Obsessive-Compulsive Disorder and Hyperphagia in a Boy with Fragile X Syndrome: A Case Report

Seyed-Ali Mostafavi[1], Pouria Yazdian-Anari[2], Maryam Mahmoudi[3], Fahimeh Mirzaei[4], Reza Bidaki[5*], Mohammad Hossein Mahmoodi Meymand[6]

1. PhD Candidate, Psychiatry Research Center, Roozbeh Hospital, Tehran University of Medical Sciences, Tehran, Iran.
2. medical student, Yazd Student Research Committee, Shahid Sadoughi University of Medical Sciences, Yazd, Iran
3. Medical Student. Rafsanjan University of Medical Sciences, Rafsanjan, Iran.
4. Medical Student. Rafsanjan University of Medical Sciences, Rafsanjan, Iran.
5. MD. Assistant Professor of Psychiatry. Department of Psychiatry, Research Center of Addiction and Behavioral Sciences, Shahid Sadoughi University of Medical Sciences, Yazd, Iran.
6. Medical Student. Rafsanjan University of Medical Sciences , Rafsanjan, Iran.

***Correspondence:**
Reza Bidaki, MD. Assistant professor of Psychiatry. Department of Psychiatry, Research center of Addiction and Behavioral Sciences, Shahid Sadoughi University of Medical Sciences, Yazd, Iran.
Email: Reza_Bidaki@yahoo.com
Tel: (98) 353 724 9333
Fax: (98) 353 725 8413

Received: 11 June 2015

Accepted: 10 September 2015

Published in September 2015

Abstract

Objective: Fragile X syndrome is the second etiology for inherited mental retardation. It may concomitant with other psychiatric disorders. Intellectual disability (ID) is a state of functioning that typically begins in childhood and is characterized by limitations in intelligence and adaptive skills. We intend to introduce a male young patient with Fragile X syndrome and Obsessive-Compulsive disorder.

Case Presentation: A single, obese male patient with aggressive behaviors, intellectual disabilities, hyperphagia and Obsessive–Compulsive disorder was admitted for treatment. Behavioral problems and impaired interpersonal relationship was caused many of next problems. Also metabolic syndrome is suggested for him.

Conclusion: In a mental retard patient, obsession may not be a chief complaint, but we saw compulsive behavior dominantly in this patient. Some cultural factors like stigma, low social support and unsuitable community approach towards him may be effective in this case. So, further evaluations and treatment of psychiatric and medical co morbidities is highly needed in an adolescent with mental retardation.

Keywords: Fragile X syndrome, Hyperphagia, Mental retardation, Obsessive-compulsive disorder

Introduction

Fragile x syndrome is the most common form of inherited intellectual disability, with a prevalence 1 in 4000-6000 in male sex (1). The prevalence in males is approximately half of that in females (2). The phenotypic features of fragile X syndrome depending upon age in men. The classic physical manifestations are more obvious in adolescents: long and narrow face, prominent forehead ,prognathism, huge ears, macrocephaly, strabismus, pale blue irises, sunken eyes, arched palate, mitral valve prolapse, hyperlaxity of joints, hypotonia, soft skin over the dorsum of hands, flexible flat feet, and testicular enlargement (volume>25ml after puberty) with normal testicular function

[Downloaded from ijdo.ssu.ac.ir on 2026-11-20]

(3). There are developmental delay, intellectual disabilities, and learning problems in this syndrome (6). Males with fragile X syndrome have delayed language development. Expressive language skills are achieved more slowly than receptive language skills, and the discrepancy between expressive and receptive skills accelerate with age (6). Nearly 10 % of boys with Fragile X are aphasic (8). Attention deficit hyperactivity disorder, anxiety, hyperactivity, inattention, gaze aversion, stereotypic movement, hand flapping, hyper arousal, social anxiety are common (9-11) Males with severe intellectual problems have avoidant behaviors (12). In these patients, social skills often correlate with cognitive level and they do not avoidant familiar persons (12). Other symptoms include anxiety (nervousness, obsessive-compulsive disorder-like obsessions and perseverations), mood instability, aggressive and self-injury behaviors (13,14).

Intellectual disability (ID) is a state of functioning that typically begins in childhood and is characterized by limitations in intelligence and adaptive skills (15). The term of mental retardation has been used instead of intellectual disability, which it defines by three co-existing criteria: Significant sub-average intellectual function, Significant limitations in adaptive functioning, before 18 years of age (16) X-linked inherited genetic disorders or patterns contribute in part to the increased prevalence of ID in males, accounting for approximately 16 % of ID in male sex (17,18). It has been identified more than 90 different X-linked ID genes, affecting a wide range of cellular processes (19). Fragile X syndrome occurs in 1-2 % of individuals with ID and is one of the most common inherited disorders that cause developmental delay and ID (17,20).

In this report we intend introduce a case with Fragile X syndrome comorbid with severe aggressive behavior, hyperphagia and OCD. To our knowledge, this is the first report of the development of OCD in a patient with Fragile X syndrome.

Case Presentation

We present a case of 25 year-old single obese male who developed Obsessive-compulsive disorder (OCD) and he had criteria for Mental retardation and Fragile X syndrome. He was born in a family with low socioeconomic status. He was disable for learning and therefore was out of school. He had a typical phenotype for Fragile X syndrome. He had cluttered speech, poverty thought and speech, harsh and elongated face, large or protruding ears, and large testes (macroorchidism). He was admitted because of aggressive behavior toward his family. Physical aggression was seen toward his family selectively. He admitted twice, the first, nearly 9 months ago, because of assault and aggressive behavior. There wasn't positive psychiatric history in his family. We didn't detect any maternal problems about his mother. There weren't psychotic symptoms and didn't meet criteria for Bipolar disorder. IQ test as Raven was 69. His Body Mass Index was more than 45. He had hyperphagia and his eating behaviors and appetite was out of control. His average daily food intake was more than 4000 kcal based on 3 day food record. (table 1)

Pelvic and abdominal Sonography were normal. Blood pressure was 140/90 mm/Hg.

We started him Topiramate 100 mg at night, Metformine 500 mg twice a day, Atrovastatine 20 mg daily, Medroxy Progestrone acetate twice a day, Metoral 50 Mg twice a day, Thioridazine 25 mg twice a day, but response treatment wasn't favorable.

In second admission, we added Chlorpromazine 25 mg twice a day and

Table 1. Routine laboratory tests

Laboratory test	Result	Normal	Unit
Red blood cell count	5.23 million	4.7 to 6.1 million	cells/mcL
Triglycerides	320	200 to 499 mg/dL is High.	mg/dL
Total cholesterol	350	240 mg/dL and above is High.	mg/dL
Fasting blood sugar	210	70 to 100 mg/dL normal	mg/dL

[Downloaded from ijdo.ssu.ac.ir on 2026-11-20]

Clozapine 25 Mg at night that response was excellent. Compulsive behaviors, hypersexuality and aggression were resolved, but now he was social isolated and extra pyramidal symptoms were obvious. We didn't Karyotyping and diagnosis was as phenotypic states. We emphasized in high social supports and social skill training.

Discussion

"Obsessive-compulsive disorder is extremely rare among patient with mental retardation" (27). Our case was a mentally retarded boy who exhibits contamination obsessions, compulsive hand-washing rituals, hyperphagia, and assault behavior. Behavioral problems and impaired interpersonal relationship was cause many of next problems. Varieties of repetitive behaviors have been seen in patients with mental retardation and autism (28). Our patient showed these repetitive behaviors and also repetitive washing and bathing, as well as over eating. H. Blair Simpson and colleagues (29) have cured comorbidity of eating disorders and obsessive-compulsive disorder with a multimodal treatment program designed to tackle both problems at once. They used a cognitive-behavioral approach (integrating exposure and response prevention) for patients with OCD comorbid with an eating disorder. Current case did not fill the criteria for bulimia or eating disorder we can just use the hyperphagia term in context of a obsession and compulsive behavior. This is a very complex case and usually using medication is necessary and we decided to do so.

We prescribed Topiramate 100 mg/Hs for decline of aggressive behavior and Hyperphagia. The patient showed a suitable response.

Some psychological and cultural factors predispose mental retardation patients to psychiatric status like low self-confidence, poor social skills and in adaptive behavior, dependency, and social stigma on mental retardation (30,31). In our case, some cultural problem like stigma, unsuitable community approach to mental retardation patients, low social support were effective.

Conclusion

Further evaluation and treatment of psychiatric disorders is needed in patients with mental retardation.

Refrences

1. Turner G, Webb T, Wake S, Robinson H. Prevalence of fragile X syndrome. American journal of medical genetics. 1996;64(1):196-7.
2. Saul RA, Tarleton JC. FMR1-Related Disorders. In: Pagon RA, Adam MP, Ardinger HH, Wallace SE, Amemiya A, Bean LJH, et al. editors. GeneReviews(R). Seattle (WA): University of Washington, Seattle University of Washington, Seattle. All rights reserved. 1993.
3. Metcalfe SA, Archibald AD. Fragile X population carrier screening. Genet Med. 2012;14(3):350.
4. Fryns JP. The fragile X syndrome: A study of 83 families. Clinical Genetics. 1984;26(6):497-528.
5. Kau AS, Reider EE, Payne L, Meyer WA, Freund L. Early behavior signs of psychiatric phenotypes in fragile X syndrome. American journal of mental retardation: AJMR. 2000;105(4):286-99.
6. Budimirovic DB, Bukelis I, Cox C, Gray RM, Tierney E, Kaufmann WE. Autism spectrum disorder in Fragile X syndrome: differential contribution of adaptive socialization and social withdrawal. American journal of medical genetics Part A. 2006;140(17):1814-26.
7. Collins SC, Coffee B, Benke PJ, Berry-Kravis E, Gilbert F, Oostra B, et al. Array-Based FMR1 Sequencing and Deletion Analysis in Patients with a Fragile X Syndrome–Like Phenotype. PLoS ONE. 2010;5(3):9476.
8. Hall SS. Treatments for Fragile X Syndrome: A Closer Look at the Data. Developmental disabilities research reviews. 2009;15(4):353-60.
9. Shin S, Yu N, Choi JR, Jeong S, Lee K-A. Routine Chromosomal Microarray Analysis is Necessary in Korean Patients With Unexplained Developmental Delay/Mental Retardation/Autism Spectrum Disorder. Annals of Laboratory Medicine. 2015;35(5):510-8.
10. Lubs Herbert A, Stevenson Roger E, Schwartz Charles E. Fragile X and X-Linked Intellectual

[Downloaded from ijdo.ssu.ac.ir on 2026-11-20]

Disability: Four Decades of Discovery. American Journal of Human Genetics. 2012;90(4):579-90.
11. Gecz J, Shoubridge C, Corbett M. The genetic landscape of intellectual disability arising from chromosome X. Trends in genetics : TIG. 2009;25(7):308-16.
12. Mundhofir FEP, Nillesen WM, Van Bon BWM, Smeets D, Pfundt R, van de Ven-Schobers G, et al. Subtelomeric chromosomal rearrangements in a large cohort of unexplained intellectually disabled individuals in Indonesia: A clinical and molecular study. Indian Journal of Human Genetics. 2013;19(2):171-8.
13. Coccaro EF, Lee R, Vezina P. Cerebrospinal fluid glutamate concentration correlates with impulsive aggression in human subjects. Journal of psychiatric research. 2013;47(9):1247-53.
14. Baker BL, Neece CL, Fenning RM, Crnic KA, Blacher J. Mental Disorders in Five Year Old Children With or Without Developmental Delay: Focus on ADHD. Journal of clinical child and adolescent psychology : the official journal for the Society of Clinical Child and Adolescent Psychology, American Psychological Association, Division 53. 2010;39(4):492-505.
15. McNally RJ, Calamari JE. Obsessive-compulsive disorder in a mentally retarded woman. The British journal of psychiatry : the journal of mental science. 1989;155:116-7.
16. Matson JL, Bamburg JW, Mayville EA, Pinkston J, Bielecki J, Kuhn D, et al. Psychopharmacology and mental retardation: a 10 year review (1990-1999). Research in developmental disabilities. 2000;21(4):263-96.
17. Simpson HB, Wetterneck CT, Cahill SP, Steinglass JE, Franklin ME, Leonard RC, et al. Treatment of obsessive-compulsive disorder complicated by comorbid eating disorders. Cognitive behaviour therapy. 2013;42(1):64-76.
18. Karno M, Golding JM, Sorenson SB, Burnam MA. The epidemiology of obsessive-compulsive disorder in five US communities. Archives of general psychiatry. 1988;45(12):1094-9.
19. Dong G, Lu Q, Zhou H, Zhao X. Precursor or Sequela: Pathological Disorders in People with Internet Addiction Disorder. PLoS ONE. 2011;6(2):14703.

[Downloaded from ijdo.ssu.ac.ir on 2026-11-20]