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Tiaprex in The Management of Charles Bonnet Syndrome: A Case Report

KEYWORDS: Charles Bonnet syndrome, hallucinations, Tiaprex.

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ABSTRACT Charles Bonnet Syndrome, a diagnosis common in elderly population, usually present with pleasant hallucinations. But in some cases it may cause significant distress which warrants an early diagnosis and adequate management. Review of literature suggests a variety of management options in psychiatric and ophthalmic realm. We discuss successful management of complex visual hallucinations with the use of tiaprex, an atypical antipsychotic.

Introduction

The phenomenology of Charles Bonnet Syndrome (CBS) was first described in 1769 by a Swiss philosopher named Charles Bonnet, while the eponym CBS was coined by a neurologist named George de Morsier in 1936 [1-2]. CBS terminology is used in literature to define a condition in which the individual experiences recurrent or persistent complex visual hallucinations, most commonly pleasant [3]. Despite a long history of recognition of the syndrome in literature, a stringent diagnostic criteria has not been established [4-5]. This is because of the differences in opinion among researchers regarding the inclusion of visual impairment as an important aspect for the CBS diagnosis [6]. Many authors emphasize the importance of impaired visual acuity which may be due to a lesion anywhere in the visual pathway [7-8] and that of impaired brain function [9], for CBS to develop. In support for impaired brain function there are literature evidences of patients experiencing CBS hallucinations who subsequently developed dementia [10-12]. Social isolation causing a more general sensory deprivation has also been proposed as an important contributory factor for its development [13]. The "release" and "sensory deprivation" theories have been proposed as the possible explanation for CBS occurrence. Former theory postulates release of defective electrochemical impulses due to a lesion in visual pathway, thereby causing visual hallucinations. While the latter proposes a reduced sensory input to brain due to an ocular lesion, thus producing spontaneous discharge by neurons at the level of the retina or cortex [14-15]. CBS cases have also been segregated by researchers into typical and atypical CBS. Cases of typical CBS are exceedingly rare and usually presents with complex visual hallucinations with full insight and without any neuropsychiatric disorder. While most cases of CBS present with visual hallucinations in presence of a neuropsychiatric disorder or with the total lack of insight into the unreality of hallucinations, they are called CBS plus or atypical CBS cases [16]. The prevalence of CBS increases with aging population, as in elderly people the occurrence of vision and cerebral disorders increases [17-18]. Various prevalence rates have been reported, probably due to the use of different definitions and diagnostic criteria, as well as the reluctance of patients to seek medical care because of fear of being labeled with some psychiatric illness [19]. There is also no clear evidence-based medical treatment guideline for the condition. Although proposed management strategy includes a good liaison among psychiatrist and ophthalmologist. First step involves rectifying the underlying cause of visual impairment if possible. Also Interesting to note is the fact that the hallucinations may cease with the progress of visual deterioration to total blindness [20]. Literature on treatment of visual hallucinations in CBS shows drug treatment only partially successful. Although there have been anecdotal evidences of successful treatment of CBS cases with a variety of different medications, which include olanzapine [21-22], risperidone [23], haloperidol [24-25], mirtazapine [26], melperone [27], valproate [28-30], carbamazepine [31], donapezil [32], cisapride [33], ondansetron [34], gabapentin [35] and tiapride [36]. Historically visual hallucinations have been majorly described as a manifestation of mental disorders and thus a negative stigma is attached to them. Thus the individuals who have insight into the unreal nature of their visual hallucinations may get disturbed by the possibility of a mental disorder and this fear might cause their reluctance to discuss in detail their unreal visual experiences with physician [37-39]. Thus acknowledging the fact that visual hallucinations in some cases may be experienced by healthy people due to visual impairment is important. Being non-judgmental towards an individual's experiences and giving reassurance regarding the benign nature of visual hallucination, may have a therapeutic effect. Psychoeducation of patient and family members may provide support and reduce the caregiver's burden [39-41]. This suggest the importance of clinicians to be well verse with CBS so as to avoid misdiagnosis and appropriate management of the condition.

This case report describes a patient who fulfilled the criteria for typical CBS and showed complete remission of symptoms with low dose tiapride.

Case Report

An eighty five year old male patient, a retired businessman of high socioeconomic status came by self with daughter with complaints of visualizing unreal images since past three months. He gave a 35 year history of diabetes mel-

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litus for which he was on regular treatment. Along with this he also gave history of a hypermature cataract in left eye since 5 years and age related macular degeneration in right eye since 3 years with a combined vision of just light perception. Thus he was largely home bound with minimal social contacts since a long time.

He was apparently alright 3 months back when he got surprised to have crystal clear visual experience of a man with beard and a black hat sitting on a moving swing. He used to see it multiple times a day most frequently in the evening times. Gradually over a period of seven days he began to see trains passing by him, as if he was sitting at a railway station. Over a month he started seeing even more events which included people crossing the road and a black cloud passing by him. Initially he used to see these visions for some times in a day which used to continue for 15 to 20 minutes each time, but the visual experiences gradually increased to almost throughout the day, as if he was watching a movie. His combined eye vision was just light perception but these visual experiences were perceived with extreme clarity described by him as detailed, vivid and occurring in external space. Although these visions were non-threatening and a source of amusement at start, the patient with time got increasingly concerned about them as he was aware about their unreal nature. He tried to consciously control them, but was mostly unsuccessful in doing so. Thus the visual experiences which were a source of amusement gradually turned bothersome and unwanted.

These visual experiences were never accompanied by clouding of consciousness or other perceptual disturbances. There was no past or family history of any psychiatric illness or substance use. There was no history of memory problems or cognitive decline in the patient. His physical and neurological examination was within normal limits with normal gait, posture and tone of body. There were no signs of aphasia, agnosia, apraxia or visuo-spatial problems. His mini-mental status examination score was 27/30. His routine blood and urine examinations, venereal disease research laboratory test, electroencephalograph (EEG) and magnetic resonance imaging (MRI) scan were within normal range. His psychodiagnostic evaluation with Rorschach and Thematic Apperception Test was within normal range and revealed no pathological finding.

On mental status examination the patient was conscious, cooperative, oriented to time/place/person with normal psychomotor activity and reaction time. His speech was spontaneous, relevant, coherent with normal rate, tone and volume. His affect was euthymic and was having no thought disorder. In perceptual domain he had multiple visual hallucinations which consisted of people crossing the road or trains passing by or a man with beard and a black hat sitting on a swing or a huge black cloud passing by him. His judgment was intact and insight was present.

He was diagnosed as a case of Charles Bonnet Syndrome. Tablet tiaprex at a low dose of 25 mg per day was started, over which within a period of 2 weeks the patient perceived 25 percent improvement. The frequency of hallucinations decreased to 10-12 times a day and the visions were getting increasingly blurred. Gradually over a period of two months the dose was increased to 200 mg a day over which the patient perceived full remission with no visual experiences. The patient is continued over the same dose and is free from all hallucinations throughout the follow up period of 6 months.

Discussion

This case report describes visual hallucinations with intact insight in a patient having diminished vision. Among the various risk factors for developing CBS described in literature our patient presented with visual impairment, social isolation and age above 65 years. Complex visual hallucinations may appear in a variety of different conditions, while specifically in CBS it arises from lesions at various levels of the visual system causing visual impairment. The patient presented with age-related macular degeneration which is the most frequent cause for CBS described in literature. He had a decrease in visual sensory input which could have provoked visual hallucinations by the release or sensory deprivation hypothesis proposed for CBS. The symptoms started with episodic onset which progressed later with hallucinations present almost throughout the day. The outcome and duration of symptoms in CBS is usually variable and unpredictable with the visual hallucinatory experiences lasting from seconds to days to years [39]. Although the visual experiences started with amazement in our patient but gradually became burdensome and a matter of concern for him, which led him for psychiatric consultation. In previous researches emotional burden and generation of unpleasant responses to visual hallucinations have been described [42-43], which highlights the importance of early diagnosis and management. As per previous reports on CBS, it seems necessary to rule out cognitive impairment and thus our patient was evaluated for signs and symptoms of dementia and scored on MMSE which were within normal range [10-12]. In our case, pharmacological intervention was necessary as the hallucinatory experiences got bothersome to patient. Previously published case reports on treatment of CBS have described the use of antipsychotic or anticonvulsant drugs with variable outcomes. Favorable treatment response to tiapride as an add on drug in two cases of schizophrenia with persistent auditory hallucinations has been described recently [44]. Also in literature successful management of visual release hallucinations with tiapride has been mentioned [36]. Thus we started the patient on atypical antipsychotic tiapride and achieved full remission of symptoms. Although positive outcomes in CBS are common with spontaneous regression of symptoms and partial responses are attributed to spontaneous recovery [44], our patient showed complete remission of his visual hallucinations on tiapride which seems less likely to occur by the above mentioned ways. Our finding is in accordance with the literature evidences and we support the use of tiaprex in CBS cases where the hallucinations are bothersome to the patient.

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