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Case report

Coexisting gender dysphoria, cross-dress and bipolar schizoaffective disorder: a psychopathological conundrum

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SUMMARY

The co-occurrence of Schizophrenic psychoses (with sex changes delusional experiences or themes) and gender dysphoria has been debated since the end of 19 th century with different accents: as an association of sexual psychopathy and paranoia, for the possible etiopathogenetic role of bi-sexuality and gender dysphoria in different psychic disorders, as psychopathologies that needed to be accurately differentiated, and, at last, as co-occurring conditions that can interfere each to the other.

The Authors describe in detail the clinical case of a 54 years old man who in relation with the periodic sharpening of his schizo-affective maniform psychosis, showed temporary shameless, grotesque and even bizarre cross-dressing behavior and sex re-assignment request. In euthymic or depressive phase, his transexual behavior was much less evident or even repressed. The psychopathological implications of this rare but impressive co-occurrence are discussed.

Key words: schizophrenia, gender dysphoria, cross-dressing, bisexuality, non-binary, sexual reassignment

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According to earlier epidemiological estimates about 1/37000 (0,0027%) males and 1/103.000 (0,00097%) females seek reassignment surgery [1]. The prevalence is getting higher and, in 2007, a Belgian study [2] found out values of about 1/12900 (0,0077%) in males and 1/33800 (0,0029%) in females. The percentage of people seeking sex surgery reassignment is nowadays increasing because of the progress in technical surgery, safe and successful today more than ever [3,4]. Clearly, the wider ethical acceptance and political support of gender-fluid patterns of sexuality makes their affirmation easier. Recent evidence shows ever higher rates of gender dysphoria in females (from 1/3000 to1/15000), overcoming the percentage in males [5]. However, the surgical sex reassignment and gender affirmation practices are ever more requested both from males to females than from females to males without any deep prior psychological and psychopathological assessments [4,6].

The prevalence of schizophrenic psychoses is about 1-2% in adults' people, and the prevalence of sex change delusions of sexual transformations in schizophrenia are not so rare as Krafft-Ebing [7] believed. According to Borras et al. [8], 20% of delusional experiences in schizophrenia have gender change or other genital modification's themes. The co-occurrence of schizophrenia and gender dysphoria, and even more of schizophrenia and other psychotic disorders with sex change delusions and sex gender reassignment should be considered very rare, but possible [9]. Hence, the literature on the co-occurrence is sparse and anectodic [5,10]. However, from a psychiatric standpoint, the relations between the two conditions

are difficult to disentangle [8,11,12] and represent a true psychopathological conundrum.

A brief recall to myths and literature

The transformation of a sex gender in another is a myth found in universal mythologies [13]. In Greek mythology, the most quoted sex change is that of Tyresisias (Τειρεσίας) that, in the most common tale, lived seven years as woman and seven years as a man (after having killed alternately the male and the female exemplary of a couple of snakes during their coupling). So, Zeus and Era called him as a judge to establish which sex would have felt more pleasure during sexual activities (Tyresias answered that women felt it nine times more than man).

Among the many literary example, we may quote Orlando, the character of Virginia Woolf [14], who changes sex in the centuries to meet different heroes of English literature, but the more pertinent for psychopathologic reflection is the main character of Roman Polansky's movie "The tenants" [15], drawn from Roland Topor's homonymous novel [16]. Trelkowski, the name of the character of a polish immigrate, played by Director Polansky himself, develops a delusional belief that people living in the same palace compel him to transform himself into the previous female tenant of his accommodation, who at last committed suicide jumping out of the window. In a growing dramatic climax of delusional and hallucinatory experiences, Trelkowski acts a complete cross-dress: he begins to wear female dress and uses the cosmetics of the earlier female tenant, then buys and wears female wig and heeled shoes before committing suicide dressed as his predecessor.

An experience of gender transformation is described also in Pier Paolo Pasolini's last, unaccomplished novel, "Petrolio" [17]: the great Italian writer was homosexual, but was deeply concerned with sexual paraphilias during his last year of life (see also his last film "Salò e le 120 giornate di Sodoma" – [18]). In the novel Pasolini tells two times the experiences of sex change of his main character "Carlo" who, in front of a glass, sees "the little sore that was his new sex".

Psychiatric literature

The psychiatric literature about psychopathological aspects co-occurring with gender dysphoria is not conclusive. Anyway a lot of delusional experiences of changing sex has been referred in the course of history of psychiatry.

Krafft-Ebing [7] described some cases in which the gender dysphoria and cross-dressing (*antipathic sexual instinct*) progresses in a psychotic conditions and called them *Metamorphosis sexualis paranoica.* In such circumstances the subject feels the sensation of *trasmutatio sexus*.

The most famous self-report of delusional sex-change (or gender transformations) experience is the one described by the remarkable Daniel Paul Schreber autobiography [19] "Memories of a nervous ill" (the case of the "President Schreber" analyzed by Freud directly on the basis of the writing – [20]): in his long hallucinatory paranoid experience, which needed two successive years long admissions in the Clinical Psychiatry in Lipsia ad in Sonnenstein Psychiatric Hospital between 1884 and 1895, Schreber, in the context of a wider well-systematized mystic experience felt two times his male genitalia transformed in female genitalia, thanks to "divine miracles". His description evokes up to date surgical procedures of sex reassignment: "The emasculation happened in this way: external male organs (scrotum and penis) were retracted inside the body and transformed in the corresponding female sexual organs, at the same time internal sexual organs transformed themselves…".

Until the end of the 20th century, most of psychiatrists believed that gender dysphoria was a delusional disorder (delusion of sex change): they were subtyped as delusion of no longer being a male or female, delusion of being a neuter or both sexes simultaneously, delusion of being of the opposite sex [21]; the last type was the most pertinent to discuss the relationships between trans-sexualism (gender dysphoria) and schizophrenia. In the last decades, some reports described isolated cases of co-occurrence of gender dysphoria and Paranoid schizophrenia [11,12,21-26]. They try to suggest that psychotic disorders should be excluded in people who ask for endocrine and chirurgical interventions of sex change, since sex change delusions are not so rare in clinical practice and self-reports of psychotic patients and can represent a subtype of somatic delusion and hallucinations [27]; these case of "pseudo-trans-sexualism" should be carefully differentiated from simple gender dysphoria beliefs [11,12]. These delusional beliefs can sometimes remit spontaneously or with antipsychotic medication [24,25]. Psychiatric diagnoses comorbidity with gender dysphoria has been studied in last years with opposite aims; Dhejne et al. [3] found no evidence of higher prevalence with respect to the general population, while the prevalence of substances and alcohol abuse, anxiety, social anxiety, alexithymia and mood disorders is much more relevant [3,9,28-31]. The suicidal risk is higher than in the general population [3,28] and suicidal risk and the occurrence non-suicidal self-injuring behavior are more common than in the general population as well as in gay, lesbian, and bisexual people [29-32]. Nonetheless, the updated prevalent paradigm is to consider change sex (sex change) ideas or drives as normal, if not associated with other psychotic symptoms or experiences [9]. Updated reviews reverse traditional models supporting that gender dysphoria conditions may be traumatic and contribute to the presentations of psychotic symptoms;

both hormone therapy and culturally-sensitive and gender-affirming approaches may help in their prevention and treatment [5,10].

Our interest in this topic rises up in our work (teamwork discussions) about the case of a fifty-four-years old bipolar schizoaffective male patient, who periodically present showy and even grotesque and manneristic cross-dressing that partially remits in course of antipsychotic treatments (haloperidol and clozapine), even if auditory verbal hallucinations persist. Remarkably, when the cross-dress behavior is at its acme, he gets in contact with specialized centers for endocrine and surgical sex reassignment. We tried to better understand his feelings reconstructing in detail his illness history in search of mutual interactions between gender dysphoria and psychotic disorders.

The clinical sketch

B. M. is a fifty-four-years old male that has been treated in our service discontinuously since fifteen years up to now, though more regularly during the last three years, due to the worsening of his psychopathology and behavior.

The early clinical disturbances referred by the patient are correlated to alcohol abuse in his late twentieth. He was treated in a public service until his recovery and alcohol abstinence. The onset of clearer psychiatric disorders is not well described by the patient, but was likely characterized by opposite mood oscillations. In the earlier stages of his illness, he consulted academic services and private psychiatrists, even though we could not find any trace of those treatments. The severe acoustic hallucinatory psychotic disorders have risen probably after his forties and have become chronic and neuroleptic resistant. In recent years, the course of the illness is characterized by alternation of maniform and depressive-like phases associated with reference delusions and auditory verbal hallucinations, especially malevolent comments of his acts, suggestions, and orders, so that a DSM-5 [1] diagnosis of Schizoaffective psychosis, bipolar type, continuous, has been consensually formulated.

The patient refers that doubts about his own sex gender arose in early childhood, but were repressed by his mother to protect him from his domineering father ("early-onset gender dysphoria" [1]). He doesn't refer sexual abuse but a single homosexual intercourse with an older boy at about ten years old.

His parents were merchants and the family was wealthy. Indeed, some unsuccessful attempts were made to push him to continue parental activities or to undertake any job whatsoever.

He has never lived with his family and, after his father's death when he was thirty-five, he has been living with his mother, which nowadays is eighty-years old upon writing. His sister lives autonomously and struggles to support and protect him.

He refers (in the phases of mood stabilization) to be shy and privacy-protective about his sexual life.

At about thirty-five, he has had a relationship with a woman, but he refers that, during sexual intercourses, sometimes, he was compelled to imagine that she was a little child. Therefore, he used to interrupt the intercourse, realizing his pedophile tendencies. Anyway, from this relationship, a male child is born, in virtue of the female's will, and is adolescent upon writing. The patient has not looked after his child for many years, due to the obsessive phobia of being a pedophile. However, he has maintained distant relations with him and his mother, and currently meets him at his mother's house.

He doesn't refer about any other relationship, neither heterosexual nor homosexual. During the phases of mood stabilization, he is not involved in sexual activities, and even he pretends to be asexual or non-binary.

The clinical picture changes drastically during maniform phases. He speaks continuously about his homosexuality and argues that his main source of sexual pleasure is a point of the rectus five centimeters inside his anus, looks for sexual activities with professional transsexuals and he's obsessed by cross-dressing activities.

In the last years he has been admitted sometimes in our psychiatric guard, even mandatorily, due to the sharpening of auditory hallucinations and aggressive behavior against his mother, this time following her refuse to give him money to buy very expensive female dresses. His insight is very low, both regarding his psychotic conditions, both his inappropriate, grotesque cross-dressing during maniform phases. He has developed the somatic delusion that the voices come from a device installed in his neck and he consulted a surgeon to remove it. Other paranoid ideas concern other persecutory themes. The response of these symptoms to drug treatments (haloperidol, clozapine, lithium) is quite low, and the course of the different phases follows spontaneous remissions.

Concerning his cross-dressing, during manic-like phases, his look is very impressive and astonishing: he dresses as a provoking prostitute, but the result is quite grotesque and obscene. For example, under the very short skirt he shows with evidence the remainders of his male genitals under the slip, and the hair of his breast stick out the pink top, matched with very expensive twelve-centimeters heeled pink shoes. Dressed as such, he is used to walk in his little town's streets and pubs completely untouched by judgments of the many people that know him. In these phases, he completely loses the sense of glamour that he would like to flaunt; moreover, he looks for homosexual passive intercourses with transexuals. At last, he took an appointment to begin the long process of surgical sex reassignment, starting with expensive private mastoplasty and buttocks rounding for which he gave a consistent advance

payment to the surgeon. In this last phase, he racked up debts for over than twenty thousand euros, so that his parents prompted for a juridical protection. The surgeon was mandated to give back the account.
Despite his cross-dressing, during manic phases he is sometimes uninhibited with young women and, at the end of the maniform phase, the cross-dress behavior remits and the patient tries to flaunt a more masculine behavior; in the past, he used to mimic more masculine behavior like driving expensive cars, looking for androgenic treatments and trying to have sexual intercourses with escorts.
In the stabilization or slightly depressive phases, he, as we have already written (as already mentioned), presents himself as a shy person, and defines himself as asexual, angelic, non-binary. He becomes critic about his transsexual behavior, especially for the moral damage he causes to his parents and his son.

Discussion and conclusions

The question of the relations between psychotic experiences, gender dysphoria and gender fluid disposition has a long tradition in psychoanalytical literature, beginning with Freud's repeated reflections and papers about the role of bisexuality in the genesis of "Hysteria" and paranoid delusions [33,34]. Due to their speculative nature, these kinds of observations have been increasingly outdated in the scientific literature and psychopathological reflections. However, the question is up to date not irrelevant since both the high rates of psychopathology, especially major depressive, phobias and adjustment disorders [30] in addition of the anticonservative behavior associated with gender dysphoria [28,31], as well the not so rare rates of sexual change themes in paranoid schizophrenics, still continue to rise it in clinical practice. Are there some psychopathological links between the two different conditions, psychosis and gender dysphoria? And, if yes, which?
The case we have described is quite interesting, for the intertwining of different psychopathological dimensions such as psychosis, obsessive phobias and abnormal personality traits, whose combinations with gender dysphoria and changes in sexual orientation during different phases of the patient's illness supported different, unstable, intriguing but also misleading clinical pictures. Our case study, that has astonishing similarities with that reported by Subbash Bhargava and Sujata Sethi [26], supports with clear evidence that schizophrenia and gender dysphoria with cross-dressing are independent conditions that can co-exist. In the patients history, schizophrenic paranoid symptomatology refers to non-sexual persecutory themes and, as well as the functional social impairment, has become chronic over the course of the years. However the mood oscillations influence pervasively the expression of gender dysphoria. Outside the manic phases, the patient still complaints of delusional themes but does not mention his gender-conflict. On the contrary, during the manic phases the gender conflict and the exorbitant cross-dressing tendencies are at their acme and represent the largest part of his psychopathology. The independence between psychosis, changes in sexual orientation and gender has been shown in the pioneering study by Hyde and Mckenna [29] on a pair of twins, which however did not exclude that the two conditions are two poles of a spectrum of disorders. Other explanations are: delusional and hallucinatory experiences can drag cross-dressing or desires to change sex, or, as in our case, can make the outing easier and their gestaltic and behavioral expression more pronounced. In our case, the cross-dressing during acute manic-like relapses of the illness was grotesque and shameless in its expression, quite different in its phenomenology from "normal" cross-dressing. Its theatrical expression might be considered a symptom of mania, though unrelated to hallucinatory experiences or verbal auditory hallucinatory orders that talk about other topics or simply comment patients' actions. Psychosis, then, could only modify the expression of gender identity disorders, as already suggested [5,22], making it more pronounced, completely void of glamour in its expression and sometimes bizarre in its subjective motivations. We can attribute these effects to a deep disinhibition of inner-locked dispositions.
The coexistence of psychotic disorders should always be investigated in people looking for sex reassignment surgery (as well as it should be in dysmorphophobic disorders in people looking for aesthetical or bariatric surgery). Apart from the cases in which transient delusional experiences supports them, the sex reassignment request can rise up but also be given up with the remission of expansive phases of bipolar or schizoaffective disorders, so that ethical and legal ("normative") questions can easily be put forward.

Conflict of interest statement

The Authors declare no conflict of interest.

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Author contributions

Both Authors contribuited to the clinical observation, the writing of the paper and the bibliographic research.

Ethical consideration

The research was conducted ethically, with all study procedures being performed in accordance with the requirements of the World Medical Association's Declaration of Helsinki.
Written informed consent was obtained from each participant/patient for study participation and data publication.

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