



Atypical presentation of Cytomegalovirus endotheliitis: A case report

Journal:	<i>Ocular Immunology and Inflammation</i>
Manuscript ID:	NOII-2010-0084.R1
Manuscript Type:	Case Reports
Date Submitted by the Author:	n/a
Complete List of Authors:	Chiang, Chun-Chi; China Medical Univsesity Hospital, Ophthalmology
Keywords:	cytomegalovirus (CMV), endotheliitis, corneal edema, keratic precipitates (KPs), valganciclovior



1
2
3
4 **Atypical presentation of Cytomegalovirus endotheliitis: A case report**
5

6
7 Chun Chi Chiang, MD^{1,2}, Yi Yu Tsai, MD PhD¹
8
9

10 ¹Department of Ophthalmology, China Medical University Hospital, Taichung,
11

12
13 Taiwan
14

15
16 ²Institute of Medical Molecular Toxicology, Chung Shan Medical University,
17

18
19 Taichung, Taiwan
20
21

22
23
24
25
26 Correspondence to: Yi Yu Tsai, MD PhD, Department of Ophthalmology, China
27

28
29 Medical University Hospital, Taichung, Taiwan
30

31
32 Telephone: 886-4-22052121 ext. 1141
33

34
35 Fax: 886-4-22059265
36

37
38 e-mail address: elsa10019@yahoo.com.tw
39

40
41 The authors have no proprietary or financial interest in any material or device
42
43 mentioned.
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

Abstract

Purpose: We report an atypical case of cytomegalovirus (CMV) endotheliitis who presented with chronic corneal edema without keratic precipitates and intraocular pressure elevation.

Design: Case reports

Methods: A complete ophthalmologic examination was performed. PCR was used to amplify the DNA of HSV, VZV, and CMV in samples of the aqueous humor to rule out viral endotheliitis.

Results: Severe bullous keratopathy in the temporal part of the cornea, but there were no KPs nor elevated IOP noted through the whole course. PCR detected the DNA of CMV in an aqueous sample, and the corneal edema subsided with oral valganciclovir.

Conclusions: CMV endotheliitis may present as corneal edema alone without typical features such as coin-shape KPs or IOP elevation.

Introduction

There are more and more reports of corneal endotheliitis caused by cytomegalovirus (CMV) in immunocompetent hosts.¹⁻³ The most unique ocular features of corneal CMV endotheliitis are keratic precipitates (KPs) and elevated intraocular pressure (IOP).¹⁻³ Here we present an atypical case with chronic corneal edema and positive of CMV infection.

Case Reports

1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

A 74-year-old Taiwanese male had bullous keratopathy of unknown cause referred for keratoplasty. He presented with blurred vision in his right eye for 2 months. On examination, visual acuity was 6/200. Intraocular pressure was 14 mmHg. Temporal two thirds of cornea had epithelial and stromal edema, but there was no KPs **nor significant anterior chamber reaction.** (Fig. 1). **Corneal endothelial cell counts were unavailable due to severe corneal edema.** Herpetic keratitis was suspected. Topical steroids (Prednisolone 1 % QID) and oral valaciclovir treatment were given for 1 week. However, the corneal edema persisted without any improvement **in the following 2 weeks. Informed concern was obtained and an anterior chamber paracentesis was performed. Qualitative PCR analysis revealed the presence of the cytomegalovirus genome but negative HSV and VZV DNA in the aqueous confirming the diagnosis of CMV keratitis.** Oral valganciclovir was started and 2 weeks later the corneal edema dramatically subsided. (Fig. 2) Six weeks after valganciclovir treatment, the endotheliitis became quiescent and keratoplasty was no longer necessary. Endotheliitis remained quiescent for 4 months and valganciclovir was discontinued. (Fig. 3) In the all treatment period, there were no KPs nor elevated IOP found. **However, the corneal endothelial cell counts were only 529 cells/mm² left, with a significant loss compared with the normal**

1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

fellow eye 2432 cells/mm².

For Peer Review Only

Discussion

In case with chronic corneal edema but no IOP elevation nor KPs development, CMV endotheliitis still should be considered as a possible etiology. In the article of Anshu et al, there were 2 presumed CMV endotheliitis patients presented with glaucoma but without KPs.⁴ Koizumi reported eight CMV endotheliitis patients, though all patients had KPs, there were two patients presented with normal IOP.² Hence, KPs and IOP elevation may not always coexist in CMV endotheliitis.

Miyanaga et al reported that the CMV might affect at the endothelium and cause endothelial cell loss.⁵ In our case, the corneal endothelial cell counts were significant loss 6 months later, which was compatible with their results.

Because the CMV endotheliitis may mimic as Fuchs' heterochromic iridocyclitis(FHI), Posner Schlossman syndrome, endotheliitis in patients with presumed HSV/HZV keratouveitis, and pseudophakic bullous keratopathy,² but this patient was no iris anomaly nor depigmentation, no corneal guttata, no IOP elevation, no intraocular surgery before. The viral etiology was most likely and PCR confirmed the CMV infection. However, in this kind of atypical presentation of CMV endotheliitis, it might be overlook and misdiagnose. If keratoplasty were performed in those atypical corneal CMV infection cases, the CMV endotheliitis might still recur after surgery without antiviral therapy.

1
2
3
4 In summary, CMV infection should be suspected in any chronic corneal edema
5
6
7 patients even without KPs and elevated IOP. Aqueous PCR analysis can confirm the
8
9
10 diagnosis and avoid unnecessary surgery.
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

For Peer Review Only

References

1. Chee SP, Bacsal K, Jap A, et al. Endotheliitis Associated with Evidence of Cytomegalovirus Infection. *Ophthalmology*. 2007;114(4):798-803.
2. Koizumi N, Suzuki T, Uno T, et al. Cytomegalovirus as an etiologic factor in corneal endotheliitis. *Ophthalmology*. 2008;115(2):292-297.
3. van Boxtel LA, van der Lelij A, van der Meer J, et al. Cytomegalovirus as a cause of anterior uveitis in immunocompetent patients. *Ophthalmology*. 2007;114(7):1358-62.
4. Anshu A, Chee SP, Mehta JS, et al. Cytomegalovirus endotheliitis in Descemet's stripping endothelial keratoplasty. *Ophthalmology*. 2009;116(4):624-30.
5. Miyanaga M, Sugita S, Shimizu N, Morio T, Miyata K, Maruyama K, Kinoshita S, Mochizuki M. A significant association of viral loads with corneal endothelial cell damage in cytomegalovirus anterior uveitis. *Br J Ophthalmol*. 2010;94(3):336-40.

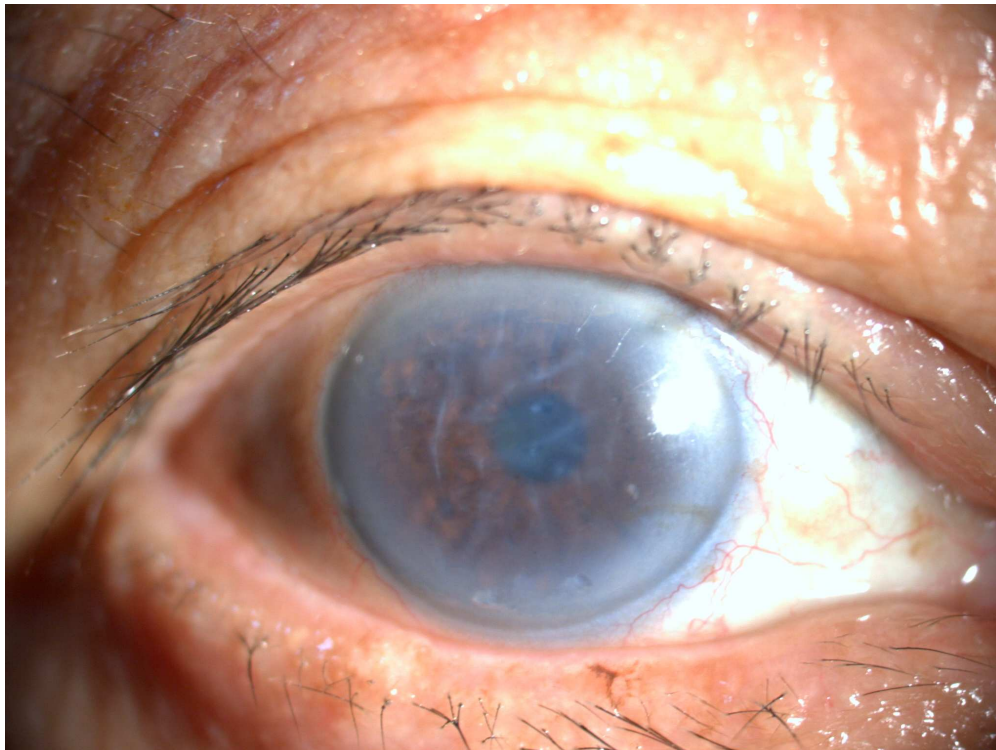
Figure legends:

Fig 1. Diffuse corneal edema with subepithelial bullae involving the temporal two thirds of the cornea. There was no visible keratic precipitates.

Fig 2. Considerable decrease in swelling after 2 weeks of valganciclovir treatment

Fig 3. Corneal edema complete resolution after 16 weeks of valganciclovir treatment

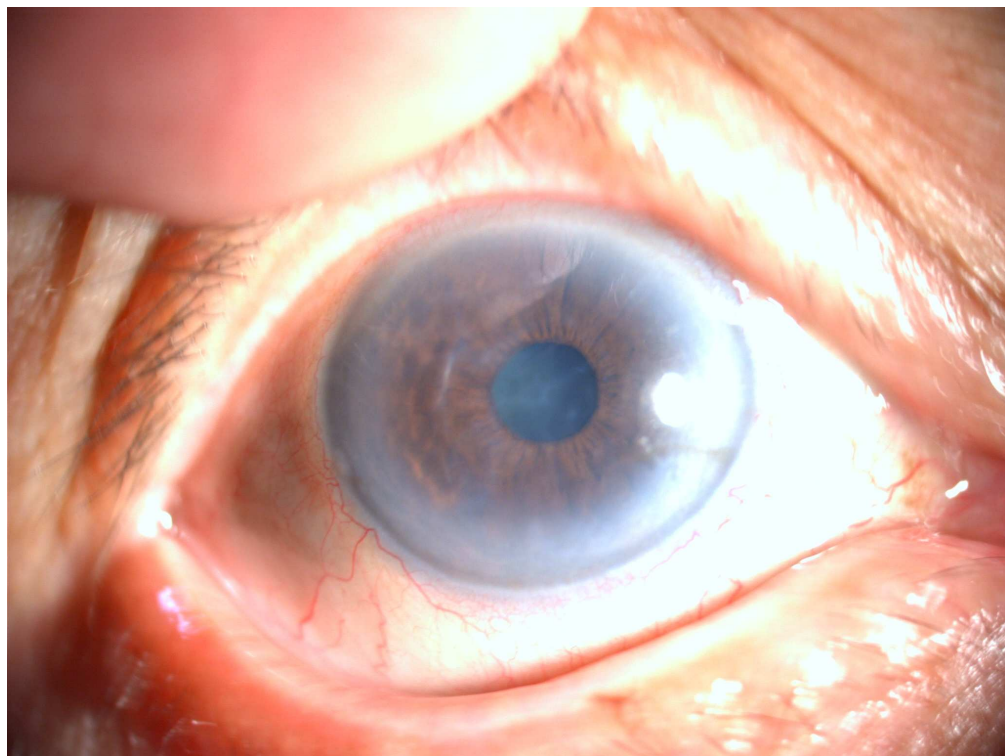
1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60



722x541mm (72 x 72 DPI)

iew Only

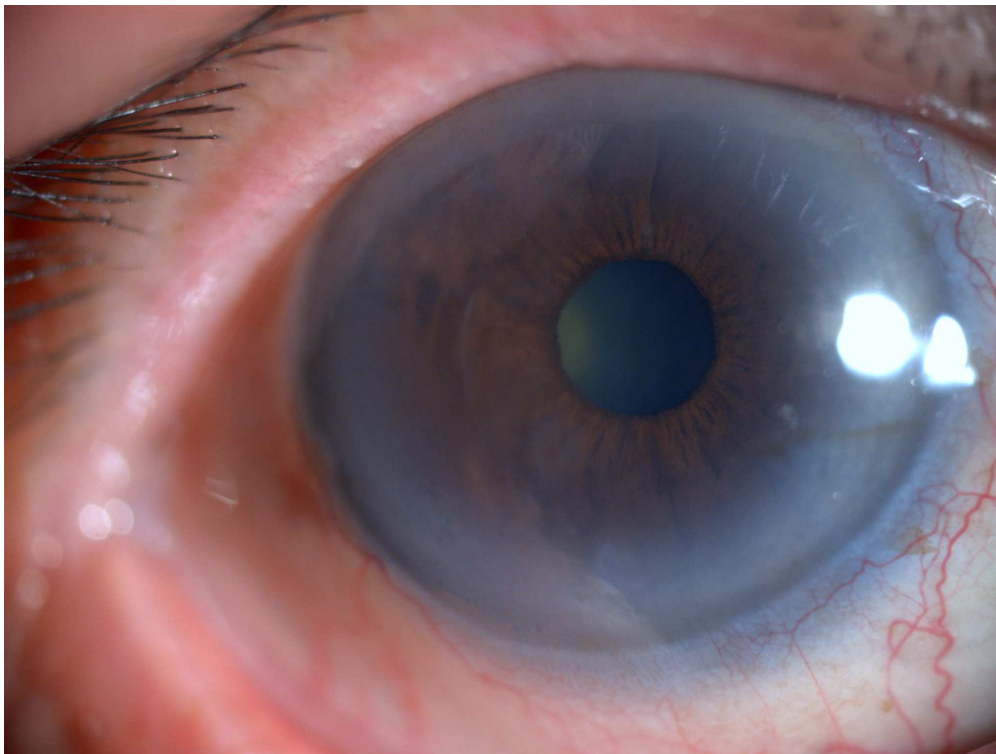
1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60



722x541mm (72 x 72 DPI)

ew Only

1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60



722x541mm (72 x 72 DPI)

ew Only