Kawasaki Disease Concurrent with Visual Conversion Disorder in Child: A Case Report and Review of The Literature

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Case Report

Keywords:

Posted Date: February 25th, 2022

DOI: https://doi.org/10.21203/rs.3.rs-1343819/v1

Abstract

Kawasaki disease (KD) is a self-limited vasculitis of medium- and small-sized vessels that occurs mainly in children between 5 months and 6 years of age. Conversion disorder is a type of somatoform disorder which is rare occurred in children under six years old. KD accompanied with conversion disorder has never been reported in the literature. We report a 4-year, 6-month-old female KD patient presenting with visual conversion disorder. She presented with classic KD symptoms, including high fever, rash, strawberry tongue and conjunctival and pharyngeal congestion. In addition, the patient displayed transient right vision loss during KD, while her ophthalmology and laboratory tests showed that there were no abnormalities that would result in vision loss; thus, psychogenic factors (conversion disorder) were considered after a simple test to confirm the functional vision loss of the right eye. The case could provides a differential diagnosis for clinical Kawasaki disease combined with vision disorder.

Introduction

Kawasaki disease (KD), also known as mucocutaneous lymph node syndrome, is a self-limited vasculitis that predominantly occurs in Asian children, and the onset age of KD is between 6 months and 5 years(1-3). KD is characterized by a few classic findings, including high fever, strawberry tongue, skin rash, bilateral conjunctival congestion, polymorphous exanthema, and extremity changes(4). Ocular damage is a common manifestation of KD, such as nonexudative conjunctivitis and anterior uveitis. However, functional vision loss is an unusual ocular finding in KD.

Conversion disorder, namely, functional neurological symptom disorder, is a type of somatoform disorder and is thought to be a complicated and puzzling disease by psychologists and physicians. Conversion disorder has long been recognized as a cause of subjective visual field defects(5). However, vision loss is much less common symptom of conversion disorder in young children(6, 7).

To date, Kawasaki disease concurrent with visual conversion disorder has not been reported. In this review, we report a preschool child with Kawasaki disease accompanied by visual conversion disorder. It provides a differential diagnosis for classic Kawasaki disease combined with organic vision disorders.

Report Of A Case

A previously healthy 4-year, 6-month-old (4y6m) female was transferred to our hospital because of a one-week history of high fever (39·1 °C) accompanied by abdominal pain and skin rash. Symptoms started 7 days before, a physical examination revealed rash, superficial lymph nodes, conjunctival and pharyngeal congestion, chapped lips, strawberry tongue, and chest auscultation with rough breath sounds. She had no sick contacts, recent travel, or prior illicit drug use. Admission laboratory blood tests revealed the following: white blood cells, 9·3×10^9/L; red blood cell count, 4·53×10^12/L; hemoglobin, 124·0 g/L; platelet count, 268×10^9/L; neutrophils, 81·31%; and lymphocytes, 13·22%. Urine examination showed the following: white blood cells, 20/μl; white blood cell, 4/HPF; ketone, 2+; urine specific gravity, 1·020; erythrocyte sedimentation rate, 18 mm/h; and CRP, 73·6 mg/L. She was treated with acyclovir, cefotiam, and vitamin B6 but did not improve and developed maculopapular diffuse rush on the day following hospitalization (2nd day after the

onset of fever). Then, she was given 7·5 g/day γ-globulin for 2 days, while the fever was still intermittent (39·3 °C). Therefore, she was transferred to our hospital for further evaluation.

On admission to our hospital, the laboratory findings were as follows: white blood cells, 15·7×10^9/L; red blood cell count, 3·64×10^12/L; hemoglobin, 101·0 g/L; platelet count, 268×10^9/L; neutrophils, 84·7%; lymphocytes, 9·2%; and neutrophil count, 13·3×10^9/L. No dilatation of coronary arteries was found. Neck ultrasonography showed enlarged lymph nodes on the right side of the neck. All other findings in the physical examination were unremarkable. The patient was diagnosed with KD and treated with γ-globulin 2 g/kg/day and aspirin 600 mg per day. The diffuse skin rash was diminished accompanied by periungual peeling of fingers (Figure 1). The peak of fever decreased on the 2nd day after admission, and the laboratory results are summarized in Table 1. On Day 3, the patient had a short and sudden loss of right vision with no cause that lasted for 20 min and self-recovered after the attack. Ophthalmology tests revealed unremarkable abnormalities in the fundus (see Figure 2), ophthalmic artery color Doppler and pupillary reaction. Although bilateral visual field examination of the patient demonstrated scattered blind spots, the confidence of the results was low considering the ability to understand based on the age of the patient (see Figure 2). However, the loss of right vision reappeared on the morning of Day 4 for 10 min, and examinations continued to show no abnormalities. Then, the vision loss was self-recovered again. The ophthalmologist and neurologist suggested visual-evoked potentials (VEPs), brain MRI+MRA and EEG, and neck color Doppler to explore the causes; all the imaging results demonstrated no abnormal findings, neurological diseases and carotid stenosis and optic neuritis were excluded. Salvia miltiorrhiza polyphenolate and tafluprost eye drops were given to the patient to improve circulation and reduce intraocular pressure. The loss of vision appeared 5 times intermittently over the next few days, and the longest loss lasted for 2 hours. The causes remained unknown. Consultation with a specialist occurred, and the possibility of conversion disorder was first considered; however, whether the vision loss was due to conversion disorder needed to be confirmed. On Day 8, there was an important finding that the patient's mother told us: the patient could paint normally with the shadow of the left eye during the period of the transient loss of right vision (Figure 3). The "painting test" was performed again during vision loss, and the results were the same as before. Based on the clinical findings, examinations and mother's description, the diagnosis of visual conversion disorder was made. In the next few days, the laboratory blood tests were measured again and are summarized in Table 1. The amount of aspirin was reduced to 75 mg per day. The loss of vision disappeared, and the fever was resolved.

Comment

Kawasaki disease is an acute nonspecific systemic vasculitis related to immune dysfunction that mostly occurs in East Asian infants and young children under 5 years old(1). KD patients also present with ocular disorders, and the most common forms of ocular dysfunction are nonexudative conjunctivitis and anterior uveitis. In this review, we report an unusual KD patient accompanied by vision loss due to conversion disorder.

Visual disorders are occasionally reported in KD as manifestations or accompanying diseases. To date, a total of 9 cases of Kawasaki disease with visual dysfunction have been reported(8-16) (see Table 2). The etiology, ocular manifestations, sex and age of these patients were analyzed. All of the patients had no

previous history of eye diseases. All patients presented with various degrees of monocular or binocular vision reduction or loss, with 30% of vision Lost. Among the organic factors contributing to visual disorders, the ratio of ocular to extraocular (cerebral vasculitis) disease was 8:1. Conjunctival congestion occurred in 77·8% (7/9) of patients and is one of the typical symptoms of Kawasaki disease. In addition, the ages of these patients ranged from 4–18 years old, and the ratio of males to females was 2:7. The pathogenesis of vision loss (VL) is mainly divided into two categories: organic factors and functional vision loss (FVL). Organic VL could be the result of ocular or neuropathic diseases, whereas the pathogenesis of functional vision loss (FVL), which is opposite to organic vision loss, often has some underlying psychological basis(17). Conversion disorder is one of the common causes of FVL(18). In the present patient, vision loss was functional and psychogenic, due to conversion disorder, unlike the above-described organic ocular disorders. To the best of our knowledge, KD with or accompanied by conversion disorder in children or adults has not been previously reported in the literature.

Conversion disorder, also called hysteria, may appear in the form of a physical or psychological disorder(19). Sensory symptoms, including altered, reduced, or absent skin sensation, vision, or hearing, are common manifestations of conversion disorder. Research has revealed that physical diseases, especially acute injury, could induce the onset of conversion disorder in one-third of patients(20). In our patient, the vision loss of the patient was sudden and nonorganic, which could not be explained by neurological or medical conditions, which supported the diagnostic criteria for conversion disorder(3). However, it is worth noting that the onset age of our patient was rare among those with conversion disorder. A study demonstrated that conversion disorder mostly occurs between the ages of 12 and 16 years (21-23), and the prevalence is higher in females than in males(24). Moreover, there are clear psychological inducers before the onset of the disease in most cases, such as family conflict, academic difficulties and some diseases(23, 25-27). In addition to KD, the characteristics of our patient could also be of great importance regarding contributions to the occurrence of visual conversion disorder. Although the specific pathogenesis of hysterical vision loss is not clear, we could suggest that the two-sided personality of the patient (i.e., shy in the hospital and lively in the family environment), persistent high fever resulting from KD, and the anxiety of her parents could have promoted the occurrence of conversion disorder. It has been suggested that the occurrence of symptoms might be an adaptive defense against anxiety in children(28).

Another important finding was that parents' advice could be useful to the diagnosis of conversion disorder in young children. Conversion disorders are uncommon in children and often lack specific markers, which makes their clinical assessment challenging, as they do not easily fit standardized clinical care guidelines(29). In our case, the mother of the patient conducted a simple test to help the doctor identify the functional vision loss in the child. The simple "painting test" was similar to the vertical prism dissociation test used in ophthalmology to identify functional monocular vision loss(17).

Conclusion

this case reminds us that psychogenic factors should be considered when organic diseases are excluded for unexplained ocular symptoms in children with KD. Our findings also provide a differential diagnosis for patients with KD with ocular damage and a warning for pediatricians regarding the early onset age of

conversion disorder in children. In addition, we suggest that the combination of pediatricians and parents will be more effective in the diagnosis and treatment of childhood diseases.

Abbreviations

KD
Kawasaki disease
CRP
C reactive protein
VEPs
visual-evoked potentials
VL
vision loss

Declarations

Acknowledgement

We would like to thank the patient's mother for providing permission to share the patient's information.

Authors' contributions

JJZ, and JML designed the research proposal. JML, YYZ and HQZ analyzed the clinical data. YYZ and JML wrote the paper. All authors read and approved final draft of the manuscript.

Funding

This work was supported by the Health Commission of Henan Province (no.Wjlx2020049).

Availability of data and materials

The data that support the findings of this study are available from the corresponding author upon reasonable request.

Ethics approval and consent to participate

Parental informed consent for publication was obtained. This study was approved by the Ethics Committee of The First Affiliated Hospital of Zhengzhou University

Consent for publication

Written informed consent was obtained from the patient's legal guardians for publication of this case report and any accompanying images.

Competing interests

The authors declare no conflict of interest.

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Tables

Table 1 The laboratory results

Test		2nd day	6th day	13th day	Reference range
Hematology	WBC	12·1×10^9/L	11·06×10^9/L	6·59×10^9/L	3·5-9·5×10^9/L
	RBC	3·44×10^12/L	3·55×10^12/L	3·6×10^12/L	3·5-5·5×10^12/L
	Hb	96 g/L	99 g/L	111 g/L	115–150 g/L
	PLT	364×10^9/L	796×10^9/L	538×10^9/L	125–350×10^9/L
	neutrophils	75·8%	79·3%	59·2%	40%–75%
	lymphocytes	14·1%	10·4%	27·5%	20%–50%
	ESR		85 mm/h	50	0–20 mm/h
Coagulation	APTT	43·4	36·2	36·5	26–40 s
	D-dimer	0·66 mg/L	0·21	0·11	<0·3 mg/L
	Fib		3·97	3·43	2–4 s
	TT	5·5 mg/L	16·1	15·4	10–18 s
Blood chemistry	GGT	76 U/L	49	29	0–58 U/L
	Albumin	26 g/L	34·5 g/L	36·6	32–52 g/L
	Globulin	46·2 g/L	44·2 g/L	41·6	20–35 g/L
	cholinesterase	3·2 KU/L		8·7	4–15 KU/L
	proBNP	3900 pg/ML	298	99·9	0–97·3 pg/mL
	TG	2·27 mmol/l	1·91 mmol/l	1·11	<1·7 mmol/l
	HDL	0·38 mmol/l	0·63 mmol/l	0·94	>0·91 mmol/l
	CRP	11·42 mg/L	3·28 mg/L		<3 mg/L
	PCT	0·254 ng/ml	0·12 ng/ml		
	IL	618·37 pg/ml	69·25 pg/ml		
	BNP		298		
ZD	CK	25 U/L	27	26	26–192 U/L
	CK-MB	24 U/L	26·9 U/L	22·6	0–25 U/L

WBC, white blood cell; RBC, red blood cell; Hb, hemoglobin; PLT, platelets; PT, prothrombin time; APTT, activated partial thromboplastin time; FIB, fibrinogen; Alb, albumin; ALT, alanine transaminase; AST, aspartate transaminase; g-GGT, gamma-glutamyl transferase; ESR, erythrocyte sedimentation rate.

Table 2 9 cases of Kawasaki disease with visual dysfunction

Case	Sex	Age	Side	Vision	Intraocular pressure	Conjunctival congestion	Pathology (functional or psychogenic)	fundus
1(8)	f	18	Both	reduction	-	yes	Anterior uveitis	normal
2(9)	m	11	both	reduction	10/12 mmhg	yes	Disciform keratitis	-
3(10)	m	6y9m	both	-	-	yes	Transient unilateral oculomotor palsy	-
4(11)	f	12	both	reduction	-	-	Global inflammatory involvement of ocular segments	-
5(12)	f	7	both	reduction	-	yes	Crystalline-like keratopathy	normal
6(13)	f	12	both	reduction	normal	yes	Crystalline keratopathy	normal
7(14)	f	4	both	loss	-	-	Cerebral vasculitis	Slight peripheral retinal fibrosis
8(15)	f	4	both	reduction	normal	yes	Retinal vasculitis	bilateral pale optic and discs extensive sheathing and sclerosis of all retinal vessels
9(16)	f	9	right	loss	-	yes	Ophthalmic artery obstruction	-
present	f	5	right	loss	normal	no	Conversion disorder	normal

Figures

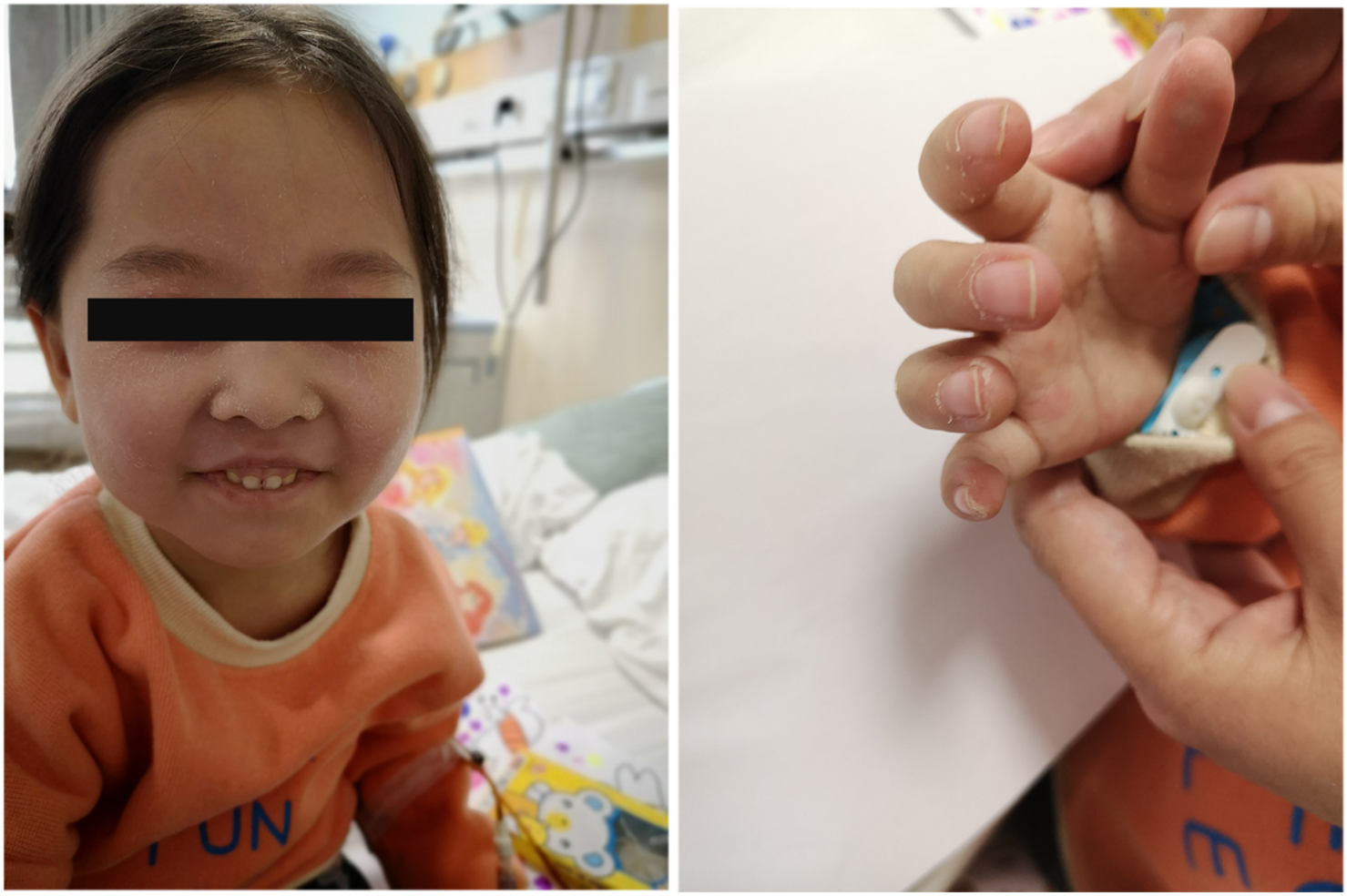

Figure 1

Classic symptoms of KD: Periungual peeling of the face, fingers and toes.

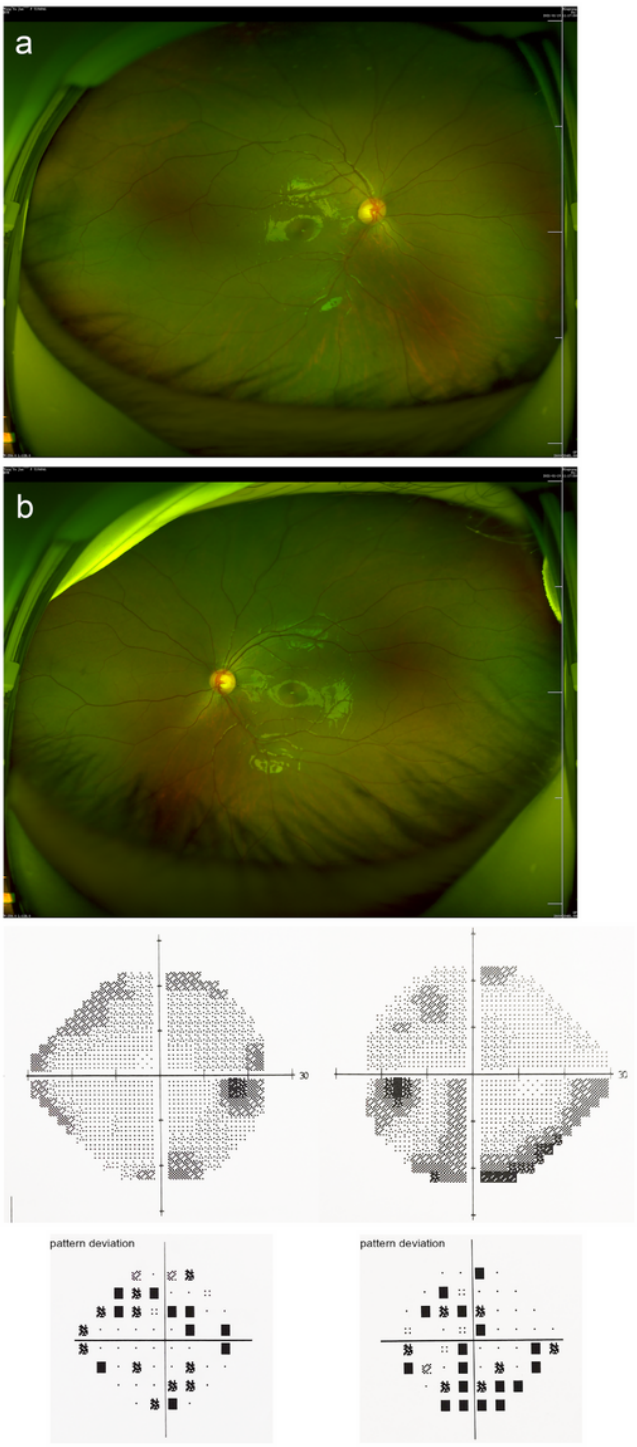

Figure 2

(A) Fundus photograph of the patient: right (a) and left (b);

(B) Bilateral visual field examination of the patient.

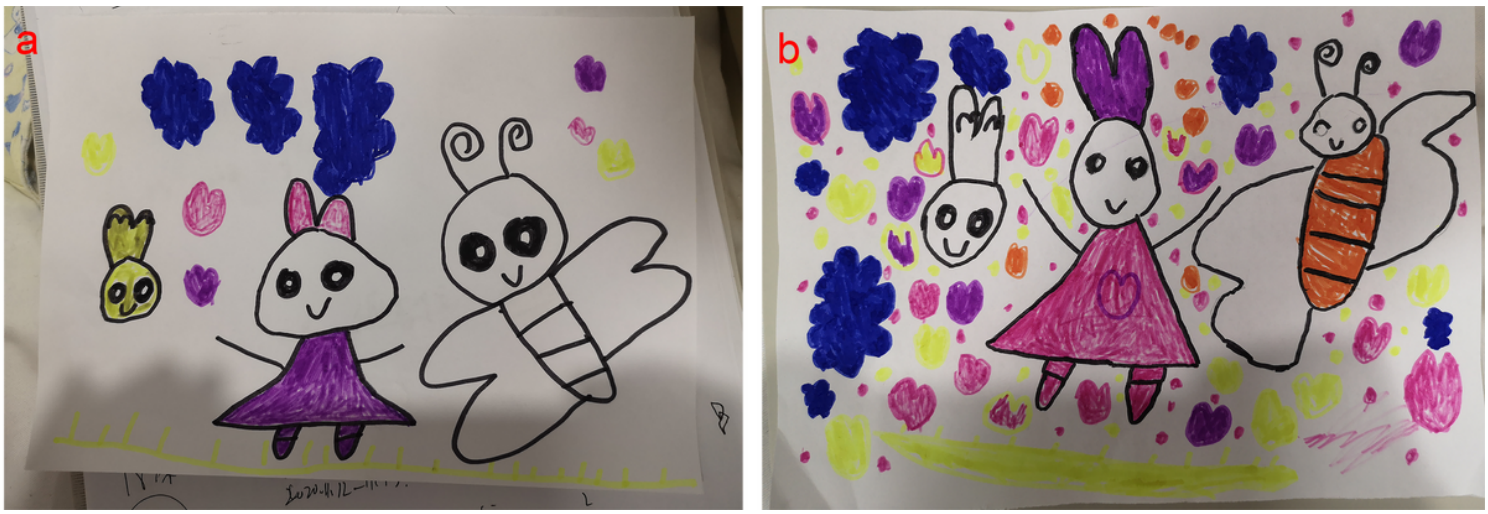

Figure 3

Simple test for functional vision loss. a: unfilled painting; b: filled painting by the patient with the shadow of the left eye.