CASE REPORT

VISUAL REHABILITATION OF BILATERAL POSTERIOR LENS LUXATION IN CHILDREN: A CASE REPORT

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ABSTRACT

Introduction: Dislocated lens can be subluxated (partial) or luxated (complete), which can cause visual impairment in children. Conservative visual rehabilitation is an option for luxated lens treatment.

Purpose: This study reports the possibility for visual rehabilitation of bilateral of bilateral luxated lens in children.

Case Report: A 3-year-old boy came to the outpatient clinic of Dr. Saiful Anwar General Hospital with complaints of blurred vision on both eyes. The child would always bring objects close to his eyes since the past year. There was no history of ocular injury nor development disorder. Uncorrected visual acuities with LEA symbol on both right and left eyes were 6/114. The best-corrected visual acuity (BCVA) of the right eye was 6/45 and left eye was 6/9 with both S+11.00. Iridodonesis and aphakic lenses were found during slit-lamp examination of both eyes. The intraocular pressures were 14.2 mmHg on the right eye and 17.3 mmHg on the left eye. The lenses were seen in the vitreous cavities during funduscopy and ultrasonography examination. After 1 month of using spectacles, the BCVA of the right eye was 6/18 and that of the left eye was 6/18 with the binocular BCVA was 6/15. Lensectomy with pars plana vitrectomy was planned to prevent complication.

Conclusion: Conservative visual rehabilitation is important to prevent amblyopia in children with luxated lenses and surgical treatment is needed to prevent complication. In this patient, aphakic spectacles were given for visual rehabilitation pending operative treatment.

Keywords: visual rehabilitation, luxated, aphakic, spectacles, lensectomy

INTRODUCTION

Dislocated lens can be defined as the displacement of the crystalline lens from its normal position, which can be partial or complete. Subluxation is the condition in which the lens is not in its normal position but still within the pupil area, whereas luxation is when the lens is detached from the ciliary body, which can be displaced anteriorly (anterior chamber) or posteriorly (vitreous body). Dislocated lens may be caused by damage or dysfunction of the zonule of Zinn. Trauma accounts for most dislocated lens cases (around 50%), but underlying genetic disorders may also play a role. [1, 2]

The displacement of lens can result in other refractive errors, such as myopia, astigmatism, or aphakic hyperopia. An anteriorly dislocated lens can entail increased intraocular pressure, while a posteriorly dislocated lens may lead to damage to the retina, for

instance, retinal ablation or vitreous hemorrhage. Dislocated lens can also cause amblyopia and, even, a permanent decrease in vision. In addition to identifying the cause of dislocated lens, the necessary visual rehabilitation should also be taken into account. Conservative visual rehabilitation may include the use of spectacles or contact lens. Surgical intervention can be in the form of a lensectomy by placing an intraocular lens. Therefore, early detection and visual rehabilitation of dislocated lens cases are essential for preventing visual disorders and blindness in children. [1, 2]

This case report describes a 3-year-old boy whose both lenses were posteriorly luxated into the vitreous body. However, no conclusive cause has been determined, which may point out the result of a genetic predisposition. In a child with aphakia as young as 3, the subsequent step is to consider whether the necessary visual rehabilitation can be conservative or will surgical interventions be needed. Hence, this case report discusses the probable causes of lens luxation and the undertaken visual rehabilitation should the lens be dislocated in the vitreous body.

CASE ILUSTRATION

A 3-year-old boy came to the eye polyclinic of dr. Saiful Anwar Hospital, Malang, complaining of needing to put objects close to his eyes to look at them. He had complained about this condition for the past year without other complaints of his eyes. There was no history of trauma in the patient. He has been immunized according to his age, and his growth and development have been appropriate to his age. During pregnancy, his mother had her health routinely controlled at her obstetrician, and no complaints were found. The patient was born at term through cesarean section because of polyhydramnios. His birth weight was 2,480 grams. There was a sinistral undescended testis that was already operated back in 2018. Other family members of the patient do not have a similar complaint to the patient. The patient is the younger of two siblings.

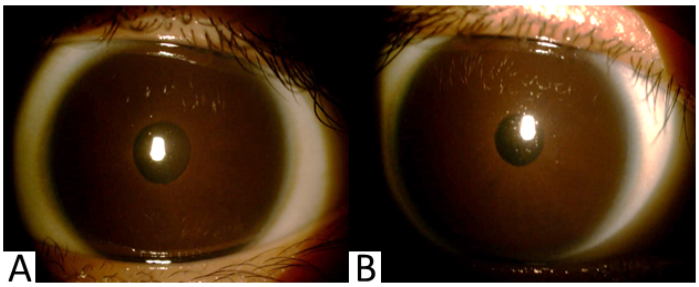

Figure 1. The right eye (A) and the left eye (B). There are iridodonesis and aphakia on both eyes.

Upon measuring the patient's visual acuity using the LEA symbols, the uncorrected visual acuity of the right and left eyes was 1/19M ≈ 6/114. The best-corrected visual acuity of the right eye became 6/45 with +11.00 D spherical lens, while the left eye had a best-corrected visual acuity of 6/9 with +11.00 D spherical lens. Streak retinoscopy of the right and left eye also obtained +11.00 D. The examination of his anterior segment using a slit lamp revealed iridodonesis and aphakia in his right and left eyes. There were normal intraocular pressures on both eyes, with 14.2 mmHg in the right eye and 17.3 mmHg in the left eye.

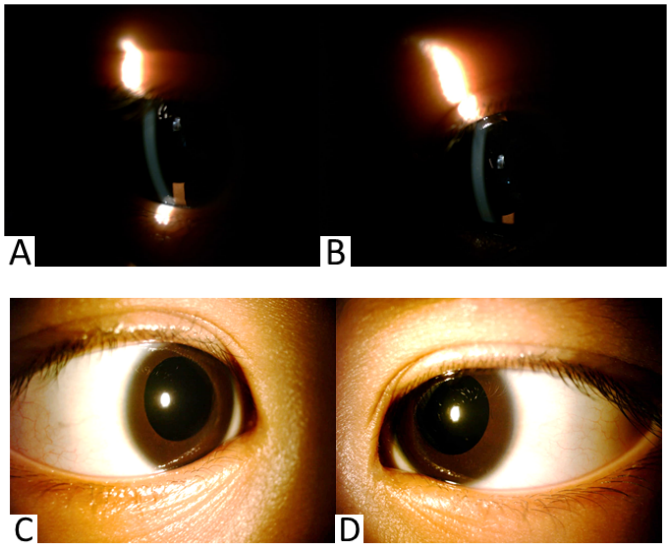

Figure 2. The right eye (A&C) and the left eye (B&D) with dilated pupil. In both photos, there were no signs of lens in the pupil.

The examination of the patient's posterior segment using fundoscopy showed that the lenses of the right and left eyes were in the vitreous body. Upon the eye ultrasonography examination, there were features of the lens (biconvex structure) in the right and left vitreous bodies with positive echodensity in the anterior vitreous media with high spikes.

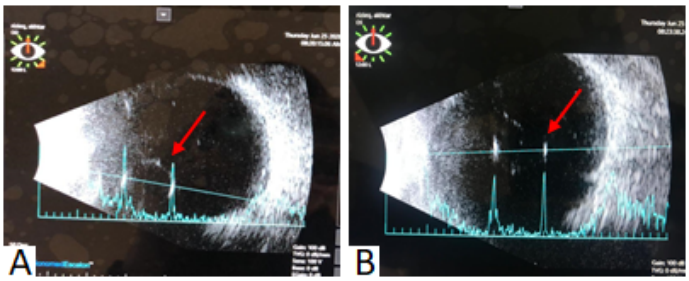

Figure 3. USG of the right eye (A) and the left eye (B). The lenses can be found in the vitreous body of the right and the left eye.

During the inspection of the patient's body posture, there were no other disorders such as high or short stature. There were no deformities on his extremities, with normal fingers and toes. His blood examinations, which include complete blood examination, kidney function,

liver function, blood sugar, and TORCH infection screening, were within normal limits. His chest plain radiography result was within normal limits with no aortic dilation. Based on a consultation with the Department of Pediatrics, the patient showed no abnormalities.

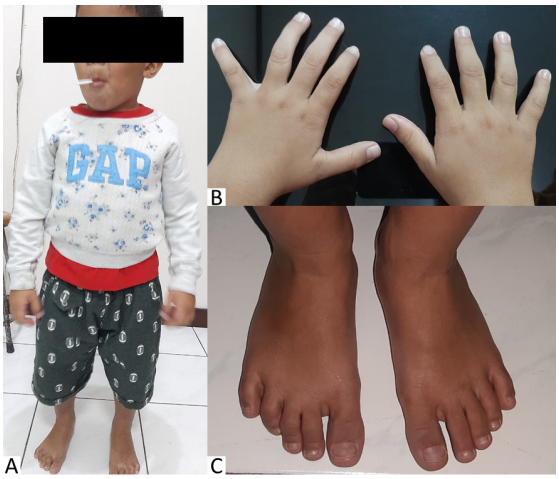

Figure 4. The patient's stature (A), his fingers (B), and his toes (C). No abnormalities in the patient's posture, fingers, nor toes.

The patient was diagnosed with ODS aphakia due to suspected bilateral isolated lens dislocation and was planned to undergo ODS lensectomy with pars plana vitrectomy. While waiting for his surgery, the patient was prescribed spectacles for visual rehabilitation. The power of his spectacles was ODS S+11.00 to see from close and afar, and from a distance of 10 cm, the patient was able to read at a size of 1,6M. After a month had passed, the patient went for medical control and, with the spectacles worn, the right and left eyes' visual acuity was 1/3M ≈ 6/18 and binocular acuity reached 1/2.4M ≈ 6/15. On the following month's examination, the patient's visual acuity remained with both right and left eyes at 1/3M ≈ 6/18 and binocular acuity reached 1/2.4M ≈ 6/15.

DISCUSSION

Dislocated lens in children stems from abnormalities of the zonule (weakness or rupture), thus allowing the displacement of the lens. The prevalence of dislocated lens is reported to reach 6.4/100,000. In children, dislocated lens can give rise to refractive errors and impaired development of vision with varying degrees of severity. This condition can either be congenital or acquired, with congenital dislocation of lens commonly associated with developmental disorders or other systemic diseases (genetic disorders), whereas acquired dislocated lens occurs due to trauma process. If a trauma process is not identified, then it is likely that genetic disorders are at play. Congenital dislocation of lens can appear during birth or spontaneously occur after birth. Congenital anomalies are usually marked with bilateral and

symmetrical abnormalities. [2-4] This patient complained of his condition when he was 2-3 years old, and, based on history taking, there was no history of trauma. The probable cause of the dislocation (luxation) of this patient's lenses is genetic factors because the condition was bilateral.

Subluxated lens can cause increased lens curvature, thus resulting in myopia. The displacement of lens position may also lead to irregular astigmatism not correctable using by cylindrical lenses, and it can trigger amblyopia, strabismus, or monocular diplopia. Anteriorly displaced lens increases intraocular pressure and decreases endothelial cell count, not to mention anterior uveitis. If the lens is located in the vitreous body, it can result in retinal ablation due to free movement of the lens or uveitis (lens-induced) and phacolytic glaucoma should capsule rupture take place. Examinations can be carried out using a slit lamp to identify the presence of iridodonesis or phacodonesis. Inspecting the pupil's dilation is also necessary to evaluate the lens equator and to identify which region experiences zonule weakness or rupture. However, the examinations conducted should be complete and thorough. Exhaustive history taking and eye examinations involve assessing best-corrected visual acuity, intraocular pressure, anterior (slit lamp) and posterior segment (fundoscopy), USG, and other systemic tests (chest plain radiograph, laboratory, and echocardiography). [2, 3] During the patient's visual acuity test, his best-corrected visual acuity was 6/15 with S+11.00 D on both eyes, and, according to the anterior segment examination, there was iridodonesis and no feature of lens in the pupil. The lenses were found inside the vitreous body according to the fundoscopy and USG. Because the patient had his lenses posteriorly luxated, the next step was to observe whether or not there were any complications, such as increased intraocular pressure or retinal ablation. The patient also had undertaken a complete blood test and chest plain radiograph with no abnormalities found. Visual acuity test in children can be influenced by various factors, for example, they are tired, drowsy, or not focused during the test, which may undermine the result. As a result, the test should be iterated or evaluated the next time the patient comes for a visit in the event that they are deemed uncooperative.

Commonly, congenital dislocation of lens occurs on account of underlying systemic or genetic disorders, e.g., Marfan syndrome, Weill-Marchesani syndrome, or Homocystinuria. Congenital disorders usually occur at birth and bilaterally. In a study conducted in Denmark, 68.2% of patients are patients with Marfan syndrome; similar findings were also observed in studies performed in China and Pakistan. [3, 4]

Marfan syndrome is an autosomal dominant genetic disorder, which is the mutation of the FBN1 (encoding fibrillin-1), an important component of the extracellular matrix. Marfan

syndrome is marked with cardiovascular (dilated aortic root), ocular (subluxated lens), and skeletal (pectus deformity, wrist and thumb sign, hindfoot deformity) abnormalities. The stature of children with Marfan syndrome are tall, slender, and have arachnodactyly (long, spider-like fingers and toes in contrast to the palm of their hand and the sole of their foot). The diagnosis of Marfan syndrome can be established despite the absence of a family history having a similar condition. The Ghent nosology is used to facilitate an accurate diagnosis of Marfan syndrome, in which dilated aortic root and subluxated lens play central role in its diagnosis. Ocular manifestations of Marfan syndrome include superonasal subluxation of lens, myopia, cataract, glaucoma, and retinal ablation. [3, 5-7]

Weill-Marchesani syndrome is an autosomal recessive genetic disorder that causes the mutation of the ADAMTS10, ADAMTS17, LTBP2 genes or autosomal dominant genetic disorder of the FBN1 gene closely linked to fibrillin-1. Clinical manifestations of Weill-Marchesani syndrome contrast with those of Marfan syndrome, which are short stature, rotundity, and brachydactyly (disproportionately short toes and fingers due to unusually short bones). Meanwhile, ocular manifestations of Weill-Marchesani syndrome comprise inferonasal subluxation of lenses, microspherophakia (spherically-shaped lens), which gives rise to high-degree myopia, glaucoma, and cataract. On the other hand, the systemic manifestations may consist of stiffness in joints, thickening of the skin, and abnormalities of the heart (cardiac defects or abnormal heart rhythm). [3, 8, 9]

Homocystinuria is an autosomal recessive genetic disorder characterized by the disorder of an individual's metabolism due to a deficiency of cystathionine beta-synthase (CBS). This results in increased accumulation of cystine inside the blood, which impairs connective tissue, muscle system, nervous system, and cardiovascular system. One of the ocular manifestations of homocystinuria is inferonasal subluxation of lens, which may also be accompanied with corneal opacities, congenital cataract, iris atrophy, NII optic nerve atrophy, and retinal detachment. Furthermore, its systemic manifestations include osteoporosis, mental retardation, epilepsy, thrombophilia, and, even, pulmonary embolism in severe cases. Even though patients with homocystinuria present some similar conditions to those with Marfan syndrome, which is arachnodactyly, the differences lie in the presence of mental retardation, relatively larger arm length compared to body height, and the different direction of the lens subluxation. [3, 6]

Upon eye examination, the patient presented with posterior luxation of both right and left lenses, indicating abnormalities of his zonules. Due to its bilaterality and possible onset during birth—though only detected when the patient was already 3 years old—this case was believed to stem from congenital (genetic) factors. The three most frequent origins of congenital

dislocation of lenses in children are Marfan syndrome, Weill-Marchesani syndrome, and homocystinuria with the majority being Marfan syndrome. Despite the fact that these syndromes are primarily the result of other systemic diseases such as cardiovascular, skeletal, and nervous system diseases, this patient did not exhibit such disorders. Furthermore, based on family history, other family members reported no diseases nor complaints analogous to those shown in this patient. Having said this, it does not completely rule out the possibility that genetic factors underpin lens luxation. The diagnosis of Marfan syndrome can still be established regardless of whether or not there were family histories of the condition and other systemic complaints, and only based on lens dislocation with a genetic abnormality on the FBN1 gene.

Isolated congenital dislocation of lens is the displacement of lens that is hereditary and not accompanied with other systemic disorders, be it skeletal abnormalities nor dilated aorta. This condition can be caused by the autosomal dominant genetic disorder of the Fibrillin-1 (FBN1) gene, autosomal recessive genetic disorder of the genes ADAMTS-Like4 (ADAMTSL4) or Prolyl 3-Hydroxylase 2 (P3H2). Fibrillin microfibrils make up zonules in the lens and are expressed by the FBN1 gene. A number of studies have mentioned that the gene ADAMTSL4 is frequently found localized in the eyes and plays a role in the formation of fibrin. Genetic testing is needed to determine any genetic disorders in children. [10-13] Even though the patient do not show other systemic disorders, this study do not dismiss the likelihood that the luxated lens are due to genetic factors. The mutation of FBN1 or ADAMTSL4 may be the reason for this patient's condition. In other words, genetic testing is highly advisable for this patient, though the limitations in the hospital did not make it possible to do so.

Therapy for ocular disorders in children is for visual rehabilitation. This allows for the correction of ametropia, the prevention and treatment of amblyopia, and the restoration of fusion and stereoscopic function in patients who will then be able to improve the prognosis of their vision. Visual rehabilitation in adult patients differs highly from pediatric patients, as the condition of children's eyes are still in their growth phase. This allows for the development of their eyes' refractive elements, i.e., axial and curvature elongation of the corneas. The indications for visual rehabilitation in children are visual impairments, such as best-corrected visual acuity <20/40 on the best eye, decreased field of vision, central field loss, decreased contrast sensitivity, and difficulty in seeing in poorly-lit settings. [14-16] The patient in this study complained of an ocular abnormality that is luxation, which causes visual impairment. Rehabilitation in this case was undertaken to correct ametropia and to prevent amblyopia in such an early age. The patient's uncorrected visual acuity was 6/114 on both eyes, which was

able to be corrected with S+11.00 into 6/18, or less than 6/12 on the best eye. Therefore, the patient can be considered to have undergone visual rehabilitation.

Therapy for dislocated lens can be performed conservatively or through surgical interventions. Conservative therapy may include prescribing spectacles or contact lenses. As the development of techniques and surgical tools for cataracts took place, techniques for operating dislocated lens in children also progressed to some extent, despite the differences in their risk level. One might have to resort to operative procedures when the dislocated lens causes severe visual impairment or conservative (non-operative) therapy is no longer effective. Determining the choice of therapy in children takes place after conducting a comprehensive examination based on the degree of lens opacity, the magnitude of zonular abnormality, visual function, patient age, equipment availability, operator ability, and parental preference. [3, 4, 14, 16] Visual rehabilitation may employ conservative measures pending the necessary surgical procedures to prevent worsening of visual impairments.

Spectacles can be given to children with indications of binocular aphakia, contact lens intolerance, or in the event that intraocular lens implant is not yet feasible. If a child only has monocular aphakia, then spectacles may not have to be the primary choice and contact lens can be selected if possible. To date, spectacles remain the refractive correction of choice for infants and children with binocular aphakia. Some of the benefits of spectacles are more convenient refractive eye exam for spectacles, affordable price, safe use, ease of change, and adjustable lens design according to the required distance (monofocal and bifocal). However, the use of spectacles also has several drawbacks, such as the lenses being thick and heavy, which can lower compliance in their use. The spectacles can also lead to magnification, disrupted field of vision, and ring scotoma due to the sheer size of their lens. Also, they are not cosmetically appealing. [14, 16, 17]

Contact lenses may be the therapy of choice for those with monocular aphakia. Its advantages over spectacles are their comfort during use, more cosmetically appealing than spectacles, the absence of magnification, and they do not disrupt the field of vision. Nevertheless, the shortcomings of using contact lenses are its challenges in measuring their proper size due to the fact that children are sometimes uncooperative, they can easily get lost, and relatively higher cost because of its more complex production process. Contact lenses may also increase the risk of conjunctival or corneal infections and hypersensitivity reactions. The types of contact lenses that can be used are soft contact lenses or rigid gas permeable (RGP) contact lenses. Soft contact lenses are more elastic and can encompass the entire corneal surface, which feel more comfortable to use and has faster adaptation time compared to RGP.

RGP can be prescribed for children with high astigmatism (>2D) and irregular corneal surface. Bear in mind that RGP lenses are less elastic, more rigid, present relatively higher discomfort during use, and have longer time for adaptation than soft contact lenses. Either way, the use of contact lenses in children poses risks of losing them when crying or rubbing their eyes. [14, 16-19]

The patient in this study was given spectacles as opposed to contact lenses because of his bilateral aphakia. Adding to the fact that he was also still 3 years old, further discussion with his parents concluded that spectacles were more preferable because of his daily activities and the possibility of losing his contact lenses during use. There were also other matters of concern, such as the risks of infections and the relative affordability of spectacles in comparison to contact lenses.

To the present day, there has yet been a standard of the proper time for operating dislocated lens in children. Although, some indications for operative treatment may include diplopia not correctable using spectacles, BCVA $\leq$ 0.3, lens equator being located in the middle of the pupil (non-correctable ametropia), lens opacity, and further complications such as secondary glaucoma, uveitis, or retinal ablation. Lensectomy that can be performed comprises intracapsular lens extraction (ICCE), manual irrigation/lens aspiration, phacoemulsification, or pars plana lensectomy. An approach using pars plana lensectomy is selected when the lens has been completely and posteriorly displaced (complete luxation) to the point that the lens is detached from the zonule. Pars plana lensectomy should be carried out promptly if the posterior luxation of lens has inflicted damage to the retina, e.g., retinal ablation, cystoid macular edema, proliferative vitreoretinopathy, and vitreous hemorrhage. After sclerotomy is made during pars plana lensectomy, vitrectomy is then performed to cleanse the vitreous body attached to the lens, followed by phacofragmentation (cleaning the lens using phaco power). A number of studies mentioned that pars plana lensectomy can be performed on children as young as 1 year, though risks of hypotonia, vitreous hemorrhage, retinal ablation, and endophthalmitis should still be taken into consideration. In bilateral cases, surgical intervention is performed on the eye with a more severe condition, followed by the other eye. [3, 20-23] In this patient, pars plana lensectomy was performed on both eyes due to both lenses present in the vitreous body. Despite no other retinal abnormalities being found in nor complained by the patient, exhaustive observations must still be made before operative procedures were carried out to prevent complications.

Based on their location, implant placements are divided into anterior chamber intraocular lens (IOL) and posterior chamber intraocular lens (IOL). The implantation of IOL in the anterior chamber may carry complications such as damage to the corneal endothelium,

which can result in corneal decompensation (bullous keratopathy), hyphema, anterior uveitis, and secondary glaucoma, thus not recommended for children. On the other hand, IOL in the posterior chamber is more advisable because the IOL is at the physiological location of the lens, in a stable position for a long period of time, and has no contact with nearby tissues, thus reducing the risk of inflammation. Anterior chamber IOL had been done on children aged 5 with the result producing correction to visual acuity but with a higher complication risk. Over time, anterior chamber IOL falls out of favor. The placement of IOL at the posterior chamber can be performed using scleral fixation or iris claw. Scleral fixation runs the risk of suture erosion, vitreous hemorrhage, retinal ablation, and endophthalmitis. Currently, iris claw has risen in popularity because it can be performed easier at a relatively shorter amount of time compared to IOL placement using scleral fixation. Scleral fixation and retropupillary iris claw have been performed on children at the age of 3 years. In its early development, iris claw was operated in the anterior chamber. However, placing IOL posteriorly using the retropupillary iris claw is deemed more physiological. [3, 24-27] So far, there has not been a best way of implanting IOL in children without capsular bag support (secondary implantation). Therefore, weighing complication risks, equipment availability, and operator ability remain crucial prior to selecting the technique to be performed.

The measurement and selection of IOL size to be implanted are essential factors in improving visual acuity. Bear in mind that children still undergo their growth period, which means that there will be changes in the diameter and curvature of the cornea, the length of the eyeballs, and the size of the capsular bag. These growths provide a challenge in determining the correct IOL size. As a result, there is no current formula specifically used to measure the size of IOL in pediatric patients, so calculations are carried out using the formula used in adult patients. The growth of eyes in children drastically takes place early in their lives, which gradually slows at the age of 2-3, closely resembling the size of adult eyes at the age of 5-6. To put it in another way, IOL implantation must consider the patient's age and refractive target. Because of the ongoing growth of eyeballs in children, there will be myopic shift after IOL has been implanted. As the IOL recipient ages, the myopic shift lessens and stabilizes approximately at the age of 20. Three approaches used in selecting IOL in children are high hypermetropia in the beginning, emmetropia in the beginning, or low hypermetropia in the beginning. High hypermetropia, in the beginning, may provide advantages of axial growth, which improves hyperopia and achieves a refractive target approaching plano when the patient grows into adulthood. Emmetropia or low hypermetropia are chosen for therapy or to prevent amblyopia or when the patient is likely to have poor compliance of wearing spectacles or

contact lenses after surgery. Refractive target needs to be considered at the contralateral eye so that, when the patient becomes an adult, the bilateral difference does not exceed 3D. Still, routine check-ups are still important to evaluate residual refraction in patients. [3, 16]

CONCLUSION

Visual rehabilitation in patients with bilateral posterior luxation of lenses in children can be performed conservatively to prevent amblyopia. Operative procedures are necessary to avert possible complications. In this case, the conservative management include aphakic spectacles pending further surgical interventions.

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