CASE REPORT

Acute Reversible Charles Bonnet Syndrome Following Eye Patch Placement

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ABSTRACT

Charles Bonnet syndrome (CBS) is characterised by recurrent vivid visual hallucinations in the presence of normal cognition. We present a case of CBS secondary to eye patching following Pars Plana Vitrectomy with an unusually acute onset in a 48-year-old woman. She presented with formed visual hallucinations that started less than 30 min after patching of her left eye. The patch was removed after 2 d, and these hallucinations persisted 2 d following eye patch removal. It is important that the ophthalmic surgeon be aware of the potential for development of CBS and offer appropriate referral and reassurance should it occur.

Keywords: Charles Bonnet syndrome, patching, visual hallucinations, acute

INTRODUCTION

The Charles Bonnet syndrome (CBS) is named after Charles Bonnet, a Swiss naturalist and philosopher, who first described the hallucinations of his 89-year-old grandfather.¹ The syndrome is characterised by the triad of complex visual hallucinations, ocular pathology causing visual deterioration, and preserved cognitive status.² The prevalence of CBS has been reported to vary from <1% to 60% in different ophthalmological populations.³ Despite many case reports, there is much to learn about the syndrome and the exact pathogenesis remains unknown. Herein we describe a case of transient CBS secondary to eye patching following ocular surgery. Eye patching is commonly used postoperatively in ophthalmology. It is important that the ophthalmic surgeon recognise CBS as a potential postoperative complication of eye patch placement.

CASE REPORT

A 48-year-old woman presented with decreased vision for 1 month in her left eye caused by cataract and retinal detachment. She had no other medical conditions and was not on medication.

Ophthalmic evaluation revealed a cataract in the right eye, visual field was full to confrontation. Applanation tonometry revealed a pressure of 13 mmHg in the left eye and 15 mmHg in the right eye. We detected an inferior retinal detachment caused by horseshoe-shaped retinal tears located at 6 o'clock in the left eye involving the macular area. Her preoperative best-corrected visual acuity (BCVA) was Left Eye: hand movements and Right Eye: 20/80. After reviewing various management options, the patient underwent Pars Plana Vitrectomy (PPV), planned subtotal retinal detachment, gas injection, phacoemulsification, and implanted intraocular lens in the left eye, under sub-tenon's local anesthesia (STLA). One hour after the operation, a pressure dressing was applied to the left eye for 2 d. At follow-up, 30 min after patching of her left eye, she began to experience complex, vivid, elaborate, and coloured visual hallucinations in her right eye. These hallucinations would last from a few minutes to 30 min, occurring usually when the lighting in the room was low, particularly in the evening. She reported complex hallucinations including men, women, and children dressed in blue clothing and white hats. She did not recognise the people who appeared in continuous motion. They moved towards her and disappeared when they reached her. She could relieve this vision

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by blinking her eyes, but the problem increased if she kept her eyes half closed. The patient denied ever experiencing auditory, tactile, or olfactory hallucinations in the past. She denied delusional symptoms. She did not hear voices associated with the visions, and the images disappeared during sleep. She found the images quite frightening, but retained insight throughout. She was alert and oriented, and her BCVA was hand movements in the left eye.

Immediately after the onset of hallucinations, a complete medical examination and laboratory evaluation were performed without any abnormalities noted. The patient had no history of cognitive impairment. No toxic, metabolic, infectious, or psychiatric aetiologies were found. Her vital signs were within normal limits and stable. The patient's complete blood count, comprehensive metabolic panel, and thyroid function tests were all within normal limits. The patient was offered magnetic resonance imaging (MRI) along with a psychological evaluation. An MRI was performed, and the radiologist reported overall unremarkable results from the brain. At the same time, we administered the Mini-Mental State Examination (MMSE), and the patient scored 30, suggesting intact cognition. At the psychological evaluation, the patient was alert and oriented to person, place, time, and situation. She was well-dressed and made good eye contact. Speech was fluent, coherent, and of appropriate rate and volume. The patient was able to sustain attention without difficulty, and there was no evidence of a fluctuating level of consciousness. Her affect was mood congruent and of full range. She reported anxiety surrounding the onset of the hallucinations and hesitancy to report the symptoms. No delusional symptoms were noted. She was able to verbalise that the visual hallucinations were "not real". Insight and judgement were intact. No involuntary movements were observed at this time. On the basis of the patient's history and presenting symptoms, we concluded that she was suffering from neither dementia nor delirium; that there was no psychiatric explanation for her visual hallucinations; and that the presentation of the patient's case was in fact consistent with CBS. In our case, constant reassurance was used. The patient seemed satisfied with our explanation, and the anxiety symptoms were resolved completely in a few days. Following removal of the dressing, the hallucinations continued albeit less frequently in the evenings for the next 2 d, before completely stopping. Her BCVA was 20/400 in the left eye after removal of the patch. At the follow-up examination after 10 months, the patient was still symptom-free.

DISCUSSION

CBS has been reported previously after sudden visual loss. This case describes an unusually acute reversible

CBS following eye patching. In one similar case, hallucinations began 2 d after patching and ceased 2 ds after the patch was removed.⁴ In another case, the hallucinations started as soon as the patch was applied.⁵ We believe that the patient's presentation was consistent with CBS because of her decreased visual acuity, the new onset of the visual hallucinations, which were both vivid and complex. The patient had insight that these hallucinations were not real, although she was startled by their intense nature. She had no history of psychiatric illness, she had a clear sensorium, and showed no significant change in her cognitive functioning.

In our case, the operation was performed under STLA, without any sedation. The STLA consisted of 4 ml of 2% lignocaine delivered with a Stevens cannula. To an extent, the effects of sedation or anaesthetic drugs have been excluded as a possible cause of the hallucinations. Closed-eye visual hallucinations have been reported following local anaesthesia. In one case, the onset of hallucinations began a few hours after the use of lidocaine and resolved approximately 3 d later, but the total dose of lidocaine used was not documented.⁶ Therefore, it is unclear whether the dosing exceeded usual ranges. A differential diagnosis for the hallucinations experienced by our case is "black-patch psychosis", which is a state of delirium occurring in the postoperative period when bilateral eye patches have been placed. These hallucinations have been reported to occur 24–48 h after eye patching and are accompanied by disorientation and memory problems.⁷

Because of the fear of a mental illness being diagnosed, patients are often reluctant to discuss these hallucinations. Some studies indicating CBS occurs up to 40–60%, whereas other studies suggest that the occurrence rate is 1% in individuals with visual impairment.³ CBS is a rare disorder with estimated prevalence of 0.4% in Asians.⁸ CBS has been described in a wide variety of ophthalmic conditions, including cataract, glaucoma, diabetic retinopathy, and optic neuropathy. The common risk factors in these patients seem to be poor visual acuity and advanced age.⁹

Its aetiology has not been fully elucidated, and the pathophysiology of CBS is poorly understood. Two commonly accepted theories for the visual hallucinations in CBS are the release and deprivation theories. The release theory suggests that hallucinations result from anomalous signals to the visual cortex from lesions in the visual pathway.² The sensory deprivation or "deafferentation" theory suggests that when the visual cortex is deprived of afferent inputs, it may show spontaneous activity leading to visual hallucinations.⁹ Positron emission tomography–statistical parametric mapping imaging studies initially revealed hypermetabolism in the right inferior temporal area and left thalamus, which disappeared after

treatment with valproic acid.¹⁰ Hypermetabolism of the thalamocortical pathway as a result of deafferentation may contribute to the pathophysiologic mechanism of CBS. Two observations in our case are particularly interesting, and add new information to understanding the aetiology of CBS. First, the visual hallucinations were temporary with eye patching. Second, the fact that the symptoms have occurred with relatively normal vision in the other eye is also of interest. Here, we suggest that postsurgical eye patch placement may mimic acute visual deficit and results in development of the hallucinations. This leads to hyperexcitability in the deafferented brain area. We propose that hallucinations may be due to hypersensitivity of the deafferented cortex and they are triggered by the activation of surviving neurons.¹¹ Therefore, stimulus from the retina, optic tract and other aspects of the brain could now elicit a visual hallucination because the neurons are more sensitive. It is theorised that a reduction of sensory input to specific areas of the brain allows some images into consciousness as hallucinations. Tan et al.¹¹ proposed that when visual recovery ceases, the sensitivity of deafferented neurons returns to the normal baseline level and the same level of stimulus is no longer sufficient to trigger hallucinations, hence explaining their termination. Thus, the visual hallucinations ceased after some time, which may be the result of cortical neuroplasticity and reorganisation.¹² On the other hand, visual impairment in the right eye caused by cataract may be necessary to elicit the hallucinations. Holroyd et al.¹³ suggested a possible explanation might be that while visual disorder is a predisposing factor for visual hallucinations, the degree of visual loss may not be related to the likelihood of developing CBS.

Although not functionally disabling, the hallucinations may be a source of significant anxiety. There are various treatments for CBS; psychotropic medications have been used to treat these patients but to date, no double-blind, placebo-controlled studies have been done to ascertain which treatments are most effective. Many patients need only education and reassurance of the benign nature of the visions. CBS is both underdiagnosed and poorly understood. It has long been considered rare because of the reluctance of patients to report the symptoms for fear of being thought mentally ill. In one study 21% of patients with CBS had not reported their symptoms to anyone, 64% had mentioned them to a family member and 15% had told their ophthalmic care provider.¹⁴ As the population continues to age, more patients will be seen with reduced vision caused by a myriad of ocular diseases, increasing the likelihood that more patients may present with CBS in the future. It is the responsibility

of the primary eye care provider to become familiar with CBS to identify it and help patients deal with its consequences, both visual and psychological.

Avoiding a psychiatric misdiagnosis, clinicians have to consider the syndrome of CBS when patients have a recent visual impairment and they begin to experience complex visual hallucinations.

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