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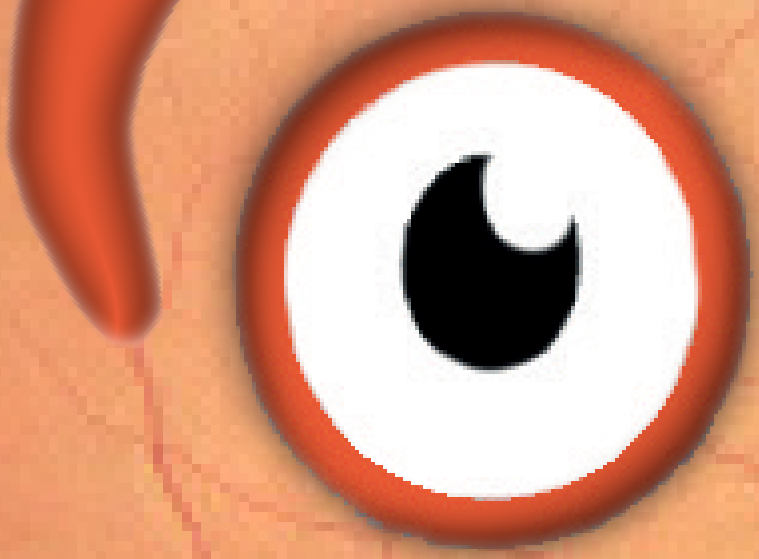

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Original Article

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Open globe injuries: associated findings, management, and visual outcomes

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ABSTRACT

Aims: The aim of this study was to conduct a retrospective analysis of the outcomes of open globe injuries (OGI) in adults diagnosed and treated at our hospital.

Methods: A retrospective review was conducted on the medical records of patients diagnosed and treated for OGI at our clinic. OGI were classified as laceration and rupture. Lacerations were further divided into penetrating, perforating injuries and intraocular foreign body. All OGI were assessed and subsequently underwent surgical repair to restore structural integrity within 24 hours of arriving at our hospital.

Results: A total of 37 cases of OGI were evaluated in this study, consisting of 28 males (75.7%) and 9 females (24.3%). The average age of the patients was 45.8±14 years. The mean age was 48.2±17.2 years for females and 45±13.1 years for males. The initial mean best corrected visual acuity (BCVA) was 2.03±0.79 logMAR, and the mean BCVA at the last follow-up was 0.77±1 logMAR. It was observed that metal objects and wood were the most common causes of injuries. Penetration and rupture were found to be the most common types of injury (45.9%). Intraocular foreign bodies were observed in 8.1%. No perforations were observed.

Conclusion: Open globe injuries continue to be a significant health issue due to their poor visual outcomes.

Keywords: Cataract, intraocular foreign body, open globe injuries, trauma, vitreous hemorrhage

INTRODUCTION

Open globe injuries (OGI) are severe ocular traumas in which the integrity of the globe is compromised, typically through a puncture, tear, or rupture of the cornea and/or sclera.[1] These injuries pose a serious risk to both vision and the structural integrity of the eye, often caused by trauma from penetrating or perforating foreign objects, blunt impact, or lacerations.[2] Open globe injuries are classified as medical emergencies due to the significant risk of vision impairment, infection, and other serious complications that can result in permanent damage to the eye or even its loss.[1-3] Prompt identification and intervention are crucial to protect vision and minimize further harm to the eye's sensitive structures, such as the retina, optic nerve, and lens.[2,3] Treatment typically involves surgical repair, infection prevention, and secondary surgeries for rehabilitation to help recover as much function as possible.[1-4]

Imaging, when used alongside a comprehensive ophthalmologic examination, can be an invaluable tool in assessing traumatic eye injuries.[5] Imaging characteristics indicative of globe rupture include changes in the normal shape or size of the eye, discontinuity of the sclera, intraocular air, a deepened anterior chamber, and the presence of an intraocular foreign body.[5]

To our knowledge, no published study has addressed OGI in our region. The purpose of this study was to conduct a retrospective analysis of the outcomes of OGI in adults diagnosed and treated at our hospital.

METHODS

The study was carried out with the permission of the Tokat Gaziosmanpaşa University Non-intervention Scientific Researches Ethics Committee (Date: 04.03.2025, Decision No: 25-MOBAEK-090). This study was conducted between 2021 and 2025 at the Department of Ophthalmology, Faculty of Medicine, Tokat Gaziosmanpaşa University, in accordance with the principles of the Helsinki Declaration. Patients 18 years of age and older with open globe injury were included in the study. Patients with closed globe injury, as well as those with inadequate follow-up duration, were excluded from the study.

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A retrospective review was conducted on the medical records of patients diagnosed and treated for OGI at our clinic. Data were extracted from the patient files, which included information on age, gender, cause of trauma, best corrected visual acuity (BCVA), findings from both anterior and posterior segment examinations, presence of intraocular foreign bodies, computed tomography findings, treatment provided, surgical procedures, and follow-up period. The visual acuity scores were transformed into LogMAR units.

Open globe injuries were classified according to the system developed by the ocular trauma classification group.[6] Patients were classified into rupture, penetrating injuries, perforating injuries, and intraocular foreign body categories. All patients were 18 years of age or older. All OGI were assessed and subsequently underwent surgical repair to restore structural integrity within 24 hours of arriving at our hospital.

Descriptive statistics were used to analyze the data, with the results presented as mean±standard deviation and percentages. The comparisons between the two groups were made using the Mann-Whitney U test. Comparisons between paired samples were made using the Wilcoxon test. Results were considered significant when the p-value was below 0.05.

RESULTS

A total of 37 cases of OGI were evaluated in this study, consisting of 28 males (75.7%) and 9 females (24.3%). The average age of the patients was 45.8±14 years. The mean age was 48.2±17.2 years for females and 45±13.1 years for males. There was no significant difference between females and males in terms of age (p=0.571). The mean follow-up duration was 12.1±11.1 months.

The initial mean BCVA was 2.09±0.77 logMAR, and the mean BCVA at the last follow-up was 1.07±1.15 logMAR. There was a significant difference between the initial and last follow-up mean BCVA (p=0.001). The final visual acuity was no light perception in 5 cases (13.5%). When we compared the affected zones and visual acuity, the best final BCVA was seen in zone I injuries, followed by zone II and zone III, respectively. The initial mean BCVA was 1.93±0.92 logMAR in zone I, 2.06±0.63 logMAR in zone II, and 2.53±0.21 logMAR in zone III. The final BCVA was 0.67±0.93 logMAR in zone I, 1.11±1.20 logMAR in zone II, and 1.98±1.13 logMAR in zone III.

It was observed that metal objects and wood were the most common causes of injuries. Table 1 shows the causes of OGI. We evaluated the affected zones and found that 19 injuries (51.4%) were in zone I, 9 injuries (24.3%) were in zone II, and 9 injuries (24.3%) were in zone III. Penetration and rupture were found to be the most common types of injury (45.9%). Intraocular foreign bodies were observed in 8.1%. We did not observe any perforations. The most common associated findings in OGI are vitreous hemorrhage, cataract, and hyphema. The associated findings are shown in Table 2.

Table 1. The etiological causes of OGI

Cause	n	%
Cutting/piercing metal object	12	32.4
Wood	9	24.3
Stone	4	10.8
Traffic accident	4	10.8
Glass	3	8.1
Blunt trauma	2	5.4
Cow horn	1	2.7
Fall	1	2.7
Other/unknown	1	2.7

OGI: Open globe injuries

Table 2. Anterior and posterior segment findings associated with OGI

Findings	n	%
Traumatic cataract	18	48.6
Vitreous hemorrhage	12	32.4
Hyphema	11	29.7
Iridodyalisis	6	16.2
Eyelid laceration	5	13.5
Retinal detachment	4	10.8
Intraocular foreign body	4	10.8
Nucleous dislocation	3	8.1
Canalicular laceration	1	2.7

OGI: Open globe injuries

Primary suturation was carried out in all cases. Two cases with no light perception underwent evisceration due to phthisis bulbi during follow-up. There was uncontrolled pain in one case with no light perception at follow-up, and this patient also underwent evisceration. In total, 3 patients underwent evisceration.

Lens clearance with phacoemulsification was performed in 4 cases during the first surgery. After primary suturing, cataract surgery with phacoemulsification and intraocular lens (IOL) implantation was performed on 5 cases. Pupilloplasty was added to one of these surgeries in the same procedure. Cataract surgery with phacoemulsification and scleral-fixated IOL implantation was performed in 2 cases. Secondary IOL implantation was performed in one case. Keratoplasty was performed in two cases due to corneal opacity. Keratoplasty surgeries were performed in different hospitals. Pars plana vitrectomy was performed for 4 patients: one for nucleus dislocation, two for vitreous hemorrhage and retinal detachment and the last one for intraocular foreign body and vitreous hemorrhage. There was one case in which the patients did not want to undergo a second surgery due to retinal detachment.

There were 3 cases with intraocular foreign bodies. Two of the 3 intraocular foreign bodies were located in the lens, and the other one was in the retina. Primary suturation, intraocular foreign body removal, lens clearance with phacoemulsification, and IOL implantation were performed in one case during the first surgery. Primary suturation, intraocular foreign body removal, and lens clearance with phacoemulsification were performed in the second case. Primary suturation was carried out for the third case in the first surgery. Then, phacoemulsification, IOL implantation, and pars plana vitrectomy with intraocular foreign body removal from the retina were performed in the second surgery.

DISCUSSION

Open-globe injuries are a common cause of blindness, typically resulting from sharp objects or blunt trauma. These injuries are characterized by a laceration or rupture in the

cornea or sclera, leading to the exposure of the inner contents of the eye to the external environment.[7] Open globe injuries were classified as laceration and rupture.[6] Ruptures occur due to blunt trauma, where increased intraocular pressure causes the eye wall to rupture at its weakest points. Lacerations, on the other hand, are caused by sharp trauma, where an object cuts or punctures the eye. Lacerations can be further divided into penetrating and perforating injuries. If the object remains inside the eye, it is referred to as an intraocular foreign body.[3,6]

The male patients are more affected than females, with a male-to-female ratio of about 3:1 in this study. Altıntas et al.[8] reported a male-to-female ratio of 3.12 in our country, which is similar to ours. Ozbek et al.[9] reported a 4:1 male-to-female ratio, also from Turkiye. Another study from Turkiye indicated a high male-to-female ratio.[10] In general, there is a male predominance in OGI in the literature.[1,4,8-12] This may be related to occupational exposure and the risk-taking behavior in men.

It was found that the mean age of the patients was 45.8±14 years in our study. It was 48.2±17.2 years for females and 45±13.1 years for males. Yıldırım et al.[10] reported the mean age as 37.82±26.07 (range: 1-95 years). The mean age ranged from 24 to 37 years in studies from Turkiye, which included pediatric cases.[1,8-11] However, we included only patients aged 18 or older. We believe that the higher mean age in our study is due to this difference. Cro et al.[12] also reported a relatively high median age, similar to ours.

We presented that the initial mean BCVA was 2.09±0.77 logMAR, and at the last follow-up, it was 1.07±1.15 logMAR. We observed the best BCVA in zone I OGI, followed by zone II and zone III OGI. We also observed that the most affected zone was zone I, followed by zone II and zone III. Batur et al.[1] indicated that the mean initial BCVA was 2.25±1.10 logMAR, and the mean final BCVA was 1.25±1.19. They also observed that zone I was the most affected zone, and the best BCVA was in this zone. Lee et al.[13] reported that the mean BCVA was 2.04±0.97 logMAR at presentation, and the final mean BCVA was 1.40±1.16 logMAR. These results suggest that a poor visual outcome can be observed after OGI, especially in zone III, and are similar to ours.

When we evaluated the trauma etiologic factors, it was determined that cutting/piercing metal objects were the most common cause (32.4%), followed by wood (24.3%), stone (10.8%), traffic accidents (10.8%), and glass (8.1%) as other common causes. Yıldırım et al.[10] observed that the most common etiologic causes were 28.5% cutting/piercing tools, 13.9% wood, and 9.9% glass, which is similar to our results. Cutting/piercing metal objects, wood, glass, blunt trauma, stone, traffic accidents, and falls were generally observed as the most common causes of OGI in the literature.[1,8-11]

We noted traumatic cataract (48.6 %), vitreous hemorrhage (32.4 %), hyphema (29.7 %) and iridodyalisis (16.2 %) as the most frequent anterior and posterior segment findings associated with OGI. Cro et al.[12] detected vitreous hemorrhage (66 %), cataract (35 %) and hyphema (34 %) as most common associated findings of OGI. In a report from Turkiye, hyphema (33.1 %), vitreous hemorrhage (12.6 %) and iridodyalisis (11.9 %) were determined as prevalent associated findings of OGI.[10] These findings are similar to other studies in the literature.[2,3,14]

CONCLUSION

Open globe injuries continue to be a significant health issue due to their poor visual outcomes. The fact that affected individuals are often of a young age further emphasizes the importance of this issue. Therefore, it is crucial to investigate the causes of eye trauma and identify preventable or correctable factors.

ETHICAL DECLARATIONS

Ethics Committee Approval

The study was carried out with the permission of the Tokat Gaziosmanpaşa University Non-intervention Scientific Researches Ethics Committee (Date: 04.03.2025, Decision No: 25-MOBAEK-090).

Informed Consent

Because the study was designed retrospectively, no written informed consent form was obtained from patients.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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Original Article

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Evaluation of demographic characteristics and management in cases of paralytic strabismus

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ABSTRACT

Aims: To evaluate the demographic characteristics of cases of paralytic strabismus followed up in our Strabismus Unit and to report on their management and long-term outcomes.

Methods: The records of 98 patients diagnosed with paralytic strabismus between 2010 and 2024 at the Strabismus Unit of Fırat University Eye Diseases Clinic were retrospectively reviewed. The diagnosis, etiology, management, and outcomes of the patients were evaluated.

Results: The average age of the patients was 16.8±18.5 years, with 48 (48.9%) being female and 50 (59.1%) being male. The average follow-up period was 7.1±6.7 years. The most commonly affected cranial nerve was the 4th cranial nerve, observed in 63 patients (64.2%). The 6th cranial nerve was affected in 25 patients (25.5%), and the 3rd cranial nerve was affected in 10 patients (10.2%). While 4th cranial nerve palsy was mostly congenital, the etiology of 3rd and 6th nerve palsy was due to underlying neurological, traumatic, and vascular causes. Spontaneous recovery was observed in 10 patients, Botulinum toxin injection was administered to 10 patients, and surgery was performed on 75 patients. Three patients were prescribed prismatic glasses.

Conclusion: Comprehensive clinical examination, detailed medical history, and neurological and systemic evaluations are required for the effective management of patients with paralytic strabismus. If managed correctly, treatment success is quite high.

Keywords: Paralytic strabismus, cranial nerve palsy, etiology, management

INTRODUCTION

Paralytic strabismus is a type of strabismus that occurs as a result of partial or complete paralysis of the cranial nerves that control eye muscle movement. Patients typically present with diplopia, limited eye movement, strabismus, or abnormal head position. The most commonly affected nerves are the 6th (N. Abducens), 4th (N. Trochlear), and 3rd (N. Oculomotor) cranial nerves (CN).[1] A detailed eye examination, along with a thorough neurological and systemic examination, is crucial for determining the etiology and planning treatment in the diagnosis of paralytic strabismus. Paralytic strabismus can be congenital or acquired. In congenital cases, anatomical developmental pathologies of the muscles are often responsible for the etiology, while in acquired cases, vascular diseases, trauma, brain tumors, and neurological diseases are often implicated.[2-4]

Paralytic strabismus is an important clinical condition that negatively affects patients' daily lives, social relationships, and functional abilities. Especially in adults, the sudden onset of double vision can cause serious problems in daily life and work by disrupting visual comfort. In children, however, diplopia is not always clearly expressed, and compensatory changes in head position can, over time, place strain on the musculoskeletal system, leading to postural abnormalities. While spontaneous resolution is not expected in congenital cases, considering these long-term effects, early diagnosis and treatment in the pediatric patient group can prevent both visual and physical complications. In acquired cases, improvements may be observed over time.[5,6] Therefore, the treatment approach varies depending on the patient's age, the duration and cause of paralysis, and the severity of the patient's symptoms.

The aim of this study is to report the demographic characteristics, treatment modalities, and outcomes of patients followed up in the strabismus unit due to paralytic strabismus.

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METHODS

The study was carried out with the permission of the Fırat University Hospital Scientific Researches Evaluation and Ethics Committee (Date: 13.02.2024, Decision No: 2024/03-48). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki. In this retropspective study, the medical records of 98 patients with paralytic strabismus who applied to our clinic between 2010 and 2025 were retrospectively reviewed. Patients diagnosed with paralytic strabismus due to 3rd CN palsy, 4th CN palsy, and 6th CN palsy were included in the study. Patients with diplopia due to myopathic causes such as thyroid ophthalmopathy, myasthenia gravis, Duane retraction syndrome, Brown syndrome, ocular fibrosis syndromes, and chronic progressive external ophthalmoplegia were excluded from the study.

The patients' age, gender, diagnosis, complaints at the time of presentation and duration of complaints, affected eye, etiological causes, best-corrected visual acuity (BCVA), examination findings, treatment method applied, and follow-up information were evaluated.

Statistical Analysis

The data obtained in the study were analyzed statistically using SPSS version 25.0 software (Statistical Package for the Social Sciences version 25.0, SPSS Inc., Chicago, IL, USA). The data were analyzed using descriptive statistical methods. A one-way ANOVA test was used to compare the mean ages between groups. If a significant difference was detected, a post-hoc analysis test was performed to determine which groups differed. The significance level for statistical analyses was set at $p<0.05$.

Mean±standard deviation was used for continuous variables, and percentage values were used for categorical variables.

RESULTS

A total of 98 patients with paralytic strabismus were included in the study. The mean age of the patients was 16.8±18.5 years, and 48 (48.9%) were female and 50 (59.1%) were male. The most commonly affected cranial nerve was the 4th cranial nerve, observed in 63 patients (64.2%). The 6th cranial nerve was affected in 25 patients (25.5%), and the 3rd cranial nerve was affected in 10 patients (10.2%). The average follow-up period was 7.1±6.7 years. The demographic results of the study are presented in Table.

Table. The demographic results for cases of paralytic strabismus

	3rd CN (n: 10)	4th CN (n: 63)	6th CN (n: 25)
Age	31.9±27.3	14.2±13.8	13.5±19.8
Gender (F/M)	4/6	30/33	14/11
Bilaterality	0	8	2
Diplopia	1	8	20
Abnormal head position	0	35	5
Congenital	2	52	2
Acquired	8	11	23
Mean follow-up duration (years)	3.6	7.1	9.8

CN: Cranial nerves, F: Famele, M: Male

The mean age of the 10 patients with 3rd CN palsy was 31.9±27.3 years (Table). Two patients had congenital cases, and 8 had acquired cases. Among the acquired cases, 2 involved trauma, 2 involved diabetes, 2 involved neurological factors (aneurysm, intracranial tumor), and 2 had no identifiable cause. All 10 patients with cranial nerve palsy had restricted gaze in all directions except outward, and 4 patients had ptosis and 1 had diplopia. Five of these patients (50%) had significant exotropia in the primary position, with an average deviation of 23.7±8.5 PD. Spontaneous improvement was observed in 4 of the 10 patients during follow-up, and *Botulinum* toxin injection was administered to 2 patients. One patient who received a *Botulinum* toxin injection experienced recurrence after 6 months and underwent surgery. In 4 patients with partial paralysis, lateral rectus recession and medial rectus resection were performed on the paralyzed eye, resulting in orthophoria in 3 patients. Surgical success was not achieved in two patients with complete paralysis who underwent lateral rectus recession and medial rectus resection together with eyelid surgery. In one of these patients, periosteal fixation of the lateral rectus to the lateral orbital wall was performed, but surgical success was still not achieved.

The mean age of the 63 patients with 4th CN palsy was 14.2±13.8 years (Table). In 28 cases, the right eye was affected; in 27 cases, the left eye was affected; and in 8 cases, there was bilateral involvement. Fifty-two patients had congenital cases, and 11 patients had acquired cases. Among those with acquired strabismus, 4 had trauma as the etiology, 2 had neurological factors (vascular and multiple sclerosis), and 5 had no identifiable etiological factors. Of the 63 patients with 4th cranial nerve palsy, 48 (76.1%) had hypertropia, with an average deviation of 14.1±6.3 it was measured as prism diopters (PD). In addition, exotropia was observed in 10 (20.8%) of the patients with hypertropia, while esotropia was observed in 5 (10.4%). In 53 patients, inferior oblique hyperfunction (IOHF) was observed. The Parks-Bielschowsky three-step test was performed to differentiate these patients from those with isolated IOHF, and it was found to be positive. In the treatment of 63 patients with 4th CN, inferior oblique muscle recession/anteriorization or myectomy was performed in 59 (83%) of a total of 71 paralyzed eyes, including 8 with bilateral involvement. Superior oblique muscle plication surgery was performed in 3 patients, and in 2 eyes that underwent inferior oblique muscle surgery, inferior rectus recession was performed in the contralateral eye due to surgical failure. In 44 (83%) of the 53 patients who underwent inferior oblique muscle recession surgery, orthophoria was achieved at the postoperative examination, while microtropia was observed in 4, exotropia in 3, and minimal hypertropia in 2. In 1 of the cases with postoperative hypertropia, orthophoria was achieved at the end of the 3-month follow-up period. In 2 of the patients with postoperative exotropia, additional lateral rectus recession surgery was required, while no increase in the amount of deviation was observed in the other patients during the follow-up period.

The mean age of the 25 patients with 6th CN palsy was 13.5±19.8 years. Among the acquired cases, 6 had trauma as the etiology, 10 had neurological causes such as increased intracranial pressure (ICP) or tumors, and 7 had no identifiable etiological factor. Of the 25 patients with 6th cranial nerve palsy, 17 (68%) had significant esotropia with restricted outward gaze, with an average deviation of 19.1±8.2 PD. However, nystagmus was

present in 1 patient with 6th cranial nerve palsy, and inferior oblique hyperfunction was noticeable in 1 patient. Of the 25 patients with 6th cranial nerve palsy, 8 (24%) underwent *Botulinum* toxin injection into the medial rectus muscle of the paralyzed eye; after *Botulinum* toxin injection, recurrent esotropia developed in 6 patients after an average of 7±6.6 months, and 1 patient experienced temporary exotropia lasting 3 months after injection. In addition to the 3 patients who experienced recurrence after Botox injection, a total of 5 patients underwent recession surgery of the medial rectus muscle and resection surgery of the lateral rectus muscle. Orthophoria was achieved in all patients who underwent surgery during postoperative examination. In addition, prismatic glasses were prescribed for 3 patients with low deviation and diplopia who did not want surgery.

DISCUSSION

Eye movements are controlled by the third (N. Oculomotor), fourth (N. Trochlear), and sixth (N. Abducens) cranial nerves, which innervate the extraocular muscles in both eyes. Paralytic strabismus occurs as a result of damage to one or more of these nerves and is typically characterized by symptoms such as restricted eye movements, strabismus, diplopia, and compensatory head position.[1,2] In this study, the demographic data, clinical findings, etiological causes, treatment methods, and long-term outcomes of 98 patients diagnosed with paralytic strabismus and followed up at our clinic were retrospectively evaluated.

When we look at our study results in terms of age, the average age of patients with 3rd nerve palsy was higher. This finding can be interpreted as indicating that 3rd CN palsy may be more common in older age groups due to its greater association with systemic diseases.

The most common type of cranial nerve palsy observed in our clinic was 4th CN palsy, followed by 6th cranial nerve palsy. Fourth cranial nerve palsy is the most common cause in patients presenting with vertical diplopia. Additionally, due to its delicate structure and long course, the fourth cranial nerve is more susceptible to traumatic injuries compared to the third and sixth cranial nerves.[7,8] These characteristics may explain why the most common type is 4th cranial nerve palsy. Additionally, 4th CN palsy is more often congenital, and in many patients, the underlying cause cannot be identified. We believe that the result is due to patients spending more time in the relevant department in cases where trauma, neurological, and vascular secondary factors play a more frequent role in the etiology, such as sixth cranial nerve palsy, and that the number of referrals is lower because most of the symptoms are temporary, leading to more patients being referred to eye clinics when the etiology cannot be determined. In contrast, 6th CN palsy is frequently secondary to trauma, neurological, or vascular causes, and temporary symptoms often resolve with treatment of the primary condition in other departments. Therefore, these patients either present to ophthalmology at a later stage or are not referred at all.

In the study conducted by Holmes et al.,[9] the most common cranial nerve palsy was reported to be the 4th nerve, which was similar to the results of our study. Similarly, Bayramlar et al.,[10] Söylev et al.,[11] and Çakmak et al.[12] reported that 4th CN palsy was more common in their studies conducted in our country. In a study conducted in our country by Çolpak et al.,[13] 127 patients were evaluated, and the third cranial nerve palsy was found to be the most common type. However, unlike our study, the average age of this patient group followed up in the neuro-ophthalmology unit was 58.87±13.6, which is considerably higher than the average age in our study. However, unlike the results obtained in our study, there are numerous studies showing that the sixth cranial nerve is the most commonly affected nerve.[4,14,15] In a study by Park et al.[14] that included children and adolescents with acquired paralytic strabismus, cranial nerve VI palsy was observed in 35 of 66 patients, and the most common etiological cause was neoplasia (25%). In our study, acquired and congenital types of paralytic strabismus were evaluated together, and neurological factors were found to be the most common cause in acquired paralysis.

In a study conducted by Priya et al.,[16] 90 pediatric cases were retrospectively examined. In the study the most common cause of ocular motor cranial nerve palsy was congenital involvement of the 3rd and 4th cranial nerves. While congenital involvment was the second most common cause of 6th cranial nerve palsy. In our study, the most common cause of 4th cranial nerve palsy was congenital. However, contrary to the study by Priya et al., the most common reason for 3rd and 6th cranial nerve palsy was acquired. This may be due to the higher mean age of the patients in our study.

Paralytic strabismus can present with variable clinical findings depending on which cranial nerve is affected. Fourth cranial nerve palsy is often characterized by findings such as hypertropia, vertical diplopia, and compensatory head position, which are consistent with the presenting complaints of patients diagnosed with fourth cranial nerve palsy in our study. The high frequency of IOHF may be explained by the decrease in the antagonistic effect of the inferior oblique muscle secondary to paralysis of the superior oblique muscle and its relative overactivity.[17] Sixth cranial nerve palsy typically presents with marked esotropia and restricted outward gaze. In third cranial nerve palsy, ptosis, mydriasis, restricted eye movements in all directions except outward gaze, and often exotropia are encountered. In our study, these clinical findings were also observed to a large extent in our patient groups.

When evaluating the surgical outcomes in our study, the high rate of orthophoria achieved after inferior oblique recession surgery in patients with fourth cranial nerve palsy (83%) demonstrates that this method is an effective and reliable treatment option. The low recurrence rate and minimal residual deviations after surgery support the success of this technique. In 4 cases with high vertical deviation but no significant inferior oblique hyperfunction, surgical reinforcement of the superior oblique muscle was performed. Although inferior oblique muscle surgery is more commonly performed, superior oblique muscle surgery should be considered in necessary cases.

In 9 of 10 patients with paralytic strabismus who received *Botulinum* toxin injections, orthophoria was achieved, while in 1 patient, temporary consecutive exotropia was observed. However, recurrence of strabismus was observed in 7 of the patients who received *Botulinum* toxin injections. This finding indicates that *Botulinum* toxin injection offers a temporary solution and that surgical intervention may be necessary for permanent alignment. In a study by Metz et al.,[18] 22 of 29

patients (76%) who presented with acute sixth cranial nerve palsy and received *Botulinum* toxin injection in the early stage were successful. In a similar study by Holmes et al.,[19] conservative treatment was compared with *Botulinum* toxin application in patients with acute traumatic sixth cranial nerve palsy, and similar improvement rates were observed in both groups. In conclusion, *Botulinum* toxin injection can be used as a first-line treatment, especially in younger age groups or in patients who are not suitable for surgery. A conservative treatment approach may also be preferred, and surgical treatment options may need to be evaluated in patients who experience recurrence after *Botulinum* toxin treatment. When planning treatment, a personalized approach should be adopted based on the patient's preferences and complaints, and the most appropriate treatment method should be determined through a multidisciplinary evaluation.

In our study, third cranial nerve palsy was found to be the least common involvement among cases of paralytic strabismus (10.2%). This finding is consistent with the literature, as the third nerve contains both motor and parasympathetic fibers anatomically, making isolated nerve palsy rare and usually accompanied by other systemic findings. Due to the possibility of serious conditions such as neoplasia, meningitis, encephalitis, intracranial aneurysm, and substance poisoning in its etiology, these patients often present to neurology or emergency departments and may be referred to the ophthalmology department less frequently or at a later stage.[14] Therefore, the low incidence of third nerve palsy in our study can be explained by both the fact that patients present with systemic diseases and symptoms apart from ptosis, mydriasis, or strabismus, and by referral practices in the healthcare system.

Another important finding based on the data we obtained is that the underlying etiological factor could not be determined in 14 patients (14.2%). In particular, in pediatric patients, this situation may be explained by the difficulty of obtaining an accurate and detailed medical history, the forgetting of previous traumas, or the delayed diagnosis of some systemic diseases. In addition, some cranial nerve palsies are congenital and may go unnoticed for many years, which can lead to confusion in distinguishing between congenital and idiopathic types. At this point, a multidisciplinary approach is important for the proper management of both the diagnosis and treatment process.

CONCLUSION

As a result, comprehensive clinical examination, detailed medical history, and neurological and systemic evaluations when necessary are required for the effective management of patients with paralytic strabismus. In addition, the use of imaging methods in suspicious cases may contribute to elucidating the etiology. Long-term follow-up of patients is also extremely critical for the sustainability of treatment outcomes and the prevention of potential complications. Our study aims to contribute to clinical practice in this field and may guide future prospective studies with larger sample sizes. However, our study has certain limitations due to its retrospective design. The reliability of patients' subjective symptoms could not be fully assessed, and some clinical findings may have been incompletely recorded. Although the overall number of patients was sufficient, the small sample size in the 3rd CN palsy group may have contributed to heterogeneity among the patient groups. The fact that all surgeries were performed by a single surgeon may limit the generalizability of surgical decision-making and outcomes.

ETHICAL DECLARATIONS

Ethics Committee Approval

The study was carried out with the permission of the Fırat University Hospital Scientific Researches Evaluation and Ethics Committee (Date: 13.02.2024, Decision No: 2024/03-48).

Informed Consent

Because the study was designed retrospectively, no written informed consent form was obtained from patients.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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Comparative effectiveness of monocanalicular and bicanalicular silicone intubation in congenital nasolacrimal duct obstruction

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ABSTRACT

Aims: This study aimed to evaluate and compare the clinical success and outcomes of monocanalicular (MCI) versus bicanalicular silicone intubation (BCI) in pediatric patients with congenital nasolacrimal duct obstruction (CNLDO).

Methods: A retrospective, comparative study was conducted on 41 eyes of 41 children diagnosed with CNLDO, treated with either monocanalicular silicone intubation (MCI, n=16) or bicanalicular silicone intubation (BCI, n=25). All procedures were performed by 2 oculoplastic surgeons. Tube removal was planned for the 3rd postoperative month. Treatment success was defined as a fluorescein disappearance test of 0 and a Munk score of 0-1, 3 months after tube removal.

Results: Forty-one eyes of 41 patients were analysed in 2 groups as MCI (n=16) and BCI (n=25). The mean age was 43.1±17.9 months (between 24 months and 72 months). Twenty-five (61%) of the patients were females and 16 (39%) were males. Twenty-one (51.2%) of the patients were right eyes and 20 (47.8%) were left eyes. Treatment success was achieved in 93.7% of the MCI group and 88% of the BCI group. There was no statistically significant difference between the MCI and BCI groups in terms of age, gender distribution, and laterality (p>0.05)

Conclusion: Monocanalicular and bicanalicular intubation showed similar success rates in treating CNLDO. However, monocanalicular intubation provided greater tube stability and technical simplicity, making it a practical option in pediatric cases.

Keywords: Congenital nasolacrimal duct obstruction; lacrimal drainage system; silicone tube intubation

INTRODUCTION

Congenital nasolacrimal duct obstruction (CNLDO) is the leading cause of epiphora in childhood.[1] The spontaneous cure rate in the first year of life ranges from 62.8 to 95.0%.[2,3] While the management of CNLDO in children younger than one year is usually hydrostatic massage and treatment with topical antibiotics.[4,5] probing is the preferred method with high success rates in cases of unresolved epiphora in children older than one year.[6,7] However, the success rate of probing decreases with advancing age.[8] Nasolacrimal silicone intubation has been recommended as the primary procedure after failed probing and in children older than 24 months.[9] Bicanalicular intubation (BCI) and monocanalicular intubation (MCI) methods are used for this purpose. In recent years, a number of studies examined the outcomes of these 2 intubations in different age group.[10-12] The aim of this study was to compare the success rate and complications of MCI and BCI in children with CNLDO between 2 and 6 years of age.

METHODS

Study Design

The study, which was conducted in accordance with the principles of the Declaration of Helsinki and Good Clinical Practices, was approved by the Başakşehir Çam and Sakura City Hospital Scientific Researches Ethics Committee (Date: 27.11.2024, Decision No: 284).

This retrospective study was conducted on pediatric patients diagnosed with CNLDO who underwent silicone tube intubation between March 2022 and April 2024 at Başakşehir Çam and Sakura City Hospital, a tertiary referral center in İstanbul. Inclusion criteria were; 1: Age between 24 and 72 months; 2: History of unresolved epiphora despite conservative treatment or failed probing; 3: Clinical diagnosis of CNLDO based on symptoms and diagnostic tests.

Patients were divided into two groups according to the type of intubation performed: MCI or BCI. A total of 41 eyes from

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41 patients were included, with 16 undergoing MCI and 25 undergoing BCI.

Diagnosis and Scoring System

Diagnosis of CNLDO was based on a history of persistent tearing, a positive fluorescein dye disappearance test, and evaluation using the Munk score. The Munk score is a standardized 6-point scale to assess epiphora severity:

- **0:** No epiphora
- **1:** Occasional epiphora requiring dabbing less than twice daily
- **2:** Epiphora requiring dabbing 2-4 times daily
- **3:** Epiphora requiring dabbing 5-10 times daily
- **4:** Epiphora requiring dabbing more than 10 times daily or constant tearing
- **5:** Constant tearing with continuous dabbing

Treatment success was defined as a fluorescein disappearance grade of 0 and a Munk score of 0-1 at the 3rd month after tube removal.

Tube Intubation Procedure

All nasolacrimal intubations were performed by two oculoplastic surgeons under general anaesthesia. BCI was performed with a Crawford intubation tube (FCI, Paris, France). Before intubation, a cotton pad containing 2% lidocaine and 0.0125 mg/ml adrenaline was placed in the nasal cavity for 10 minutes. After dilating the upper and lower punctum and probing with Bowman probe, the metal probes in the silicone tube were passed through the upper and lower canaliculi into the nasolacrimal duct and nasal cavity. The distal ends of the probes were grasped with a small paediatric nasal forceps and the metal ends were removed and connected to each other. MCI was performed with a Monoka tube (FCI, Paris, France). After the metal tip was inserted into the nasolacrimal duct through the upper punctum and canaliculus, the tube was pulled out of the nose in a similar manner. The proximal head of the tube was fixed to the ampulla with a slight pull of the metal probe on the nasal tip. The tube was then cut at the entrance of the nasal cavity.

Patients received antibiotics with topical corticosteroids for 7 days and nasal spray for 3 days postoperatively. Patients were examined at 1 week after intubation to look for early complications such as corneal abrasion or tube dislodgement.

All tubes were removed under local anaesthesia. Intraoperative and postoperative complications were recorded.

Statistical Analysis

Mean, standard deviation, median lowest, highest, frequency and ratio values were used in descriptive statistics of the data. Distribution of variables was measured by Kolmogorov Simrnov, Shapiro-Wilk test. Independent sample T test was used in the analysis of quantitative independent data. Chi-square test was used in the analysis of qualitative independent data, and Fischer test was used when chi-square test conditions were not met. Statistical analysis was performed using SPSS software (version 28.0, IBM, Chicago, US).

RESULTS

A total of 41 patients who underwent MCI surgery in 16 eyes and BCI surgery in 25 eyes were evaluated. The mean age of the patients was 43.3±17.6 months and ranged from 24 months to 72 months. Twenty-five (61%) patients were females and 16 (39%) were males. Twenty-one (51.2%) of the patients were right eyes and 20 (47.8%) were left eyes (Table 1).

The mean age was 41.6±18.6 months for the MCI group and 44.6±16.8 months for the BCI group (p=0.319). There were 10 females (62.5%) and 6 males (37.5%) in the MCI group and 15 females (60%) and 10 males (40%) in the BCI group (p=0.872). In terms of laterality, 10 cases (62.5%) in the MCI group were right eyes and 6 cases (37.5%) were left eyes, while 11 cases (44%) in the BCI group were right eyes and 14 cases (56%) were left eyes (p=0.247) (Table 2).

In the MCI group, the intubation tube was absent in one patient (6.3%) at the first-month follow-up. In contrast, five cases of early tube dislodgement occurred in the BCI group-four (16%) within the first month and one (4%) in the second month-while tube removal was completed by the third month in twenty patients (80%) (p=0.628). There was a history of probing in 11 patients (68.8%) in the MCI group and 11

Table 1. Demographic characteristics of patients, probing history, laterality and success rate

		Min-max	Median	Mean±SD/n-%
Age (month)		24.0-72.0	40.0	43.3±17.65
Sex	Female			25, 61.0%
	Male			16, 39.0%
Probing history	(-)			19, 46.3%
	(+)			22, 53.7%
Laterality	Right eye			21, 51.2%
	Left eye			20, 47.8%
Tube	Bicanalicular			25, 61.0%
	Monocanalicular			16, 39.0%
Time to remove the tube (month)	1st			5, 12.2%
	2nd			1, 2.4%
	3rh			35, 85.4%
Presence of complaints in the 3rd month after surgery	(-)			37, 90.2%
	(+)			4, 9.8%

Min: Minimum, Max: Maximum, SD: Standard deviation

Table 2. Comparison of statistical results of MCI and BCI patients

		Bicanalicular tube		Monocanalicular tube		
		Mean±SD/n-%	Median	Mean±SD/n-%	Median	P
Age (month)		44.6±16.8	44.0	41.6±18.6	33.5	0.643[t]
Sex	Female	15, 60.0%		10, 62.5%		0.872[X²]
	Male	10, 40.0%		6, 37.5%		
Probing history	(-)	14, 56.0%		5, 31.2%		0.121[X²]
	(+)	11, 44.0%		11, 68.8%		
Laterality	Right eye	11, 44.0%		10, 62.5%		0.247[X²]
	Left eye	14, 56.0%		6, 37.5%		
Time to remove the tube (month)	1st	4, 16.0%		1, 6.3%		0.628[X²]
	2nd	1, 4.0%		0, 0.0%		
	3rd	20, 80.0%		15, 93.7%		
Presence of complaints in the 3rd month after surgery	(-)	22, 88.0%		15, 93.7%		0.544[X²]
	(+)	3, 12.0%		1, 6.3%		

[t]Independent sample test / [X²]Chi square test (Fischer test), MCI: Monocanalicular intubation, BCI: Bicanalicular intubation, SD: Standard deviation

patients (44%) in the BCI group. No canalicular complications were seen in both groups. Results regarding age, gender distribution and laterality were similar between BCI and MCI groups. Treatment success defined as a fluorescein disappearance grade of 0 and a Munk score of 0-1 was achieved in 15 of 16 eyes (93.7%) in the MCI group and in 22 of 25 eyes (88%) in the BCI group. There was no statistically significant difference in treatment success between the two groups (p = 0.544) (Table 2).

DISCUSSION

CNLDO is a common lacrimal system disorder in children. While most cases improve in the first 12 months with conservative treatment, probing has been recommended as the most effective method in cases between 12-18 months.[7,13] Although silicone intubation is generally used in case of failure of conservative treatment and probing, it is also recommended as the primary procedure in children over 1.5-2 years of age because probing success decreases with age.[14] The silicone tube acts as a temporary stent and prevents obstruction and fibrosis in the nasolacrimal duct during wound healing.[15] For this purpose, MCI and BCI methods have been evaluated in various studies for silicone intubation.[16-19] In BCI, the silicone tube is passed through both canalicular systems into the nasal cavity, while in MCI, the tube is passed through a single canaliculus into the nasal cavity. This exposes a single canaliculus to trauma in MCI. Studies have shown that monocanalicular or bicanalicular tube use has similar success rates on treatment success and there is no statistically significant difference.[16,20] However, although some authors have shown that BCI and MCI have similar success rates, they prefer monocanalicular tube intubation in terms of ease of placement and removal of the tube, as well as lower incidence of canalicular trauma.[10,19] Our findings support previous literature showing no significant difference in success rates between monocanalicular and bicanalicular intubation techniques.

In previous studies, a low success rate was observed in the BCI and MCI groups with age.[19,21] Rajabi et al.[22] reported that the success rate decreased with advancing age in the BCI group, but age had no effect on success in the MCI group. In this study, we did not observe a meaningful difference in the success rates in children aged 2-6 years who underwent BCI and MCI.

The timing of removal of silicone intubation tubes has been reported as the 3rd month in most studies.[10,16,19] Katowitz et al.[12] suggested that, beyond preventing re-epithelialisation and adhesions, silicone tubes may enhance surgical success by applying physical pressure that stimulates bone growth during early childhood. This is supported by higher failure rates in patients who lost the stent before 3 months or were older than 4 years. El- Essawy et al.[23] evaluated the relationship between the results of silicone intubation and the time of tube removal and reported that early tube removal (approximately two months) did not affect the success rate in children younger than 2 years of age, but in older children, the tube should be removed after at least 3 months. Peterson et al.[24] also reported that removal of the tube before 4-6 weeks significantly increased the necessity for reoperation in children older than 2 years. İn a study by Lim et al.[25] regarding the longer retention of the tubes, they concluded that retention for longer than 12 months showed a significantly lower success rate. In line with most previous studies, silicone tubes in our study were also removed at the third postoperative month.

In studies on the placement of MCI tubes in the upper or lower canaliculus, there are studies indicating that in patients in whom monocanalicular tubes are placed in the upper canaliculus, especially depending on the length and size of the head of the tube, corneal abrasions or ulcers may occur.[21,26] However, Katowitz et al.[12] reported that the complication rate of MCI tubes placed in the upper canaliculus is lower than those placed in the lower canaliculus because the upper punctum is usually more medial and further away from the corneal limbus compared to the lower punctum. In our cases, MCI tubes were placed in the upper canaliculus due to its favorable anatomical accessibility and because, in the event of eye rubbing, the lower canaliculus is more likely to come into contact with the ocular surface. This approach aimed to reduce the risk of corneal irritation and we did not observe any complications in the cornea and conjunctiva in the patients we evaluated postoperatively.

Possible complications of BCI include punctal cleft, displacement of the tube, infection, corneal abrasion, and the tube remaining in place after cutting the cantalum part of the tube.[19,23,25,27] In 1 of the patients who underwent MCI, we could not observe the tube in place in the 1st month. Epiphora symptoms of this patient continued. In 5 patients in whom BCI was removed earlier due to displacement of the tubes, we observed that postoperative epiphora complaints persisted.

During BCI application, tubes removed from the inferior meatus are connected for tube stabilisation. There are different recommendations and studies in the literature. Andalib et al.[16] tied the BCI tube ends with 8 or 10 knots and removed the tube under general anaesthesia, Lee et al.[19] fixed the distal ends to the lateral nostril with 7-0 nylon without tying them together for easy removal of the tubes during BCI, so that the proximal ring of the tube close to the punctum was held with forceps and the tube was removed under topical anaesthesia. In this study, BCI tubes were secured with two square knots. For removal, the tube was grasped from the lower punctum with forceps, gently rotated to expose the knot, and cut just below it with scissors. This method allowed safe removal in the outpatient clinic under local anesthesia, without the need for a second general anesthesia.

Unlike many previous studies that include a broader age range or combine outcomes from various surgical techniques, our study specifically focuses on children aged 2 to 6 years-a group in which probing success typically declines and intubation becomes more relevant. Moreover, the study highlights the practical advantage of MCI, including its stability and simplified outpatient removal, supporting its use in routine pediatric practice without compromising success rates.

Limitations

This study has several limitations. First, its retrospective design may be prone to inherent biases related to data collection and patient selection. Second, the sample size was relatively small, and all procedures were performed at a single tertiary referral center by only two surgeons, which may limit the generalizability of the findings. Third, the follow-up period was limited to three months after tube removal, and long-term outcomes could not be assessed. Lastly, treatment success was defined using clinical grading scales, which, although standardized, may still include subjective components.

CONCLUSION

This study confirms that both MCI and BCI silicone intubation are effective for the surgical treatment of CNLDO in children aged 2 to 6 years. While overall success rates were similar, MCI was associated with fewer early tube dislodgements and offered greater procedural simplicity. These findings, in alignment with prior literature, support the consideration of MCI as a practical and efficient alternative, particularly in clinical scenarios where technical ease and reduced intervention time are prioritized.

ETHICAL DECLARATIONS

Ethics Committee Approval

The study was carried out with the permission of the Başakşehir Çam and Sakura City Hospital Scientific Researches Ethics Committee (Date: 27.11.2024, Decision No: 284).

Informed Consent

Because the study was designed retrospectively, no written informed consent form was obtained from patients.

Referee Evaluation Process

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Conflict of Interest Statement

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Financial Disclosure

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Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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Case Report

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Bilateral acute depigmentation of the iris: a case report

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ABSTRACT

Bilateral acute depigmentation of the iris (BADI) was first described in a case series from Turkiye in 2006. The condition is characterized by bilateral acute depigmentation and discoloration of the iris stroma and pigment dispersion. It mainly affects young females and characterized by acute bilateral stromal depigmentation, without other pathological ocular findings. A 21-year-old woman presented with a history of sudden onset discoloration of the eyes, originally noticed by her family. At ophthalmological examination, best corrected visual acuity was 0.1/1.0 on the Snellen scale. Amblyopia caused by a refractive error (+6 diopters) was present in the right eye. Symmetrical, diffuse iris stromal depigmentation was observed, but no signs of anterior segment uveitis or iridocyclitis or any ocular complaint. Dilated fundus examination was normal. Intraocular pressures were 16/18. The patient was diagnosed with BADI. Topical corticosteroid therapy was immediately initiated. The patient has been under follow-up for approximately six months, and her irises have remained depigmented.

Keywords: Bilateral acute depigmentation, topical steroid, young female

INTRODUCTION

Bilateral acute depigmentation of the iris (BADI) is a rare clinical condition that has recently begun appearing in the literature. The first case series was initially published in Turkiye.[1]

BADI presents with sudden onset bilateral, symmetrical pigment loss and discoloration findings in the iris stroma. The aetiology is uncertain, although viral infections such as HSV have been implicated.

The condition is rarely encountered in the clinical setting, and case reports in the literature are limited. Although BADI is self-limiting, topical steroid drops are used to halt pigment dispersion and suppress symptoms.

Pigments are particularly distributed in the anterior chamber and anterior chamber angle.[2-4] Pigment accumulated in the trabecular meshwork can cause increased intraocular pressure. Patients may present with photophobia, eye pain and red eyes. Visual acuity is generally normal, and there are no signs of inflammation, such as posterior synechiae, in the anterior chamber.[5,6]

CASE

A 21-year-old woman presented due to sudden discoloration in both eyes, which was first noticed by her relatives.

At ophthalmological examination, visual acuity was 0.1 (on the Snellen scale) in the right eye and 1.0 in the left. Amblyopia due to high-grade [+6.00 diopters (dpt)] hyperopia was present in the right eye.

Anterior segment examination revealed diffuse and symmetrical bilateral iris depigmentation. The pupils were symmetrically regular and reactive, and there was no transillumination defect. Gonioscopic examination revealed pigment accumulation in the trabecular meshwork. No signs of inflammation, such as posterior synechiae, keratic precipitates, or cells in the anterior segment, were observed. Dilated fundus examination was normal. Intraocular pressures were 16/18 (with Goldmann applanation tonometry) on the right and left, respectively. BADI was suspected, and topical corticosteroid therapy (topical 0.1% dexamethasone 4*1) was immediately initiated. The patient's medical history revealed an upper respiratory tract infection (URTI) in the previous two weeks, and that she had not received antibiotic treatment. No eye complaints were present in the family. Steroid therapy was discontinued after one month because no increase or decrease in depigmentation was observed. No pathological findings or complaints, other than iris depigmentation, were determined at six-month follow-up examination.

DISCUSSION

BADI is a rare clinical condition characterized by bilateral acute iris stromal depigmentation, and one that particularly affects young women. Visual acuity is normal, and

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although intraocular pressure may increase due to pigment accumulation in the trabecular network, this is also generally normal.[2]

Careful differential diagnosis is required, with the exclusion of diseases such as herpetic iridocyclitis with iris atrophy, pigment dispersion syndrome, pseudoexfoliation syndrome, and Fuchs iridocyclitis. Fuchs heterochromic iridocyclitis involves stellate keratic precipitates and is usually unilateral.[7] In cases of pigment dispersion and pseudoexfoliation, pigments are mostly shed from the posterior iris stroma and accumulate in the zonules, lens capsule and angle.[8] In herpetic iridocyclitis, iris atrophy is sectoral, and transillumination defect, posterior synechiae and pupillary disorders may occur.[9] Cases of bilateral acute iris transillumination (BAIT), which has also been encountered in recent years, exhibit bilateral iris transillumination. In these cases, pigment accumulation on the anterior surface of the lens, posterior synechiae and increased intraocular pressure can be observed, and blurred vision has been reported in 27% of cases. Seventy-three percent of these cases have a history of URTI and oral antibiotic use.[7,10]

Oral antibiotic use and URTI have been implicated in the aetiology of BADI.[2] A case report of two siblings from Egypt reported that both developed BADI a few months after experiencing URTI and that CMV IgG was positive in both cases. The iris depigmentation resolved completely and high intraocular pressure returned to normal following the addition of ganciclovir to the treatment.[4]

Researchers have reported that BADI may also develop due to systemic moxifloxacin use.[5] Patnaik et al.[11] reported a case of BADI developing after COVID-19 infection. The depigmentation in that case decreased with topical dexamethasone therapy. The majority of the BADI and BAIT cases reported during the COVID-19 pandemic were from Turkiye.[12-15] Some of these cases involved oral antibiotic use, while others did not. Viral infections and genetic predisposition play an important role in the aetiology of BADI.

It may therefore be concluded that genetic predisposition along with viral infections may be implicated in the aetiology of BADI. The fact that the majority of cases in the literature have been reported from Turkiye also supports this hypothesis.

In accordance with the literature, our case involved a young woman. No pathological findings other than bilateral diffuse iris stromal depigmentation were present during admission. She also had no recent history of antibiotic use. Topical corticosteroid therapy was initiated, and no abnormal findings were observed during six-month follow-up.

However, a history of mild URTI was present. We suspected that genetic and ethnic predisposition and probably a previous viral URTI had triggered BADI in the present case. The most important limitation of this report is that we had no opportunity to photograph the anterior segment, and none can therefore be provided. However, we have attempted to describe all the examination findings in detail.

CONCLUSION

The purpose of this case report is to raise awareness and contribute to the understanding of a disease that is rarely reported and whose aetiology is still unclear.

ETHICAL DECLARATIONS

Informed Consent

The patient signed and free and informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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Case Report

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Misdiagnosis and mismanagement in stage 4 best vitelliform macular dystrophy: a case report highlighting the role of multimodal imaging

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ABSTRACT

Best vitelliform macular dystrophy (BVMD) is a typically autosomal dominant retinal disorder characterized by accumulation of lipofuscin in the macula, leading to progressive visual impairment. We report the case of a 42-year-old male diagnosed with stage 4 BVMD (vitelloruptive stage) who was previously mismanaged with multiple intravitreal anti-VEGF injections under the incorrect assumption of exudative pathology. Fundus photography revealed the characteristic "scrambled egg" appearance, while optical coherence tomography and fundus autofluorescence demonstrated hyperreflective subretinal deposits and hyperautofluorescence without any evidence of fluid accumulation or choroidal neovascularization. Despite the absence of electrooculography data, the diagnosis was supported by multimodal imaging. This case highlights the importance of accurate diagnosis and staging of BVMD, and emphasizes the need to avoid unnecessary therapeutic interventions in the non-exudative stages of the disease.

Keywords: Best vitelliform macular dystrophy, best disease, anti-VEGF therapy, multimodal imaging, optical coherence tomography, fundus autofluorescence, choroidal neovascularization, misdiagnosis

INTRODUCTION

Best vitelliform macular dystrophy (BVMD), commonly known as Best disease, is a hereditary macular dystrophy characterized by lipofuscin accumulation in the subretinal space, resulting in progressive visual decline.[1-3] The condition is typically inherited in an autosomal dominant manner and most often arises from mutations in the BEST1 gene on chromosome 11q13, which encodes bestrophin-1, a calcium-sensitive chloride channel critical for retinal pigment epithelium (RPE) function.[4,5] Less commonly, mutations in other genes such as PRPH2 (formerly RDS) have been implicated.[6]

Clinically, BVMD presents with a distinctive yellowish vitelliform lesion in the macula, often described as having an "egg-yolk" appearance.[7,8] Disease staging is based on ophthalmoscopic and multimodal imaging findings, progressing from a subclinical stage to atrophic or fibrotic phases.[9] Although visual acuity can remain preserved in early stages, misdiagnosis and inappropriate management may occur, particularly in later stages when the lesion breaks down or mimics other exudative maculopathies such as age-related macular degeneration (AMD).[10,11]

Multimodal imaging, such as fundus autofluorescence (FAF), optical coherence tomography (OCT), and fundus photography, play a central role in staging and differentiating BVMD from neovascular pathologies. Importantly, the absence of intraretinal or subretinal fluid in intermediate stages, particularly in the vitelloruptive stage, can help avoid misinterpretation and unnecessary therapeutic interventions such as anti-vascular endothelial growth factor (anti-VEGF) injections.[4]

In this report, we present a case of stage 4 BVMD that was mismanaged with anti-VEGF therapy due to presumed exudative maculopathy. This case emphasizes the importance of accurate staging, appropriate imaging, and clinical judgment in avoiding overtreatment of inherited retinal diseases.

CASE

A 42-year-old male presented to our retina clinic with progressive bilateral visual decline that was more pronounced in the left eye. His medical and systemic history was unremarkable. The best-corrected visual acuity (BCVA)

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was 0.1 in both eyes. Intraocular pressure (IOP) was within normal limits bilaterally, and anterior segment examination revealed no signs of inflammation or anterior pathology.

Fundus examination revealed irregular, yellowish subretinal deposits in the macular region of both eyes, resembling the classic "scrambled egg" appearance characteristic of the vitelloruptive stage (stage 4) of BVMD. Disruption of the retinal architecture was also noted, suggestive of advanced retinal involvement.

Multimodal imaging was then performed. Fundus photography (**Figure a**) revealed fragmented yellow deposits localized within the macula, consistent with the breakdown of the vitelliform material. OCT imaging (**Figure b**) revealed hyperreflective subretinal material without intraretinal or subretinal fluid accumulation. The outer retinal layers were preserved and no disruption of the ellipsoid zone or signs of choroidal neovascularization (CNV) were detected.

FAF imaging of both eyes (**Figure c, d**) revealed marked hyperautofluorescence within the macular region. Collectively, these findings confirmed the diagnosis of stage 4 BVMD without exudative activity or CNV.

Despite these imaging results, the patient had previously undergone ten intravitreal injections of bevacizumab (anti-VEGF) at another institution under the assumption of exudative maculopathy. Additionally, oral acetazolamide therapy was initiated but was later discontinued due to lack of benefit.

Fluorescein angiography and electrooculography (EOG) were not performed. However, based on clinical and imaging findings, a definitive diagnosis of non-exudative stage 4 BVMD was established. The patient was managed conservatively with close observation and no further pharmacological interventions were pursued.

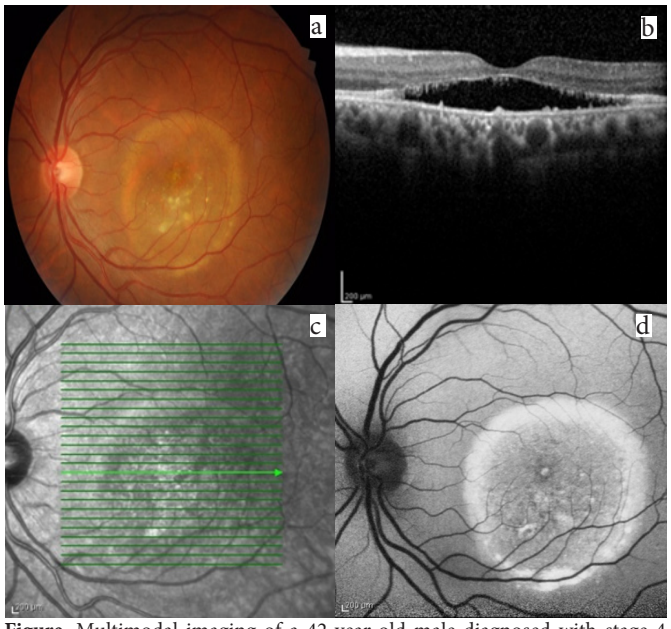

Figure. Multimodal imaging of a 42-year-old male diagnosed with stage 4 best vitelliform macular dystrophy. (a) Color fundus photograph of the right eye showing irregular, yellowish subretinal deposits in the macula resembling a "scrambled egg" pattern, (b) Optical coherence tomography scan revealing hyperreflective subretinal material without intraretinal or subretinal fluid, (c) Fundus autofluorescence image of the right eye showing intense hyperautofluorescence in the macular region, (d) Fundus autofluorescence image of the left eye showing a similar autofluorescence pattern with well-demarcated macular involvement

DISCUSSION

BVMD is an inherited macular dystrophy characterized by abnormal accumulation of lipofuscin beneath the subretinal space, primarily due to mutations in the BEST1 gene.[1-3] Although full-field electroretinogram (ERG) findings are typically normal, EOG remains a hallmark diagnostic test, often revealing a significantly reduced Arden ratio as a result of RPE dysfunction.[4-6] In our case, EOG was not performed; however, multimodal imaging provided sufficient diagnostic clarity.

Many patients with BVMD are asymptomatic in the early stages of the disease, with only subtle funduscopic changes. As the disease progresses, metamorphopsia and progressive visual loss, typically bilateral and symmetrical, become more apparent. The condition progresses through six recognized stages, although not all patients follow a strict sequential pattern.

In stage 1 (previtelliform), the retina may appear normal or show minor pigment changes, although the EOG is typically abnormal. Stage 2 (vitelliform) features a central, yolk-like yellow lesion with preserved surrounding retina. Stage 3 (pseudohypopyon) involves the accumulation of subretinal material with fluid-level separation. In stage 4 (vitelloruptive), the lesion disintegrates into a "scrambled egg" pattern with pigment clumping and early atrophy. Stage 5 (atrophic/fibrotic) is marked by the resorption of vitelliform material and RPE atrophy, making differentiation from other macular diseases more difficult. Stage 6 (cicatricial) may involve CNV and subretinal fibrosis, leading to more pronounced visual impairment. A summary of the estimated visual acuity range according to the disease stage is presented in **Table**.

Table. Estimated range of visual acuity at different stages of best vitelliform macular dystrophy

Stage	Visual acuity
Stage 1 (previtelliform)	100%
Stage 2 (vitelliform)	40-100%
Stage 3 (pseudohypopyon)	40-100%
Stage 4 (vitelloruptive)	20-100%
Stage 5 (atrophic/fibrotic)	<10-100%

Multimodal imaging is critical for BVMD diagnosis and staging. OCT, FAF, and fundus photography can delineate the disease stage, presence or absence of subretinal fluid, and potential complications such as CNV.[7,10,11] In our patient, the presence of hyperreflective subretinal material on OCT and intense hyperautofluorescence on FAF, in the absence of exudation or neovascularization, were characteristic of the vitelloruptive stage (stage 4).[5] Notably, this stage can be easily misinterpreted as exudative maculopathy due to lesion breakdown, leading to misdiagnosis and unnecessary treatment.

A major clinical error in this case was the administration of ten intravitreal anti-VEGF injections (bevacizumab) under the false presumption of CNV or exudative AMD. Although anti-VEGF therapy is a standard intervention for neovascular diseases, it has no proven benefit in BVMD stages lacking fluid or neovascular components.[8,9] Moreover, unnecessary pharmacological interventions pose avoidable risks, burden the patient financially, and delay appropriate management strategies.

This case highlights the importance of integrating clinical evaluation with targeted imaging modalities. While electrophysiological studies can aid diagnosis, careful analysis of OCT and FAF findings may provide sufficient evidence to differentiate BVMD from other mimicking conditions.[8]

CONCLUSION

Accurate staging and interpretation of multimodal imaging are essential for the management of vitelliform macular dystrophy. In the absence of fluid accumulation or neovascularization, unnecessary therapeutic interventions such as anti-VEGF injections should be avoided. This case underscores the potential consequences of misdiagnosis and reinforces the critical role of imaging-guided decision making in inherited retinal diseases.

ETHICAL DECLARATIONS

Informed Consent

The patient signed and free and informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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