СЛУЧАИ ИЗ ПРАКТИКИ
CASE REPORT

Case report
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Clinical Profile and Outcomes of Three Patients Intrapapillary Hemorrhage with Adjacent Peripapillary Subretinal Hemorrhage: A Case Series

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ABSTRACT

Objective. This case series aims to elucidate the clinical presentation, diagnostic imaging, and outcomes associated with intrapapillary hemorrhage with adjacent peripapillary subretinal hemorrhage (IHAPSH), highlighting the importance of recognizing this syndrome in young myopic individuals.

Material and methods. We conducted a single-center, retrospective, observational case series including three patients diagnosed with IHAPSH over the past seven years. Diagnostic modalities included cycloplegic refraction, fundus fluorescein angiography and photographs, optical coherence tomography (OCT), ultrasonography, and visual field testing.

Results. The study involved six eyes from three female patients aged 12 to 46. Two exhibited acute and one subacute visual symptoms. Myopic refraction ranged from −1.5 to −6.50 diopters, and initial visual acuity varied from 1.0 to counting fingers at 1 meter. Biomicroscopy showed no significant findings. OCT revealed peripapillary subretinal hemorrhages in two eyes and subretinal hemorrhage at the optic disk in another. Preretinal hemorrhage was also observed. Ultrasonography detected slight elevation at the optic disk head; no drusen were found. All hemorrhages resolved spontaneously without sequelae.

Conclusion. Hemorrhages resolved spontaneously in all cases, affirming IHAPSH's benign nature. Advanced imaging provided accurate diagnoses, distinguishing IHAPSH from others and emphasizing conservative management. No chronic sequelae were noted, and differential diagnosis prevented unnecessary treatments.

Key words: *myopia, optic disk, intrapapillary hemorrhage, peripapillary subretinal hemorrhages*

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Клинический случай

Клиническая картина и исход кровоизлияния на диске зрительного нерва и перипапиллярного субретинального кровоизлияния у трех пациентов с миопией

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РЕФЕРАТ

Введение. Представлены случаи из практики с описанием клинической картины, диагностических признаков и исходов, обусловленных кровоизлиянием на диске зрительного нерва – интрапапиллярной геморрагией с прилегающим перипапиллярным субретинальным кровоизлиянием (синдром IHAPSH) у молодых людей с близорукостью.

Материал и методы. Проведен ретроспективный анализ наблюдений у пациентов с диагнозом IHAPSH за последние 7 лет. Диагностические методы включали циклоплегическую рефракцию, флуоресцентную ангиографию и фотографии глазного дна, оптическую когерентную томографию (ОКТ), ультразвуковое исследование и определение полей зрения. В исследование были включены 3 пациента (6 глаз) в возрасте от 12 до 46 лет. Миопическая рефракция варьировала от −1,5 до −6,50 дптр, а исходная острота зрения – от 1,0 до счета пальцев на расстоянии 1 м.

Результаты. У двух пациентов наблюдались острые и у одного подострые нарушения зрительных функций. Биомикроскопическое исследование глаз не показало существенных отклонений. По данным ОКТ были выявлены перипапиллярные субретинальные кровоизлияния и субретинальные кровоизлияния в диск зрительного нерва на парном глазу. Также наблюдались признаки преретинального кровоизлияния. Ультразвуковое исследование выявило небольшую проминенцию головки диска зрительного нерва; друз не обнаружено. Все кровоизлияния разрешились спонтанно, без последствий.

Заключение: Кровоизлияния рассасывались спонтанно во всех случаях, подтверждая доброкачественный характер синдрома IHAPSH. Более детальная оценка на основании объективных исследований позволила провести точную диагностику, дифференцирующую синдром IHAPSH от другой патологии, подчеркивая отсутствие необходимости консервативного лечения. Каких-либо вторичных хронических последствий отмечено не было, а дифференциальная диагностика позволила избежать необоснованного вмешательства и дополнительной терапии.
Ключевые слова: *миопия, диск зрительного нерва, перипапиллярные субретинальные кровоизлияния, синдром IHAPSH*

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INTRODUCTION

Intrapapillary hemorrhage with adjacent peripapillary subretinal hemorrhage (IHAPSH) syndrome, first identified in 1975, is a rare, benign condition predominantly observed in myopic eyes with tilted disks. This syndrome is characterized by acute onset of non-specific visual impairment, floaters, and scotoma [1]. IHAPSH is characterized by a tilted optic disc, mixed types of bleeding, good visual acuity, benign prognosis, and rare recurrence [2]. Most cases regress spontaneously without the need for treatment. However, IHAPSH may be responsible for outer retinopathy like sequela in the long term [3, 4]. However, complicated cases can present diagnostic challenges, leading to unnecessary and incorrect treatments.

The diagnosis is primarily clinical, based on suspicion, and involves a comprehensive assessment of clinical symptoms and detailed multimodal imaging to confirm the characteristic features associated with this rare clinical syndrome [2]. It must be differentiated from other pathologies that can cause hemorrhage at the optic disk head, like optic disc drusen or peripapillary choroidal neovascularization (CNV) [5, 6]. Accurate diagnosis is essential to prevent unnecessary and inappropriate treatments in this self-resolving benign condition.

This case series underscores the significance of recognizing IHAPSH in young myopic individuals, highlighting the role of imaging techniques in understanding the pathogenesis and clinical features of this rare condition.

PATIENTS AND METHODS

This case series is a single-center, retrospective, observational study that included three patients diagnosed with IHAPSH over the past seven years (table). All patients were monitored at our clinic, and either they or their legal guardians provided informed consent. Patient records were retrieved from our archives, and relevant imaging data were extracted. Cycloplegic refractions were measured via autorefractor (Nidek ARK-510a, Tokyo, Japan). Fundus images and fundus fluorescein angiographies (FFA) were obtained using a Visucam 500 (Carl Zeiss Meditec, Jena, Germany). Optical coherence tomography (OCT) images were captured

Table

Clinical Characteristics of Patients with IHAPSH Syndrome

Таблица

Клиническая характеристика пациентов с синдромом ИГПСК

Case	Case 1	Case 2	Case 3
Age	12	19	46
Gender	F	F	F
Systemic Findings	–	–	–
Affected Eye	L	R	R
Spherical Equivalent [D]	−1.5/−1.75	−3.5/−2.25	Very high myopia/−6.5
BCVA	1.0/0.6	0.9/0.9	CF/0.2
Vitreous Hemorrhage	Yes	Yes	Yes
Optic Disc Swelling on USG	Yes	Yes	Yes
Visual Field Defect	Yes	No	Unknown
Recurrence	None	None	None
Sequelae	None	None	None
Subsequent Vision Loss	No	No	No
Family History	None	None	None

BCVA: Best corrected visual acuity, CF: Counting fingers, D: Diopter, F: Female, L: Left, R: Right, USG: Ultrasonography

using a spectral domain OCT system (Heidelberg Spectralis, Heidelberg Engineering, Heidelberg, Germany). Ultrasonographic scans were performed with a Cinescan S (Quantel Medical, Cedex, France), and visual field tests were conducted using the SITA-standard 30-2 protocol on a Humphrey Visual Field Analyzer III (HVF Analyzer III, Carl Zeiss Meditec, Dublin, CA, USA).

CASE PRESENTATIONS

Case 1

A 19-year-old female patient presented with subacute visual loss in her left eye. Cycloplegic refraction revealed −1.5 −0.5 × 180 in the right and −1.75 −0.75 × 170 in the left eye, with best corrected visual acuity (BCVA) of 1.0 in the right eye and 0.6 in the left eye. Biomicroscopic examination was unremarkable. Dilated fundus examination, along with OCT imaging, revealed peripapillary subretinal hemorrhage and preretinal hemorrhage *(Fig. 1 a, b)*. Visual field testing revealed enlargement of the blind spot and temporal arcuate scotoma in the affected left eye. Ultrasound imaging indicated mild swelling at the optic disc. Cranial and orbital magnetic resonance imaging (MRI) showed no pathology, and FFA detected intrapapillary hemorrhage with no additional findings. An 8-month follow-up revealed complete resolution of the hemorrhages and a return to a BCVA of 1.0 in both eyes.

Case 2

A 12-year-old female with no significant medical history presented with acute visual loss in the right eye. Refraction was −3.0 −1.5 × 180 in the right and −2.25 −1.5 × 170 in the left eye. BCVA was 0.9 in both eyes. Biomicroscopic examination showed no pathological findings. Fundoscopic examination revealed peripapillary subretinal and preretinal hemorrhages in the right eye *(Fig. a, b)*. Visual field testing did not detect any increased scotoma. Ultrasound and OCT imaging highlighted optic disc swelling. FFA detected intrapapillary hemorrhage with no additional findings. The hemorrhages resolved spontaneously during follow-up.

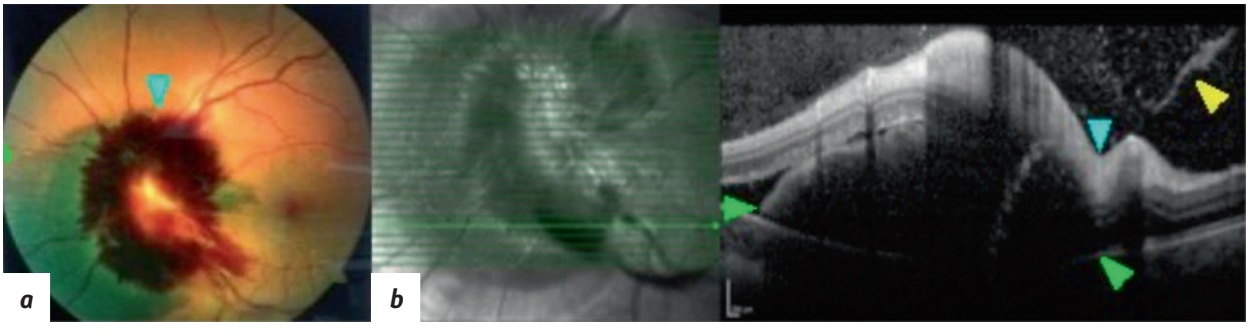

Fig. 1. Figure 1*a* displays the color fundus photograph of the left eye of Case 1. Figure 1*b* shows the corresponding area in the infrared photograph with the optical coherence tomography of the optic disk head. Blue arrowheads, indicate intraretinal hemorrhage and areas of subretinal hemorrhage are marked with green arrowheads. The area with posterior vitreous detachment is shown with a yellow arrowhead. Note the hyperreflective spots in the vitreous, which are areas of hemorrhage.

Рис. 1. Клинический случай 1: *а* – цветная фотография глазного дна левого глаза; *б* – соответствующая область на инфракрасной фотографии с оптической когерентной томографией головки диска зрительного нерва. Синие стрелки указывают на область интраретинальных кровоизлияний, зеленые стрелки – на участки субретинального кровоизлияния. Область задней отслойки стекловидного тела показана желтой стрелкой. Видны гиперрефлективные пятна в стекловидном теле, являющиеся участками кровоизлияний.

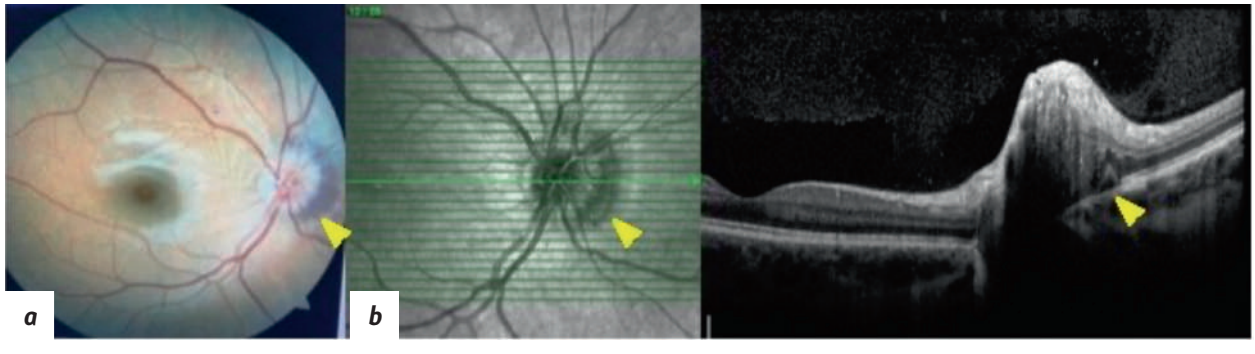

Fig. 2. Figure 2*a* displays a crescent-shaped subretinal hemorrhage area, indicated by the tip of a yellow arrow, located nasally to the optic disk within a limited area. Note the slight blurring of the optic disk margins. In Figure 2*b*, the subretinal hemorrhage corresponding to the infrared area is again indicated by the tip of a yellow arrow.

Рис. 2. Клинический случай 2. Область субретинального кровоизлияния: *а* – в форме полумесяца, обозначенная желтой стрелкой, расположенная назально относительно диска зрительного нерва на ограниченной площади. Обращает на себя внимание небольшое размытие краев диска зрительного нерва; *б* – соответствующая область на инфракрасной фотографии, отмеченная желтой стрелкой.

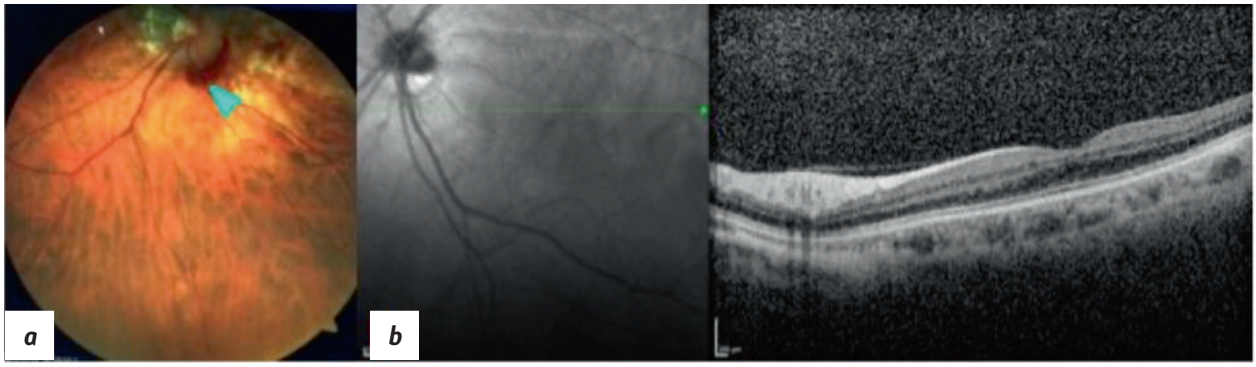

Fig. 3. Figure 3*a* shows a fundus photograph with findings of a myopic fundus and tilted optic disk, where a crescent-shaped subretinal hemorrhage area is visible nasal to the optic disk. In Figure 3*b*, due to the patient's poor fixation resulting from severe amblyopia, the OCT section could not pass through the hemorrhage area, but the captured section shows no posterior vitreous detachment.

Рис. 3. Клинический случай 3. Фотография глазного дна: а – характерные для близорукости изменения глазного дна и косой вход зрительного нерва, где видна область субретинального кровоизлияния в форме полумесяца, расположенная назально от диска зрительного нерва; *б* – из-за плохой фиксации пациента на фоне амблиопии высокой степени срез ОКТ не захватывает область кровоизлияния, но в области исследования отслойка задней гиалоидной мембраны стекловидного тела отсутствует.

Case 3

A 46-year-old female with congenital ptosis, high myopia, severe amblyopia and esotropia presented with acute visual field loss in her right eye. Visual acuity was counting fingers at 1 meter in the right eye and 0.2 in the left eye. Biomicroscopic examination was normal, and dilated fundus examination along with OCT revealed a tilted disc, extensive peripapillary hemorrhage, and intravitreal hemorrhage in the right eye *(Fig. a, b)*. Differential diagnosis included CNV and macroaneurysm. A reliable visual field test could not be performed due to severe amblyopia in the right eye. FFA did not reveal any CNV or leaking areas. Ultrasound was ruled out buried drusen. Cranial and orbital MRI findings were reported as normal. The symptoms and hemorrhages were resolved spontaneously.

DISCUSSION

IHAPSH is a clinical finding rather than the disease itself. It is very uncommon and has been reported rarely [1]. The common ocular findings in this disease include sudden onset in macular regions, increased risk in eyes with myopic tilted disks, frequent involvement of the superior and nasal sides of the optic disk, the coexistence of multiple types of hemorrhages, a benign course, and rare recurrences [7–9]. Potential pathogenic mechanisms have been proposed to include vitreopapillary traction, bleeding from fragile prelaminar vessels in swollen optic disks, hemodynamic effects of the Valsalva maneuver, and complications from optic disk edema [1].

Hemorrhages observed in our study typically resolved as expected; intrapapillary blood was absorbed within weeks, and subretinal hemorrhage regressed over months, aligning with findings reported in the literature. None of the patients in our three cases developed complete posterior vitreous detachment (PVD), consistent with previous reports [1, 2, 4, 10].

Despite significant and striking changes in the optic disk, the syndrome was considered benign and did not necessitate extensive further investigation. We performed ancillary tests, such as magnetic resonance imaging, to rule out other neuro-ophthalmological diseases, although such tests are not always mandatory. The wide spectrum of symptoms associated with vitreopapillary traction complicates the prediction of clinical outcomes for clinicians. In cases with longstanding subretinal hemorrhage, there may be pigment epithelial toxicity, which could manifest as outer retinal findings in OCT [4]; however, none of our cases developed chronic sequelae.

Differential diagnoses for IHAPSH include optic nerve head drusen, optic disk vasculitis, optic neuritis, ischemic optic neuropathy, and peripapillary subretinal neovascularization [3, 4, 7]. In cases with a suspicion of CNV, FFA can be invaluable in ruling out this condition, as demonstrated in our cases.

In conclusion, this case series enriches the existing literature on IHAPSH syndrome by detailing the benign prognosis and potential for spontaneous resolution while also highlighting the critical need for differential diagnosis. Future studies should aim to elucidate further the pathophysiological mechanisms underlying IHAPSH and to explore potential genetic or environmental factors contributing to its incidence. This could lead to more targeted surveillance strategies and therapeutic approaches in at-risk populations.

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