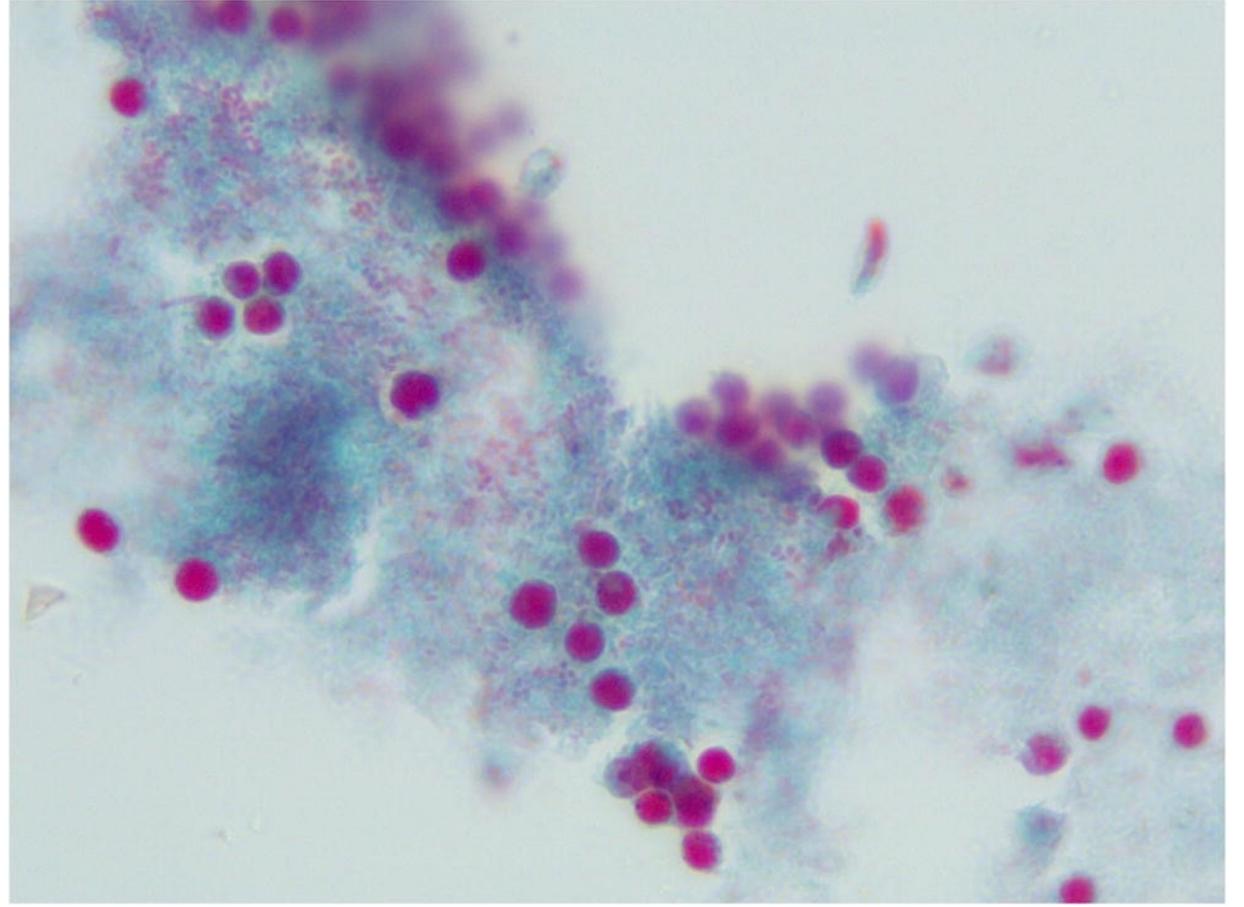
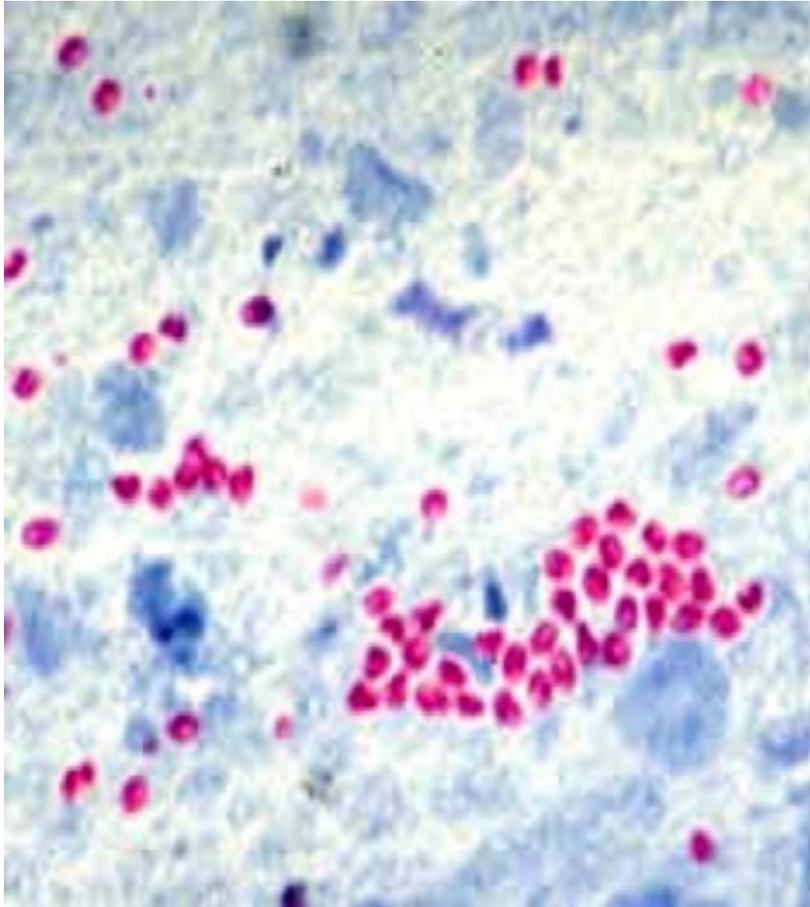
- Patient further deteriorated
- Shifted to ICU
- External ocular muscles were paralyzed with no movement of eye balls
- Relative refused for ventilatory support
- Patient succumbed on 9th day of admission

Literature review : Microsporidia

- Intracellular spore forming organisms-ubiquitous in the environment
- Approximately 220 genera & 1700 species are described
- Previously considered protozoa, now reclassified as fungi
- Clinical manifestations are diverse and include intestinal, pulmonary, ocular, muscular & renal disease.
- Diarrhoea or keratoconjunctivitis : most common presentation.
- Disease in immunocompetent and immunocompromised host
- Many species have been reported to infect humans as OI's in association with AIDS.

- Transmission
 - Not fully understood
 - Foodborne, waterborne, person to person, animal to person, Vector borne, airborne transmission may be possible
 - Myositis is associated with pleistophora spp and Trachipleistophora hominis. (T. hominis)
- Diagnosis
 - Microscopic or Genomic detection of spores in stool, body fluid or tissue specimen.
 - Light microscopy with a modified trichrom stain is used for diagnosis

Modified trichrome stains : microsporidia spores pink against blue/ green background.



Hosts and diseases associated with Microsporidia

Organism	Nonhuman species	Disease	HIV	Non-HIV
<i>Enterocytozoon bieneusi</i>	Pigs, primates	Intestinal, hepatobiliary	+	+
<i>Encephalitozoon intestinalis</i> (<i>Septata intestinalis</i>)	Some mammals	Disseminated	+	+/-
<i>Encephalitozoon hellem</i>	Parakeets	Disseminated	+	-
<i>Encephalitozoon cuniculi</i>	Rabbits, mice, cats, foxes, dogs	Corneal stroma	+	+
<i>Nosema connori</i>	-	Disseminated	+	-
<i>Nosema ocularum</i>	-	Corneal stroma	+	+
<i>Nosema</i> -like species	-	Corneal stroma	+	+
<i>Trachipleistophora hominis</i>	-	Skeletal muscle, nasal sinuses	+	-
<i>Trachipleistophora antropoptera</i>	-	Disseminated		
<i>Pleistophora</i> spp	Fish	Skeletal muscle	+	+
<i>Vittaforma corneae</i> (<i>Nosema corneae</i>)	-	Corneal stroma	+	+
<i>Microsporidium</i> spp (species not fully classified)	-	Disseminated		

UpToDate®

- Treatment

- Albendazole 400mg twice daily - Effective against most microsporidia species. Duration 3 weeks
- Other agents: Itraconazole, metronidazole, azithromycin, doxycycline, sparfloxacin, sulpha drugs, atovaquone, nitazoxanide, octreotide, paromomycin.
- Species specific diagnosis is useful for guiding treatment.
- *E. bienersi* Infection: Fumagillin 60MG daily for 14 days
- Simultaneous ART is to be started

MICROSPORIDIAL POLYMYOSITIS IN HUMAN IMMUNODEFICIENCY VIRUS–INFECTED PATIENTS, A RARE LIFE-THREATENING OPPORTUNISTIC INFECTION: CLINICAL SUSPICION, DIAGNOSIS, AND MANAGEMENT IN RESOURCE-LIMITED SETTINGS

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ABSTRACT: *Introduction:* Microsporidial myositis is a rare opportunistic infection that has been reported in HIV-infected and HIV-uninfected immunocompromised patients. *Methods:* In this study we present a retrospective analysis of 5 cases of microsporidial myositis in HIV-infected patients, including the clinical, laboratory, and histologic features, and a review of the literature. *Results:* Five young men with HIV infection [median CD4 count of 20 cells (range 14–144)/mm³] who presented with signs and symptoms suggestive of myositis underwent EMG-NCV and muscle biopsy, which revealed signs compatible with microsporidial myositis. Early and aggressive treatment led to improvement in 3 patients. Two of the 5 patients died due to a delay in diagnosis, because the spores were mistaken for *Candida* without confirmatory stains or a high index of suspicion. *Conclusions:* Myositis in HIV-infected patients with low CD4 counts should be evaluated using muscle biopsy. A high index of suspicion is required for early diagnosis of microsporidial myositis in HIV-infected patients. Early diagnosis and immediate, aggressive treatment are the keys to favorable outcomes in these patients.

Muscle Nerve 51: 775–780, 2015

opportunistic infections due to a lack of access to ART or to poor compliance with treatment. HIV-infected patients can develop a variety of muscle disorders due to infectious and non-infectious causes.^{1–10} *Trichinella* spp. (trichinosis), *Taenia solium* (cysticercosis), microsporidia, and *Toxoplasma gondii* (toxoplasmosis) are the most common parasites that cause myositis.^{4,11–13} Microsporidial myositis has been reported in HIV/acquired immunodeficiency virus (AIDS) patients, with the incidence of this infectious disease continuing to increase in immunocompromised hosts. AIDS patients with CD4 cell counts <100 cells/mm³ have the highest risk of acquiring microsporidial myositis.^{12–19} In addition, patients on posttransplant immunosuppressive therapy, patients with

Microsporidial myositis in adult-onset immunodeficiency: case-based review

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Abstract

Polymyositis is a diagnosis of exclusion. In patients with odd features, it can be of infective etiology. A high index of suspicion is required for diagnosis. A 55-year-old gentleman presented with gradual-onset proximal muscle weakness. Examination revealed mild distal weakness but no rash. Muscle enzymes were raised and tests for autoantibodies were negative. Biopsy revealed microsporidiosis. In view of this unusual infection, immunodeficiency was considered and he was found to have lymphopenia which antedated his illness. Later, he developed cranial nerve palsies due to multiple lesions in the pons. In addition, he had Cytomegalovirus viremia. Literature was reviewed to identify 20 cases of microsporidial myositis, its presentation, underlying immunodeficient state, and clinical course. Infective polymyositis should be considered in a patient with paucity of clinical and serological autoimmune features. Lymphopenia can point to underlying immunodeficiency. CMV infection could be the contributor to or bystander-effect of idiopathic lymphopenia.

Keywords: Immunodeficiency; Lymphopenia; Microsporidiosis; Myositis.

Rare Occurrence of Microsporidial Myositis Involving Masticatory Muscles

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MD | [AUTHORS INFO & AFFILIATIONS](#)

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A 45-year-old woman with AIDS on antiretroviral therapy (tenofovir disoproxil, lamivudine, dolutegravir) presented with painful mastication and generalized musculoskeletal pain for 6 weeks. She had bilateral hand contractures, with tender masseters, temporalis, and gastrocnemii. Absolute eosinophil count was 500 cells/mm³, creatine phosphokinase 66 U/L, and CD4 count 147 cells/mm³. Cranial MRI showed infiltration of masticatory muscles ([Figure 1](#)). Muscle biopsy revealed microsporidial myositis ([Figure 2](#)). Microsporidia are obligate intracellular fungi classically affecting immunocompromised hosts.¹ We present findings of rare microsporidial myositis affecting masticatory muscles. Muscle biopsy is diagnostic and excludes potential clinical (polymyalgia rheumatica, temporal arteritis), medication-related, and histologic mimics.²



Take Home message

- Rarely occurring OI in HIV – probably under reported
- High index of suspicion
- High mortality if diagnosis is delayed
- All patients of HIV having myositis : subjected for biopsy with special stains
- HIV patients having diarrhoea should be ordered for modified trichrome stains
- Whenever possible sequencing should be ordered.

- Could this be IRIS ?
- Should I have deferred ART ?
- Science of ART is DYNAMIC ?
- Fumagillin : your experience ?

Thank you

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