Case Report

DOI: https://dx.doi.org/10.18203/issn.2454-5929.ijohns20233586

Cytomegalovirus esophagitis associated with arytenoid cyst in an immunocompetent host

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Received: 10 April 2023 Revised: 07 November 2023 Accepted: 11 November 2023

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ABSTRACT

CMV esophagitis is usually encountered in immunocompromised individuals and esophagus is rarely affected in immunocompetent individuals. CMV esophagitis associated with arytenoid cyst has not yet been reported in literature to the best of my knowledge and can be considered in case of recurrence of cyst following excision. The objective of this study is to report the unique presentation of CMV in an immunocompetent patient. A middle-aged immunocompetent lady presenting with dysphagia was diagnosed to have arytenoid cyst. Biopsy from upper esophagus revealed cytomegalovirus inclusion bodies and immunological test were performed to confirm the pathology. The cyst excision lead to recurrence. It was excised again followed by a course of intravenous ganciclovir. Currently, the role of antiviral in treatment of CMV esophagitis in immunocompetent individual is not well established. We have obtained good result in this patient with a course of Ganciclovir following excision of the cyst. The rarity of cytomegaloviral upper esophageal involvement, its association with arytenoid cyst and the utility of antivirus in treatment of an immunocompetent host makes this case report unique and interesting.

Keywords: Cytomegalovirus, Esophagitis, Ganciclovir, Arytenoid, Immunocompetent, Cyst

INTRODUCTION

Cytomegalovirus infection of gastrointestinal tract was first described in 1960s. The most common site to be involved is colon. CMV esophagitis is a rare entity in immunocompetent patients and usually present with superficial or deep ulcers but some even present with necrosis and polyps. The most common presenting symptoms are dysphagia and odynophagia.

CASE REPORT

A 50 year old female patient presented to our outpatient department with history of dysphagia. She had no complaints of fever, hematemesis, pain abdomen or myalgia. Upper GI endoscopy revealed arytenoid cyst with multiple superficial mucosal ulcerations in upper esophagus (Figure 1).



Figure 1: Arytenoid cyst noted with ulcerated upper esophageal mucosa. Rest of the larynx and hypopharynx was within normal limits.

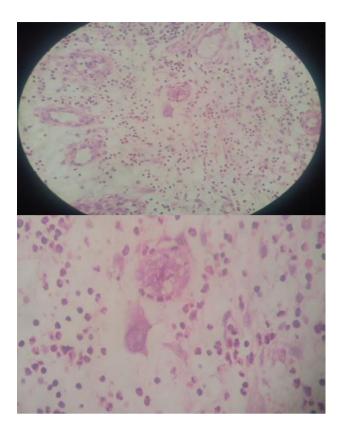


Figure 2: Endothelial cells with intranuclear viral inclusion body surrounded by neutrophilic infiltrate, endothelial cell with cytomegaly with intranuclear viral inclusion and rarefaction of chromatin of nucleus

Biopsy was taken from the upper esophagus and microscopic examination (Figure 2) revealed ulceration of epithelium, mixed inflammatory cell infiltrate and proliferation of blood vessels

Vascular endothelial cells showed cytomegaly with large nucleus and intranuclear haziness (viral inclusions). Routine blood investigations and liver function tests were within normal limits. Serology was negative for anti HIV and anti HCV antibodies and HBsAg. Ophthalmology reference was sought to rule out any pathology in the eye. The patient underwent microlaryngoscopic excision of the cyst. There was recurrence 6 months following surgery. The cyst was excised again followed by a course of slow intravenous ganciclovir 10 mg/kg per day for 21 days (other options are valciclovir, foscarnet, cidofovir or combination therapy). It was well tolerated by the patient with no side effects.

Immunological tests-Table 1-3.

Table 1: Serological test.

Cytomegalovirus Ab	Results	Reference value
IgG	143 U/ml	<12 U/ml
IgM	5 U/ml	18 U/ml

Table 2: Interpretation.

CMV IgG results	CMV IgM results	Remarks	
<12 U/ml	<18 U/ml	Nonreactive	
12-14 U/ml	18-22 U/ml	Equivocal	
>14 U/ml	>22 U/ml	Reactive	

Table 3: Interpretation of serological test.

CMV IgG	CMV IgM	Avidity	Remarks
Nonreactive	Nonreactive	Not applicable	Infection unlikely
Reactive	Nonreactive	high	Past infection
Reactive	reactive	low	Primary infection
Reactive	reactive	High	Non- primary infection

This patient had high CMV IgG levels with low IgM level indicating past infection.

The patient is on regular follow-up and has no dysphagia following treatment. A repeat upper GI endoscopy done 4 weeks following surgery revealed normal larynx with complete disappearance of the cyst and normal esophageal mucosa with healing of ulcers. Repeat upper esophageal biopsy showed normal mucosa with no evidence of CMV inclusion bodies.

DISCUSSION

CMV belongs to the group of herpes virus and is characterized by enlargement of infected cells and prominent intra-nuclear inclusions. It presents with prolonged latency in infected hosts similar to other herpes virus. Immunodeficient patients can present with severe disseminated disease whereas normal person may be asymptomatic. Though infection with the virus is common, the disease is rare and majority of the patients may be asymptomatic. CMV spreads with close contact through salivary secretions or sexual contact. Intrauterine infection can cause clinical disease presenting either as cytomegalic inclusion disease of newborn or fetal death. Primary infection in healthy adults is usually asymptomatic whereas in an immunocompromised host like AIDS patient or transplant recipient, CMV can cause fatal infection. An infected person can carry the virus for life.

CMV can cause erosive or ulcerative lesions at any location in GI tract from mouth to rectum and the symptoms depend on which part is involved.³ Wreghitt et al studied CMV manifestations in 124 immunocompetent patients and reported that they most commonly presented with abnormal LFT findings, malaise, sweats, pyrexia, muscle aches, lymphadenopathy, jaundice and arthralgia.⁴ CMV associated stomach and colon infection are more

common as compared to esophageal infection. Lim et al has reported a similar case of CMV esophagitis in an immunocompetent patient successfully treated with ganciclovir. However, in the study, patient had multiple superficial mucosal ulcerations in the larynx and upper, mid and lower esophagus. Many studies however report ulcers in middle and distal part of esophagus. CMV esophagitis reactivation is known to occur in critically ill immunocompetent individuals.

It can present as ulcerative lesions in larynx.⁸ Weile et al has reported a case of severe CMV esophagitis after corticosteroid therapy in a patient with no other cause of immunodeficiency.⁹ Primary infection can be diagnosed by demonstration of antibody using serological tests like ELISA, IF, CF and IHA. Acyclovir is useful for prophylaxis but not for treatment. Ganciclovir and foscarnet are used in treatment of CMV disease in immunocompromised patient. Intravenous Ganciclovir at dose of 10-15 mg/kg per day for 3weeks is generally preferred for active disease.¹⁰

CONCLUSION

CMV esophagitis can occur in an immunocompetent individual though it is extremely uncommon. Antibody titers and histopathology helps in diagnosis. Early diagnosis will enable us to cure this condition. Excision of the cyst with appropriate course of intravenous antiviral results in cure. Though it is considered to be self-limiting and benign condition in immunocompetent individuals and role of antiviral remains to be established, this study highlights its role in effective treatment and cure.

Funding: No funding sources Conflict of interest: None declared Ethical approval: Not required

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Cite this article as: Anchan SV, Khialani A, Raj A. Cytomegalovirus esophagitis associated with arytenoid cyst in an immunocompetent host. Int J Otorhinolaryngol Head Neck Surg 2023;9:970-2.