

Chronic Koro-like Syndrome (KLS) in recurrent depressive disorder as a variant of Cotard's delusion in an Italian male patient. A case report and historical review

Un caso di Koro-Like Syndrome (KLS) cronica, come variante di un delirio di Cotard, in un paziente italiano affetto da depressione monopolare cronica. Presentazione del caso clinico con revisione storica della letteratura

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SUMMARY. Cotard's syndrome is a delusional syndrome, first described in the 1880ies by Cotard, characterized by a nihilistic delusions about the self and/or the world. In some other cases there is an intense nihilistic belief that the patient's entire body or parts of it are disintegrated or dead. The syndrome is often associated with severe depression, but are also described neurological cases. *Koro* was described a little later from Asia and consisted in the belief that one's own genitalia are shrinking or disappearing and death will ensue thereafter, but there are many cultural variants and the syndrome may present in an incomplete form. We report on a KLS sharing more features with annihilation delusions, such as Cotard's syndrome. In KLS, the *délire de négation* may be limited to localized systems or organs. We believe that some complete and incomplete forms of *Koro*, when embedded in a depressive core, may represent a variant of Cotard's delusion. In fact, our patient did not reach a complete denial of his entire body, but rather focused on sexual identity. We analysed the psychosexual issues of our case according to Kretschmer's 1918 view of a "bipolar setting" between sthenic and asthenic characters of a patient suffering from sensitive delusions of (self-) reference. This view may allow us to relate the personological character to the genetic comprehensibility of the delusion.

KEY WORDS: Koro-Like syndromes, Cotard's delusion, recurrent depressive disorder, Kretschmer.

RIASSUNTO. La sindrome di Cotard consiste in un disturbo delirante descritto per la prima volta nel 1880 da Jules Cotard ed è caratterizzata da un delirio di negazione riguardante sia sé che il mondo circostante. Successivamente cominciarono a essere descritti altri casi in cui pazienti presentavano il convincimento delirante che l'intero corpo, o parti di esso, fossero morte o assenti. Detta sindrome è spesso associata a casi gravi di depressione maggiore, anche se sono stati descritti casi dovuti a malattie neurologiche. Il *Koro*, descritto in alcuni Paesi asiatici, consiste nel convincimento delirante che i propri genitali si retraggano nell'addome o scompaiano, con un'inevitabile morte che sopraggiungerà di lì a poco. Vengono però descritte molte varianti nelle diverse culture asiatiche rispetto a questo quadro principale, e la sindrome può anche presentarsi in forma incompleta. In questo lavoro presentiamo un insolito caso di KLS in un paziente italiano, e che presenta molte caratteristiche comuni al delirio nichilistico di Cotard. Riteniamo che sia le forme complete sia quelle incomplete di *Koro* possano rappresentare una variante del delirio di Cotard. Nel caso da noi descritto, il paziente non presentava una negazione globale del proprio corpo, ma una sensazione focalizzata sul tema della perdita dell'identità sessuale. In questo lavoro abbiamo inoltre analizzato l'aspetto psicosessuale nel quadro di riferimento del delirio sensitivo di rapporto proposto da Kretschmer nel 1918, e relativo alla costituzione "bipolare" tra caratteristiche personologiche sottostanti "steniche" e "asteniche" nella genesi di un delirio di negazione riferito a sé. Questa cornice teorica ci ha permesso di elaborare ipotesi sulle caratteristiche di personalità di base sottostanti a questo delirio.

PAROLE CHIAVE: Koro-Like Syndromes, delirio di Cotard, disturbo depressivo ricorrente, Kretschmer.

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INTRODUCTION

Cotard's syndrome is a delusional disorder introduced in the early 1880ies by the French neuro-psychiatrist Jules Cotard (Issoudun, Berry, 1st June 1840-Paris, 19th August 1889), who had worked with Charcot at the Salpêtrière in Paris. Cotard first presented a case of a "lypemanic" (with delusional depression) 43-year-old suicidal woman who believed her inside organs did not exist and who stated she needed no food, as she was immortal, but prompted others to burn her alive, as she believed fire was the only way she could give an end to her life (1). He coined the term "délire de négations" two years after (2), mistranslated into "nihilistic delusions" (3). In fact, the translation does not render the fact that Cotard's term was encompassing also other, non-delusional aspects of the syndrome, which comprised also anxiety and depression (4). The attribution of the syndrome to the man who had first described it systematically was made by Emmanuel Jean-Baptiste Joseph Régis (1855-1918) four years after Cotard's premature death (5).

Koro is a psychiatric illness characterized by acute anxiety accompanied by the belief that one's own penis (or vulva or nipples and breasts, in women) is shrinking and will dissolve in one's own abdomen, and by the fear that upon completion of this process, death will result (6). This syndrome has been variously viewed as obsessive-compulsive illness, anxiety neurosis, psychotic depersonalization and hysteria. It is a culture-bound syndrome most frequently occurring in South China, Malaysia and Indonesia, especially among people of Chinese origin. However, there are some rare case reports from the Western hemisphere.

In the DSM-IV-TR (7), *koro* is mentioned only in Appendix I (letter, not Latin number), in the context of culture-bound syndromes, as «a term, probably of Malaysian origin, that refers to an episode of sudden and intense anxiety that the penis (or, in females, the vulva and nipples) will recede into the body and possibly cause death. The syndrome is reported in south and east Asia, where it is known by a variety of local terms, such as *shuk yang*, *shook yong*, and *suo yang* (Chinese); *jinjinia bemar* (Assam); or *rok-joo* (Thailand). It is occasionally found in the West. *Koro* at times occurs in localized epidemic form in East Asian areas. This diagnosis is included in the Chinese Classification of Mental Disorders, Second Edition (CCMD-2)».

The complete Third Edition of the Chinese Classification of Mental Disorders (CCMD-3) (8) became available three years after the DSM-IV-TR, in both Chinese and English; *koro* is defined as «excessive fear in men of the penis shrinking or drawing back in to the

body. This has been associated with cultures placing a heavy emphasis on balance, or on fertility and reproduction». It may immediately be noted that the disorder is strictly confined to men, and it is broadened to encompass the concepts of ancient Chinese philosophy.

The ICD-10 (9) in its Mental and Behavioural Diseases, Diagnostic Criteria for Research manual, defines *koro* as «Acute panic or anxiety reaction involving fear of genital retraction. In severe cases, men become convinced that the penis will suddenly withdraw into the abdomen; women sense that their breast, labia, or vulva will retract. Victims expect the consequences to be fatal. Studies cite factors such as illness, exposure to cold, or excess coitus as precursors, but interpersonal conflict and sociocultural demands reportedly exert greater influence on the condition. Onset is rapid, intense, and unexpected. Responses vary, but include grasping of the genitals by the victim or a family member, application of splints or devices to prevent retraction, herbal remedies, massage, or fellatio».

It also suggests encoding it as "F48.8 Other specified neurotic disorders" or "F45.34 Somatoform autonomic dysfunction of the genitourinary system (may be used if autonomic anxiety symptoms are present)" and differentiating it from Indian "dhat" and "prameha" (10,11) or Taiwanese "shen-k'uei" (10) (acute anxiety and somatic complaints such as fatigue and muscle pain, related to a whitish discharge with urine, interpreted as loss of semen-like material in both men and women and attributed to excess coitus, urinary dysfunction, imbalance among body humours or diet) and Egyptian or other Arab "rabt (al azhâr)" (impotence or other sexual dysfunction due to sorcery or spell) (12).

The first medical reports on *koro*, a reportedly Malaysian word having to do with the tortoise's head, that may be retracted in its shell, were published in the mid-1890ies, while the term "lasa Koro", referring to dangerous penile shrinkage in Indonesian natives, had already been introduced in Western literature by Benjamin Frederik Matthes¹³ in his 1874 Buginese*-Dutch dictionary (quoted in Edwards) (14). However, the same syndrome was long known in China as *suo yang* (Mandarin), *suk yong* in Yue (Cantonese) or *shook yiang* in Wu (Shangainese) and was found in Emperor's Huang Di Classic of Internal Medicine (15). Upton (16) supports that the text was presumably not written by the Emperor himself, although he is celebrated as the father of Chinese medicine, because during these times (the Yellow Emperor is believed to have existed between 2697 and 2597 b.C.) it was cus-

*A language spoken by more than three million people, mainly in the southern part of Sulawesi, an Indonesian island.

tomary to attribute important writings to important people. At any rate, the compilation of this book began towards the end of the Chou dynasty (403 b.C. to 221 b.C.). The material is presented as a dialogue between the Yellow Emperor, who poses questions, and his physician and minister Qi Bo (Chi Po), who answers. In the *Nei Ching Su Wen*, there are sentences that might be attributed to *suo yang*, although the condition is not named as such. The Chinese believed the seminal fluid to be produced from blood in the Door of Life, a region supposedly located between the kidneys (Second Book, 6th Treatise). The kidneys, as functional units, were believed to be the same stuff with the testis. In Treatise 2 of the First Book it is twice stated that people who are disobedient towards the laws (principles) of Winter will damage their testis (kidney) and will suffer impotence during Spring. In the next Treatise, the damage to the kidney (testis) is believed to be brought about by excessive sexual activity, and there is a note by Wang Bing (Wang Ping), who edited the book, finishing it in 762 a.D., explaining that the method regulating sexual relations is the one of the wise men, who take care to match the Yin with the Yang, to preserve the Yang element and strengthen it. Other explanations, which are rooted in the Chinese culture and held to be valid for today's *koro* along with sexual activity, involve cold as a causal agent. Cold-induced feverish disease is held to pass through the genitals during the sixth day, resulting in a disorder of fullness and contraction of the scrotum (Ninth Book, 31st Chapter). In the same chapter, the Emperor recapitulates that when the disease of heat is caused by cold, the small Yang and the Absolute Yin are affected during the third day and ears become deaf and the scrotum is contracted and becomes deficient. Dietary measures are insufficient at this point and the patient is no longer able to recognise other people and will die during the sixth day. The notion that impotence or contraction of the scrotum may result in death is also found in the Second Book, 7th Chapter, where it is stated that a disease of the two Yangs will hit the heart and the spleen, and if neglected, will eventually result in loss of menses in women and insufficient semen production in men; should the disease continue, death cannot be avoided. This is the first instance in which we find in a "scientific" text the concept that loss of life-generating properties, structures and functions may ensue in death. Regarding the possibility to treat impotence, the *Nei Ching Su Wen* reports it to be treatable through acupuncture, among a host of other diseases, in Chapter 28, Eighth Book.

About the same times, penile retraction secondary to pain was somehow linked to danger of death by Hip-

pocrates (Kos, about 460 b.C. – probably Larissa, about 370 b.C.) in his writings: «A retraction of the testes and scrotum and penis are significative of severe pain and danger of death» (17). The issue of change or loss of genitalia was present in the Ancient Greek world, with Aristotle (384-322 b.C.) being quoted as having supported that «the principal reason for changing sexes is, and must be attributed to heat or cold» (Aristotle: The Midwife's Vade-Macum, Chapter X, a spurious work full of citations of people who had lived centuries apart) (18) and also demanded in his similarly spurious (19) "Problémata" «Why, who has fear, contracts his genitals?» (quoted) (20). It is remarkable that the Greeks had associated contraction of genital organs with feared death, turning into the opposite gender, and anxiety, all of them associated with classical *koro*.

In the Western World, the first account of penile retraction was on the *Lancet* (21); the author described the case of a man who saw a couple of years before, and was prompted to report the case because a similar case, though more severe, had been published in a Russian journal in the same year by Ivanov. In these two cases, shrinkage had actually occurred and was intensely anxiogenic. The first report of the culture-bound syndrome was in 1895 by Blonk (22), a military surgeon in Indonesia. The paper was in Dutch, the language that dominated the early literature on *koro*. The condition was viewed as an anxiety neurosis with hysterical features (23), or psychodynamically framed as castration fear (24). Although the element of delusional belief, often shared, that the penis will somehow disappear may lack from Western World cases, it is difficult to differentiate qualitatively the culture-bound syndrome from isolated Western cases.

Several authors attempted telling complete from incomplete forms of *koro* (25). In the incomplete forms, the belief that the penis will disappear into the abdomen or cause death is not present; however, the male patient is persuaded that he is impotent and that he lost his virility. *Koro*-like Syndromes (KLS) in Western countries are not distinct clinical entities, but represent a concomitant syndrome that requires treatment of the underlying illness.

We present a case of atypical KLS in which the patient presented with the delusion that his body would switch from male to female.

CASE HISTORY

C.M., a 58 year-old single male was admitted to our Psychiatric Hospital due to worsening of pre-existing symptoms of depression. On admission he was feeling de-

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pressed with loss of interest and drive, low self-esteem and guilt, feelings of inadequacy and of existential failure. The patient also complained of symptoms in which “everything” was over, and any solution would have been impossible to resolve his case, because he had lost not just possessions, but also the social status and power, as well as any ability to “be” in the world.

During a thorough psychiatric interview, he gave information about his illness and showed marked anxiety and fear about his penis, which he thought was gradually shrinking.

A few weeks prior to his admission, the patient felt that his penis was retracting gradually. He felt that as a result of this, he would become impotent, with no sperm, and that he would lose his gender. His belief that his body would change from male to female was supported with delusional intensity.

The patient was 18 when he first manifested concerns over his penis, which he believed will disappear, and that this would be followed by loss of his virility. He described himself as being much skinny at that time, with a poorly developed beard and no muscles. He had very low masculine self-esteem at that time. The symptoms waxed and waned for about 30 years. He lately started to grow a beard just to obtain a masculine self-image. During admission he believed that doctors would cut-off his penis to help him change his body from male to female. He eagerly sought assurance about his virility and often touched his abdomen, as he believed that some parts of the abdomen had lost function and also that he had serious physical illness.

He was the third of four children. He lived with his sister, who had been hospitalized for schizophrenia for few months. There were no other cases of psychiatric illness in his family.

His childhood was normal. He has always been shy, sensitive, polite, and had some difficulty making friends. As a child, he felt somewhat inadequate. At age 23 he abandoned his studies of architecture, after an academic failure. For the past 15 years, he had worked as a designer. Since the age of 25 he had been keeping an assiduous correspondence with many women from all around Europe and occasionally met them, but had no romantic relationships until the age of 30. Although he had some romantic involvement with women with whom he kept correspondence, at age 35 he stopped such occasional encounters because he believed that these women could perceive his loss of virility and masculinity.

The patient used minimal quantities of alcohol and did never abuse any substance. When he was 35 went to work in Libya, where he stayed for about 13 years. He was not specifically acquainted with the cultural beliefs of the Far East. He was first diagnosed as affected by major depressive disorder, recurrent type, at age 54. Since then, he was admitted to our hospital five times, about once a year. During these admissions, he had the terrifying perception that his penis was shrinking. This perception tended to subside

during the symptom-free interval of his recurrent depressive disorder. In many of these admissions the patient felt extremely inadequate and unable for routine activity, as he felt overwhelmed by even the easiest tasks. During his last admission, the delusion regarding his male-to-female body transformation presented for the first time. Tested on the Dissociative Experience Scale (DES) (26), he was found to be free from depersonalization and dissociation. On the short-form of the MMPI-2 he scored 94 on the D (Depression) scale (T-score) (normal T scores are considered those below 60). He also scored higher than normal on the Sc (Schizophrenia) (T-score=67) and Si (Social Introversion) (T-score=64) scales. These results, combined with those of the Structured Interview for the DSM axis II for personality disorders (SCID-II), which had been completed four year ago, confirmed the patient's premorbid personality, characterized by shyness, unassertiveness and some features of paranoia. Routine blood and urologic investigations were normal.

During his last admission, he was prescribed a drug combination consisting of an antidepressant (venlafaxine, 150 mg/day), an antipsychotic (olanzapine, 20 mg/day) and an anxiolytic (lorazepam, 7.5 mg/day). The patient was treated with this combination throughout his 30-day hospital stay. He was discharged with this treatment combination. At the one-month follow-up he was free from delusional ideation and depression. His Brief Psychiatric Rating Scale (BPRS) scores had dropped from 73 to 39. He provided written, informed consent for the publication of his case.

DISCUSSION

In our case, severe depressive illness led to disruption of perceptual continuity, determining a distortion in body image perception. Dissociative mechanisms, such as those leading to depersonalization, are compatible with distorted body image perception, that may lead to the perception of parts of the body as dysmorphic and dysfunctional. Depersonalization has been advocated to explain the pathogenesis of Cotard's syndrome (27), a mechanism similar to the one claimed to account for *koro* (28), however, we found no depersonalization in our case. If depersonalization is involved in this case, it should be partial. According to Berrios et al. (29), at least some delusions might be based on depersonalization experiences. Thus depersonalization might serve as a general “experiential substratum” which (modulated by different cognitive frames) will crystallize out into different delusional phenomena. In particular, it has been suggested that some forms of nihilistic or hypochondriacal delusions could have their origin in somatic depersonalization experiences, differently conceptualized by the psychotic patient.

While in KLS body-image disturbances may be limited to localized systems or organs, in Cotard's syndrome the disturbance usually extends to the whole body. In our case, fears over male-to-female body transformation does not mean discontinuity of one's own identity. Abnormality of body image may be the result of abnormal sensations (30). Our patient felt that he was impotent, with no sperm, and that if his penis retracted completely, he would change his gender and he would be "nothing". In fact, our case did not reach a complete denial of one's own body, but rather a selective change related to sexual identity; in this patient, the fear to be transformed into a woman could represent not an absolute discontinuity with respect to self-identity, but an inevitable corollary of the fact that on the face of a reduced perception of one's male identity there is an unavoidable transformation into the opposite sex. The delusional thinking linked to such psychopathological condition is in fact holothymically comprehensible, as described by Kretschmer (31) in 1918.

According to Kretschmer (31), the presence of an underlying mood disorder provides an acceptable explanation behind the body image disturbance. He argued that psychosexual conflicts led to sensitive delusions of (self-)reference. In his clinical cases of sensitive delusion of (self-)reference, the accent is on critical episodes caused by ethical-sexual conflicts. Human experience developing in the space between the self and the external world, according to this author, tends towards a "bipolar" pattern, swinging between two poles, represented by superiority feelings and high self-ideal (sthenic polarity) and those characterized by inferiority, discouragement and shame (asthenic polarity). In particular, Kretschmer's interest focuses on the psychological development of character, in which a sthenic disposition is subjected to the stimulant influence of a strong asthenic disposition that forms the opposite pole, or *vice versa*. In the first case we obtain expansive natures, whose most typical psychological manifestation is paranoia, in the second we obtain sensitive natures. Most often, perceived inefficacy and vulnerability are encountered at the background of people with strongly sthenic sensitivity. The same way paranoid people display a wide range of sthenic qualities, albeit with a hidden core of vulnerability, the sensitive shows a mainly asthenic constitution, which is however subject to a sthenic counter-reaction (31).

The internal experience of these patients is isolated; they jealously abstain from exteriorizing their feelings, which remain in a permanent tension state. They demonstrate much introspective power and insight, scrupulous morality and true altruism. We may hypothesize that the sensitive character, who usually lives in a

conscious way the conflict between grandiosity and shame, due to particularly traumatic experiences which threaten self-esteem, is forced to externalize this conflict between self-ideal and shameful inadequacy, thereby identifying his internal persecutory instances in another, external person. The feeling of self-cohesion is preserved by the angry-sthenic turnover of the breakdown caused by shame in a persecutory feeling. These subjects have an extreme sensibility, their spiritual life is isolated, their feelings remain tense, and they have a scrupulous attention to other people. In our case, these personal characteristics are well represented, mainly in their asthenic expression (feelings of inadequacy, shame) only partially counterbalanced by sthenic drives, as shown by a higher than normal score on the Pa (Paranoia) scale of the MMPI, by excessive scrupulousness, and by inadequacy feelings when facing everyday tasks.

Hence, our case bears some similarities with one case described in the mid-nineties by Wolff and McKenzie (32). These authors observed misidentification syndromes of both the Fregoli and the Capgras subtypes in a patient with shared psychotic disorder and *koro* during periods of depression. It could be possible to trace an analogy between our patient's "misidentification" of his own body and the misidentification of other people by the patient described by Wolff and McKenzie (32). Their patient could be viewed also according to Kretschmer's conceptualization, with sthenic instances promoting the paranoid, outward-directed misidentification, and asthenic ones being related with inward-directed misidentification, like *koro*-like syndrome, which should be viewed like a particular form of Cotard's delusion. In this sense we can find resemblances between the thought of Kretschmer and the modern theories of attributional style and its relationship with delusion content.

Attributional biases are *cognitive biases* which affect *attribution*, the way we determine who or what was responsible for an event or action. Such biases typically rely on *actor/observer differences*, and attribution theory provides a framework for understanding the causal explanations that individuals give for their own behaviour and the behaviour of others. *Normal subjects consistently demonstrate a self-serving attributional bias in explaining the causes of events*; that is, they tend to take credit for success (internal attribution of positive events: the "self-enhancing" bias) and to deny responsibility for failure (external attribution of negative events: the "self-protective" bias). *Such biases may serve to enhance self-esteem. Patients with persecutory delusions (with diagnoses of paranoid schizophrenia or delusional disorder), show an exaggeration of this self-serving attributional bias* (33).

In Capgras patients are thought to have a damage to neural pathways underpinning the emotional component of face recognition (34) and (35). The ensuing discordance between experiences of the way someone “looks” and the way they “feel” is thought to underpin the impostor delusion of these patients. Young et al. (36), have suggested that disruptions to the affective component of visual recognition may occur in Cotard cases as well as in Capgras cases. However, whereas Capgras patients interpret the resultant experiences in accordance with a paranoid, projective attributional style, Cotard patients interpret them in accordance with a depressive, introjective attributional style (37), like Kretschmer’s theoretical framework.

Our patient had a chronic course, supporting that sporadic KLS could be not a self-limited condition; our case is similar at this respect with the two cases from the East Indies, who were recently described by Kar (38).

It should be stated that our case does not bear all these features that render *koro* a culture-bound syndrome. He was poorly acquainted with cultural backgrounds of the Far East and had only a multi-year experience with Arabic culture. Although to date no *koro* case has been reported from Libya, some cases were described in Asian Arab patients (39,40) and patients from seven West African countries (41). However, it is unlikely that our patient could have been influenced by culture, since he had already developed his fear of penis retraction by the age of 18, long time before he visited a foreign country. Furthermore, our patient extended his uncertainties about his masculinity also to his beard, and this places him nearer to Cotard’s delusion rather than to classical, culture-bound *koro*. Psychiatrists should be more sensitive to their patients’ significance of their symptoms and to other coexisting symptoms to distinguish classical *koro* (in which, for instance, the patients may use physical maneuvers to prevent penile retraction, and many times believe they will die as consequence of Koro), from possible cases of KLS with higher affinity for Cotard’s delusion and depression, and treat them accordingly. In this regard it is important to point out the differential diagnosis of non-Asian Koro like cases with major depression (42), schizophrenia (43), bipolar disorder, as well as neurological disorders with psychiatric comorbidity, as epilepsy, brain tumors and Parkinson disease (4). It is interesting at this respect, that whereas *koro* is usually treated with antipsychotics, two cases of *formae frustae* of *koro*, one in a Caucasian (44) and one in a Japanese patient (45) were successfully treated with selective serotonin re-uptake inhibitors.

CONCLUSIONS

Our case is a KLS sharing more features with annihilation delusions, such as Cotard’s syndrome, rather than the culture-bound syndrome which we intend as *koro*. This is an only partial annihilation delusion, which shows polarity, as shown by the fear of transformation into the opposite gender. Such polarity recalls Kretschmer’s framing of sthenic and asthenic characters, which best suit our case. The intense emotional turmoil could affect consciousness by producing altered perceptual constancy, which in turn would result in delusional perception of body or one part of it. In our case, perceived sexual inadequacy characterizes the entire existence of the patient and becomes delusional upon worsening of depressive symptoms.

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