Urrets-Zavalia syndrome with intraocular metallic foreign body: thinking caused by a case report

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Abstract: This paper reports a rare case of pupil dilatation and ankylosis of the left eye of a 22-year-old male patient for half a month. The patient felt foreign body sensation, conjunctival congestion and a mild visual loss of the left eye. The right eye is normal. After a series of examination, the reason of the patient's signs and symptoms were determined by intraocular metallic foreign body in his left eye. The magnetic resonance imaging (MRI) which shouldn't be performed actually, didn't reveal the object clearly, but the subsequent CT examination confirmed the high-density foreign body in the left eye. By asking about the history of trauma again, obviously, the pupil dilatation and ankylosis of the left eye was from the intraocular metallic foreign body. Finally, the patient was diagnosed as Urrets-ZavaIia syndrome which is a rare disease in clinic. Then the patient was recommended to do eye surgery. After surgery, his left eye recovered well. In conclusion, our case reports a rare disease of Urrets-ZavaIia syndrome caused by intraocular metallic foreign body. It also demonstrates that the detailed history and complete examinations should be obtained in patients to diagnosis such rare disease.

Keywords: Urrets-ZavaIia syndrome; metallic foreign body; pupil dilatation; pupil ankylosis; case report

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Introduction

Urrets-Zavalia syndrome is a rare disease characterized by fixed dilated pupil (1). It is usually caused by penetrating keratoplasty (PK), cataract surgery, phakic posterior chamber intraocular lens, argon laser peripheral iridoplasty, and so on (2-5). Urrets-ZavaIia syndrome is easy to be misdiagnosed or missed in clinic because of few positive signs (6). To the best of our knowledge, there are few reports of Urrets-ZavaIia syndrome caused by intraocular metallic foreign body. Here, we report a case of Urrets-ZavaIia syndrome caused by an intraocular metallic foreign body which was discovered accidentally. The patient has typical symptom of pupil dilatation and ankylosis of the left eye, but the history of the patient complained is unclear at first. Above all, the case is easily to be misdiagnosed with retinal or visual pathway diseases.

We present the following article/case in accordance with the CARE reporting checklist (available at http://dx.doi.org/10.21037/acr-19-157).

Case presentation

A 22-year-old male patient, he himself suspected of being hit by something in the left eye half a month before he visited hospital. He felt foreign body sensation and conjunctival congestion of the left eye in the first few days. The right eye was normal. He did not see the doctor for treatment at first, and just used chloramphenicol eye drops by himself.

Half a month later, the patient felt mild decrease of vision of his left eye, then he went to the local hospital. After the first examination of an ophthalmologist, he was found of mydriasis of the left eye (*Figure 1*), and light reflection is slow. The vision of the left eye was 16/20 compared the

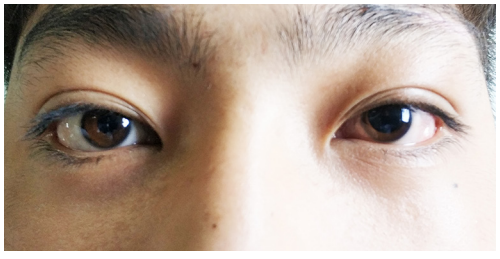

Figure 1 The pupil of the left eye is circle and centered with a 7mm-diameter, compared the right eye 3-mm diameter.

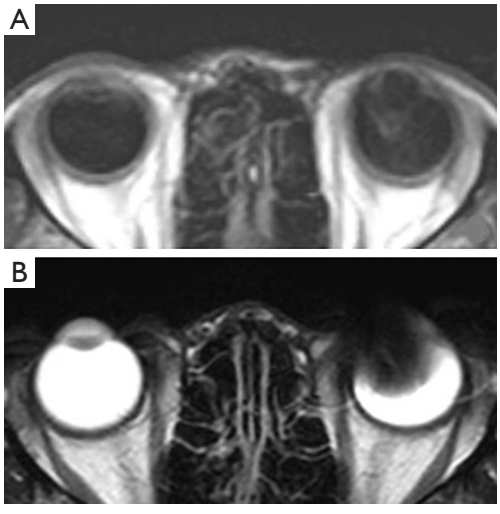

Figure 2 MRI image shows that some artifacts and abnormal signals in the left eye. No clear foreign body in the left eye is displayed: (A) T1-weighted image shows abnormal signals and structural disorder in the left eye; (B) T2-weighted image shows some artifacts of the left eye. MRI, magnetic resonance imaging.

right eye 20/20. The vitreous humor was slightly turbid of the left eye. The intraocular pressure was 15 mmHg in each eye. No other abnormalities were found. There was no medical, family, and psycho-social history including relevant genetic information about this patient. Because of few other positive signs, the ophthalmologist suggested the patient go to the neurology department for further examination. In the neurology department, the patient had brain magnetic resonance imaging (MRI) examination which shouldn't be performed actually. The MRI showed some artifacts and abnormal signals in the left eye. But MRI didn't reveal any clear foreign body in the left eye (*Figure 2*). Because of the abnormal images in MRI, the patient was suggested back to the ophthalmology department to check

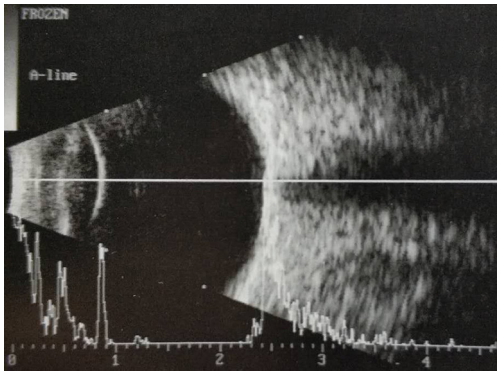

Figure 3 There is no abnormality in the A/B-ultrasound examination of the left eye.

the left eye. Then the ophthalmologist examined the patient again. It was still no other positive findings in the ophthalmology department. There was no swelling and congestion in both eyes. It was normal about eye movement and eye size. In the left eye, the corneal was transparency. It was normal about anterior chamber (aqueous humor). The iris had no pigment loss and rupture, the pupil was circular and centered with a 7-mm-diameter, but direct and indirect light reflection was slow. The lens was transparent. The vitreous humor was slightly turbid and the fundus was even clear. And there was no positive sign in the patient's right eye. Even in the A/B-ultrasound examination, there was no abnormality in both eyes, which didn't show any clear abnormal image (*Figure 3*).

After that, the patient underwent the orbital CT examination. Then a high-density foreign body like metal in the left eye was detected by CT scan (*Figure 4*). Obviously, the pupil dilatation and ankylosis of the left eye was from the intraocular metallic foreign body. At that moment, the ophthalmologist considered a rare disease in clinic: Urrets-Zavalia syndrome, because the patient had a typical sign of pupil dilatation and ankylosis of the left eye which was the characteristic of Urrets-Zavalia syndrome. Then the patient was recommended for eye surgery. After surgery, the foreign body was removed and determined to be iron (2.5 mm × 2 mm × 2 mm). Through a few days of postoperative treatment, the patient's left eye recovered well.

For some reason, the patient was followed up for only 3 months after surgery. At the last examination, we found that the pupil of the left eye had not shrunk and the light reflex had not return to normal, but the corrected visual acuity returned to the level of 16/20. *Figure 5* is the timeline of the patient's medical experience.

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

Discussion

This case is a rare disease called Urrets-ZavaIia syndrome. Urrets-ZavaIia syndrome is defined as an irreversible pupil stiffness after application or absence of a mydriatic agent (1). It is first proposed by Urrets-Zavalia, who described six cases of atrophic and mydriatic pupil associated with secondary glaucoma following PK in patients with keratoconus (7,8). Other findings of Urrets-ZavaIia syndrome are subsequently reported, including peripheral synechiae, posterior subcapsular opacity, iris ectropion, pigmentary dispersion, and glaukomflecken (1,3,5,9,10). Many factors can cause Urrets-ZavaIia syndrome, it is reported that most cases are deep anterior lamellar keratoplasty (1), PK (2), cataract surgery (3), phakic intraocular lens implant (4), argon laser peripheral iridoplasty (5), trabeculectomy (9), and so on. However, its etiology has not yet reached a conclusion. Although Urrets-ZavaIia syndrome is mostly caused by intraocular surgery, but in our case, there is no history of intraocular surgery and any other evidence. So, it is credible that the suspected trauma is the cause of Urrets-ZavaIia syndrome. It is even rare in clinical practice.

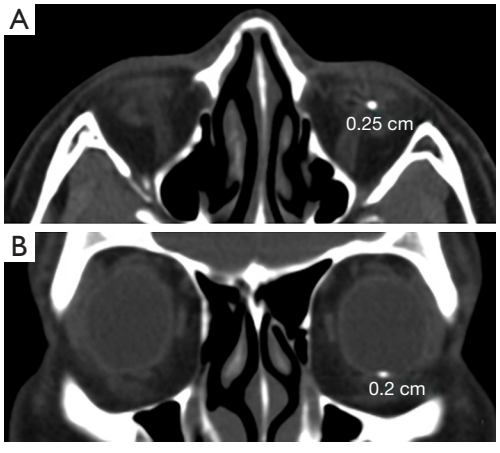

Figure 4 CT scan clearly shows the size and location of the high-density foreign body. (A) The horizontal CT scanning shows that the maximum diameter of the foreign body is 2.5 mm; (B) the coronal CT scan shows a high-density foreign body locates in the vitreous cavity of the left eye. CT, computed tomography.

In this case, the patient had foreign body sensation and conjunctival congestion of the left eye at the first few days. The patient said he suspected being hit by something in the left eye half a month ago. If the ophthalmologist raised his guard and asked more details about the history of trauma, it may arouse to suspect the possibility of an intraocular foreign body. If so, a direct CT scan may be performed earlier. But the ophthalmologist didn't notice the patient's history of suspicious trauma. The first ophthalmic examination only contained visual acuity, intraocular pressure, slit lamp and direct ophthalmoscope examination,

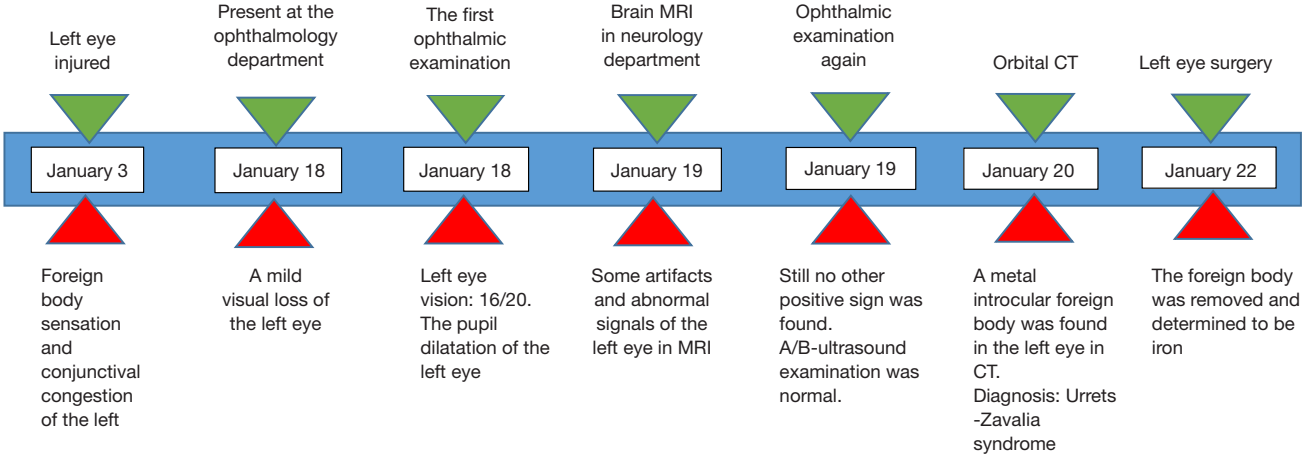

Figure 5 Timeline.

without further examinations. The result of the first ophthalmic examination only revealed the mild loss of vision, the pupil dilatation and the slightly turbid vitreous humor of the left eye. The intraocular lesion was not considered until some abnormality images was detected by MRI in the neurology department. Dramatically, the patient incompletely remembered the matter of ocular trauma. He wasn't very clear about the eye injury, because he signed the questionnaire of no metallic objects in his body before he did the MRI. So that after a further examination, the final diagnosis was determined by CT scan. Then the ophthalmologist thought of the disease, Urrets-ZavaIia syndrome. The pupil stiffness might be caused by damage to the eye tissue by metal foreign body and inflammation around it. It was possible that foreign matter entered the eyeball through the anterior sclera.

There are currently no specific treatments for diffuse pupillary atrophy. Corneal tattooing, specialty contact lenses, and artificial iris implants are options for glare reduction and appearance improvement. In focal atrophy, miotics such as guanethidine and pilocarpine have demonstrated efficacy in treating Urrets-ZavaIia syndrome and attenuating mydriasis (11). The limitation of collecting and reporting this case is that we lacked some postoperative data because the patients wasn't followed up in the hospital as required.

In conclusion, we report a rare disease of Urrets-ZavaIia syndrome caused by intraocular metallic foreign body. It also demonstrates that the detailed history should be obtained in patients to diagnosis. And complete examination is also essential to help diagnosis especially for such rare disease.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at http://dx.doi.org/10.21037/acr-19-157

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at http://dx.doi.org/10.21037/acr-19-157). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

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