



ISSN- 0975-7066 Vol 15, Issue 2, 2023

Review Article

A REVIEW ON POLYCORIA

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Received: 05 Jan 2023, Revised and Accepted: 15 Feb 2023

ABSTRACT

The presence of multiple iris holes is most commonly called pseudopolycoria, because only the central pupil has a sphincter muscle and can constrict. Iris holes without muscle tissue arise as congenital defects or develop from ocular trauma, inflammation, or degeneration and are thus not considered to be true accessory pupils. Multiple pupils, also known as true polycoria, are distinguished by the presence of a sphincter muscle around each iris hole, permitting synchronous constriction and dilation of the 2 pupils. One presumed mechanism for polycoria is a snaring or pinching off from the margin of another pupil. This patient had a small, oval-shaped accessory pupil, which may have separated from the temporal margin of the central pupil, leaving a thin bridge of no sphincter connecting tissue. The outlook for polycoria is generally good. You may not require any treatment if your visual impairment is minimal and doesn't interfere with your daily life. However, if treatment is needed, pupilloplasty has so far shown positive results. If you have polycoria, it's important to have regular check-ups with an eye doctor to monitor your vision and any changes your eyes may have. Having your eye checked regularly is also beneficial for your eyesight as a whole.

Keywords: Polycoria, Ophthalmopathology

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INTRODUCTION

Polycoria is an ophthalmopathology in which two or more pupilsare located in the iris. And also the pupil tend to be smaller than normal and separated by individual segments of iris. It is a pathological condition of the eye characterized by more than one pupillary opening in the iris. It may be congenital [1] or result from and visualization. This can mean less light enters your eye, which can dim your vision you may also have difficulty in focusing because the pupil's aren't working effectively. Direct cause of polycoria is not known. Polycoria may be caused by problems with eye development before birth. Polycoria affects the pupils and can occur in both or just one eye. A patient may not know they have polycoria until they go to an eye doctor for a comprehensive eye exam. During the visit, the eye doctor will likely perform a slit-lamp exam, which uses a special light and microscope to examine the front and back of the eyes. This exam also allows the eye doctor to see any abnormalities [2] in the pupils. The use of operational tactics (iridoplasty) is recommended. If there are contraindications to surgical treatment, contact lenses are used to eliminate a cosmentic defect and correct visual dysfunction.

Polycoria is of two type's True polycoria and Pseudo polycoria or false polycoria. True polycoria is extremely rare and is defined by an additional pupil surrounded by an intact sphincter muscle. Pseudo polycoria is characterized by a full-thickness iris defect that lacks a surrounding sphincter muscle. Because these pupillary defects lack a sphincter, when the primary pupil dilates, the accessory defects undergo reflexive constriction. The bulk of iris tumors can be diagnosed with clinical or historical criteria [3] without the need for cytologic or pathologic verification. In the case, that observation may be dangerous or a diagnosis cannot be solidified based on clinical information and historical signs, other methods of verification can be used.

Biopsy techniqueis selected based on the tumor location, size, frability, feeder or intrinsic vessels and the risk of potential scattering of tumor onto the iris surface or anterior chamber angle. Biopsy techniques include fine-needle [4] aspiration biopsy, surgical iridiocyclectomy and transcorneal tumor biopsy. We will discuss pseudo-polycoria as a complication of iris biopsy, its greatest visual consequence, photophobia, anda non-surgical alternative for management.

Polycoria has also been linked to these two other uncommon eye conditions:

- Axenfeld-Rieger syndrome: Axenfeld-Rieger syndrome (ARS) is a genetic condition that affects the eyes. Patients who have this condition may also have false polycoria-extra holes in the iris that look like multiple pupils-or a single pupil that is off center.
- Iridocorneal endothelial syndrome: Iridocorneal endothelial (ICE) syndrome is an eye disorder that can leads to distortion [5] of the iris and the pupil, including polycoria. Patients with this condition also often develop secondary glaucoma. ICE syndrome is more common in women and patients 20 to 50 y old.

Polycoria is extremely rare and other conditions are frequently mistaken of it. Polycoria is often congenital however not diagnosed until adulthood. No matter how absurd this thing sound's its real. General cause of polycoria is not known, but there are some other eye conditions that are in association with polycoria. These include (although not often) polarcataracts glaucoma, abnormally long eyelashes, abnormal eye development and poor vision. There have been cases diagnosed from age 3 to adulthood. The 2 kinds of polycoria are true polycoria and false or pseudopolycoria [6]. There are no known or proposed trends in the occurrences of polycoria based on geographical location, age, gender, or season.



Fig. 1: Eye with polycoria

History

Since true polycoria and pseudopolycoria are so rare, there is not much history on the disorders in the tradition of modern western $\,$

medicine; according to an article published in 2002, there have only been 2 cases of true polycoria since 1966. Early Chinese history names multiple legendary fig. as having double pupils, which as a result was, led to a beliefs that a child born two double pupils were destined to be a great king or sage. Polycoria is one of the manifestations of Axenfeld-Rieger syndrome, perforated iris defects are diagnosed with a frequency of 1:200, 000. There are about 35, 000 people with this nosology in the world. Polycoria occurs with the same frequency among males and females.

Xiang Yu, Hegemon of Chu, blood brother and later arch nemesis of Liu Bang, the founding emperor of the Han dynasty, was given great esteem due to his prowess in combat and auspicious double pupil. After overthrowing the Qin dynasty together, he was named Hegemon of the loose series of kingdoms he created in its wake, while he gave Liu Bang the remote province of Han. The civil war that followed called the Chu-Han contention [7], ending with a Han victory and a legendary last stand by the Hegemon. Polycoria is a congenital malformation of the iris, which is detected in the first year of a child's life. Isolated cases of acquired pathology in mature persons are described. There are no statistics on the overall prevalence of the disease. Geographical features of the spread of the disease are not observed.

Classification

True polycoria

True polycoria is when you have 2 or more pupils in the same eye. It is an extra pupil that tends to be reactive to light and medication. To be considered true polycoria the extra pupil and the principle pupil must dilate and contract simultaneously with triggers such as light and administered drug. The extraneous pupil is 2.5 mm away from the principal pupil. There isanintactsphincter muscle [8], which contracts and dilates the pupil. The prevalence of true polycoria is minimal. In an eye without polycoria, the sphincter muscle is a part of the iris that functions to constrict and dilate the pupil. The patient

with true polycoria experiences handicapped vision as well as stimulation of the retina in response to bright lights. It is said that the term true polycoria is overused and used correctly when addressing congenital deformation of the iris. You will have two or more separate pupils [9] in one eye. Each pupil will have its own intact sphincter muscle. Eachpupil will individually constrict.

Pseudo polycoria

Pseudopolycoria or falsepolycoria is less rare then true polycoria but looks similar to it in the sense that people with pseudopolycoria seems to have two or more pupils in one eye. But, in this case, the pupils don't have separate sphincter muscles [10] and can't act independently. These extra pupils are nothing but the holes in your iris. As this condition is just an imperfection of the iris, people with pseudopolycoria don't experience any vision problems. False or pseudopolycoriais a Hereditary ocular disease. Pseudopolycoria or false polycoria is less rare than true polycoria. Pseudopolycoria is still very uncommon. In these cases of pseudopolycoria that differentiates there is a 'passive constriction' that differentiates [11] the extrapupil from the true pupil during constriction and dilation. The extra pupil in pseudopolycoria is different than the extra pupil in true polycoria because it shows defects that are independent of the sphincter muscles. It is often associated with sickle syndrome, posterior polymorphous dystrophy and juvenile glaucoma.

It consists of splitting of the iris that is not contingent with the sphincter muscles at the root of theiris. In the pseudopolycoria, the holes in your irislook like additional pupils. These holesare usually just a defect of the iris and do not cause any issues with your vision. Pseudopolycoria is distinguished from true polycoria by the passive constriction of the accessory pupil when the true pupil is dilated and is characteristic of essential iris atrophy that can be associated with sickle syndrome, polymorphous dystrophy [12], and juvenile glaucoma. It occurs when greater than one pupil appears to be present but active dilatation of true pupil leads to passive constriction of the adjacent falseor accessory pupil.

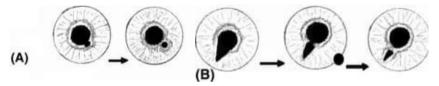


Fig. 2: (A) Snaring off a portion of the pupillary margin, (B) secondary closure of a closure

Symptoms

Polycoria may or may not cause vision issues. Symptoms of polycoria are usually a product of having more than one set of iris muscles. The iris is the colored ring of muscle around eachpupil; it controls how much light is allowed into the eye. In polycoria, the pupils are smaller than usual and each one of them has its iris. While all of the irises adjust light, it could dim your vision by reducing the amount of light [13] that enters your eye. Other signs and symptoms may include appearance of more than one "pupils"in one or both eyes, Blurry vision, Double vision(Diplopia), Glare sensitivity, Oblong shape of one or all additional pupils, A bridge of iris tissue between the pupils.

When a patient is diagnosed with polycoria, the signs [14] and symptoms shown are associated with ocular and adnexal growth abnormalities. The iris and the pupil become less effective. Signs can be present as a child; however the patient may be diagnosed later in their life time. This condition results in abnormal eye development affecting both eyes and just one. Upon gross examination the patient will typically have excessively long eyelashes. The iris become hypo plastic, making abnormally shaped pupils with prominent crypts. The crypts are little squiggly lines that radiate out around the pupil; with this condition, thick round or oval opening can beseen. On diagnosis, signs lead to symptoms where there is more thanone set of iris muscles, which controls the amount of light being brought into the eye. When the iris becomes deformed, it will disarrange the control coming in leading to blurred vision and finding it hard to

visually focus. Polar cataracts will also be present in this condition where a round, opaque malformation of distorted lens fibers is located in the central posterior [15] part of the lens showing disformality. Blindness may also be caused due to damage of the optic nerve from glaucoma.

Diagnostics

To verify the diagnosis [16], it is enough for an ophthalmologist to conduct an external examination. In most cases, 2-3 pupils are visualized, one of which is larger in size. The holes in the iris are rounded, rarely irregular in shape. Special ophthalmological examination includes:

- The study of pupillary reaction
- · Ultrasound of the eye
- A sample with mydriatics
- Biomicroscopy of the eye
- Perimetry
- Tonometry

Causes

The etiology of the disease is not fully understood. The main cause of polycoria are effect of teratogenic factors, Coloboma of the iris,

intrauterine infections, Iridocorneal endothelial syndrome, latrogenic effects, Traumatic injury, Axenfeld-Rienger syndrome. Some conditions that have been associated with polycoria are detached retina, Polar cataracts, Glaucoma, Abnormal development of the pupil margins, abnormal eye development. There is not any known direct mechanisms involved in the development of true polycoria or pseudopolycoria. There are some proposed ideas, one being that after the sphincter muscle [17] is fully formedand developed in the eve there is a severing of the pupillary margins leading to the distinction of the extra pupil and the principle pupil. Polycoria can also be caused any hole in one's iris to develop a sphincter muscle development. Another proposed theory about the cause of polycoria is intrauterine trauma, or postpartum iris trauma. If the development of the iris is hindered, the ectoderm of the eye (which forms the lens and corneal epithelium) may split, which could lead to pseudopolycoria.

Treatment

- Iris plastic surgery
- Surgical correction
- Symptomatic therapy

It may not be necessary to treat polycoria if it does not affect the patient's vision. Surgery is one treatment method for polycoria. But, since this problem is so rare, it could be pretty hard to determine the best form of treatment for this condition. However, an eye doctor recommends surgery for a patient with true polycoria if it is causing poor vision. The surgeon may treat polycoria by doing a pupilloplasty [18], an operation in which the surgeon cuts the iris "bridge "that separates the multiple pupils, joining them into a single pupils. In the case ofthe patient with true polycoria in Brazil, his doctor performed a pupilloplasty. The patient's vision improved in the affected (then surgically treated) eye, and he was happy with the results of the surgery. People with pseudopolycoria don't require any treatment as their vision isn't affected by this condition. Those who experience vision difficulties due to this condition must seek immediate medical treatment.

Polycoria has been linked to hereditary genetics and also associated with polar cataractsglaucoma, and retinal detachment. Not all cases are treated for this iris abnormality, but when casesare treated the only treatment is surgical procedure and life-long ocular monitoring that is highly recommended. Considerations for surgery are surgical correction, intraocular surgery, and/or reapproximation, as if it was being treated for glaucoma [19] or retinal detachment. Children under the age of 3, who do not seek surgical surgery, have with responded well mitotic drops that contained mydriatic/cycloplegic, allowing the separate eye sphincters to dilate and constrict together. This allows improved balance of uncorrected visual acuity. A 1-mm limbal incision would be made; a spatula in inserted through the side to elevate the iris tissue would be cut using viscoelastic material. Finally, the limbal incisions were closed with stromal hydration, and intracameral cefuroxime [20] is applied.

Surgical technique

There have been cases where there are various techniques for surgical procedures and can be performed on children and adults. One of the techniques for repair is by using a double armed polypropylene suture, where the suture is left externally on the sclerawith a knot buried in the sclera flap. This technique allows posterior fixation of intraocular lens implants in the absence of capsular support. Another technique [21] for surgery is called pupilloplasty, where the patient would be placed under retro bulbar anesthesia with the pupils being dilated with 1% tropic amide.

Pupilloplasty

One case report has shown that surgery was an effective treatment option. This type of surgery is called pupilloplasty. During pupilloplasty [22], the surgeon cuts through the iris tissue. This removes the 'bridge' that has developed between the two pupils. During this surgery, the patient is placed under retrobulbar anesthesia, and the pupils are dilated with one percent tropicamide. The surgery, in this case report, was successful and improved the individual's vision. More trials are necessary to determine whether pupilloplasty will be effective for everyone with true polycoria. However, as true polycoria is rare, there have not been enough cases to determine a success rate for this treatment

Polypropylene structure

Another surgical procedure used to treat polycoria is the use of a double-armed polypropylene suture. The suture is left externally on the sclera [23] with a knot buried in the sclera flap. This technique enables posterior fixation of intraocular lens implants in the absence of capsular support.

Genetics

The gene that is the cause of this disorder is the PRDM5 gene. The PRDM5 gene also been linked to Brittle Cornea syndrome, which is a tissues disorder of the eye, as well as Axenfeld syndrome. PRDM5 plays crucial role in the molecular composition of the eye, as well as the tissue thickness. Axenfeld syndrome occurs in the patient in a case of the mutation of the FOXC1 gene, which is a heterozygous mutation [24]. The extremely rare condition of Polycoria can also been seen in animals. There are cases of polycoria in cats and dogs. These cats look like they possess magical powers. That's because their eyes are so strikingly unusual, you might think they are otherworldly, magical beings. They are in fact [25], all normal cats, with exception of their eyes. This cat has an eye with black swirls. The cat turns out to have a rare eve condition that gives him more than one pupillary opening in the eye. That's right, his eye has multiple pupils. The condition is called polycoria, which results in a malformed iris and the extra, oddly shaped pupils. The causes may be congenital or result from a diseases or injury affecting the iris. People are speculating on what these cats may be able to see. Many believe cats with polycoria may be seeing multiple images. There could be a "ghosting" effect [26] with images superimposing over top of each other, much like a mirror maze. While others guess the cat may only see fuzzy images at best.





Fig. 3: (A) Cat with polycoria, (B) Dog with polycoria

Prognosis

Postoperative care

Hypertonic saline solution used as eye drops may be used to reduce the corneal edema the use of anti-glaucomatous topical to help improve corneal edema, and aqueous suppressants that are accompanied by miotis, include topical beta blockers, alpha antagonists, and carbonic anhydrase inhibitors. Antibiotics [27] and steroid drops for 4 w post-surgery.

Complications, benefits and associated conditions

The complications of polycoria include blurred vision, poor vision, and vision difficulties from the glare of lights. These complications of polycoria are due to a less effective iris and pupil. Pseudopolycoria, or holes in the iris that look like additional pupils, can be a part of Axenfeld-Rieger syndrome. Complications of surgical procedures are possible suture erosion through the scleral conjunctiva, or both Minor intraocular inflammations [28] during and after surgery. Improved distant and near visual acuity, little defects surrounding sphincter muscles, and normal pupillary margins.

Prevention

There are no known preventive measures for polycoria, however, genetic testing may be able to reveal genetic patterns of the disorder. Conditions such as reduced corneal thickness, are observed in people with cases of polycoria, as well as Keratoconus

[29] (keratoconus is a corneal disease has the possibility of leading to blindness and/or astigmatism). However, there are some proposals that it is caused by a dissociation of the pupil margins, a partial coloboma which is a hole in the eye, or abnormal eye tissue composition.

Case report

A 44-year-old man was referred to our clinic complaining of poor vision in his left eye that had been present since childhood. He had no history of trauma or ocular surgery, no abnormal obstetric history, and no other systemic conditions. On examination, his best-corrected visual acuity (BCVA) was 1.0 dilopters (D) (-1.00, -1.75, 105°) in the right eye and 0.5 D (-4.25, -1.75, 105°) in the left eye, with intraocular pressures [30] of 16 mmHg and 17 mmHg in the right and left eyes, respectively. Secular microscopy revealed 2, 817 cells/mm² in the right eye and 2, 882 cells/mm² in the left eye. Fundus examination was normal in both eyes, as was the right pupil, which measured 3 mm in diameter. However, there were two pupils in the left eye within a 2.5-mm central zone and measuring 1.2 and 1.1 mm in diameter. Both pupils in the left eye had a true iris sphincter and pigment epithelium. Direct and indirect pupillary reflexes [31] were normal in both eyes. After the application of tropicamide 1% drops, both pupils in the left eye dilated.

The patient's left eye is (A) light and (B) dim light (C, D) The pupillary margins are no longer puckered up following dilation with guttae tropicamide 1% and phenyiephrine 2.5%.



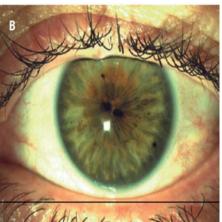


Fig. 4: A) Preoperative image of the right left(normal) eye, before pupil dilatation, B) Preoperative image of the eye polycoria before pupil dilatation





Fig. 5: A) Preoperative image of the right, after pupil dilatation, showing true polycoria, B) Preoperative image of the left eye (normal) eye, after pupil dilatation

Surgery

After performed pupilloplasty under retrobulbar anesthesia after the pupil was dilated with 1% tropicamide, following two 1 mm

limbal incisions, the anterioe iris tissue between the two pupils was then elevated with the spatula to avoid contact with the lens. We then cut the iris tissue using vitreoretinal scissors [32] inserted through the other side port.

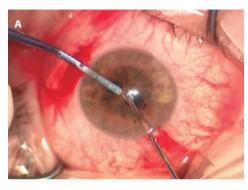




Fig. 6: A) Intraoperative image showing, iris (upper part), B) Intraoperative image showing cutting of the cutting of the iris (lower part)

After aspirating the viscoelastic material, limbal incisions were closed with stromal hydration, and the operation was completed with the application of intracameral cefuroxime. No serious intraocular inflammation [33] was reported during the early postoperative period. Postoperatively, the patient was treated with antibiotics and steroid drops for 4 w. At the first follow-up 1 mo

after surgery, the BCVA had increased to $0.9\ D\ (-3.75, -2.50, 95^\circ)$, the intraocular pressure was $18\ mmHg$, and the lens was clear. The pupil was almost round and measured $3.5\ mm$ in diameter on the first postoperative [34] day but had reduced to $2.7\ mm$ one month after the surgery. The patient was satisfied with his improved vision and better quality of life.





Fig. 7: A) Image showing the result the, B) Image showing the results after end of the postoperative day 1.1 Mo

CONCLUSION

A patient with polycoria won't need any treatment until the condition is affecting their vision and interfering with their daily life. Pupilloplasty has shown positive results for everyone who got it. If you have this rare eye condition, make sure you visit your eye doctor regularly for routine eye check-ups. having regular eye tests is also crucial to your overall eye health as well.

FUNDING

Nil

AUTHORS CONTRIBUTIONS

All the authors have contributed equally.

CONFLICT OF INTERESTS

Declared none

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