EVERLASTING FIRE - PERSISTENT MANIA: A CASE REPORT
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INTRODUCTION

Only ash remains after fire is gone. Metaphorically mania is the fire and depression its ash (Koukopoulos & Ghaemi 2009). What is the reason, when there is just fire without ash? Unipolar mania has been described in the literature. Different terms have been used like recurrent mania, chronic mania and persistent mania. But these are scarce diagnoses in psychiatric routine. This topic has got little attention in research of affective disorders, but cases have been reported over the time (Maihi et al. 2001, Solomon et al. 2003, Kraepelin 1921). To fulfill the diagnosis of chronic mania, prominent manic symptoms for more than two years without remission are required (Hare 1981). But no consistent definition of this disorder is available in literature. A contentious issue related to this topic is, if unipolar mania is a separate entity (Harish et al. 2005). Yazici et al. showed, that unipolar mania differs significantly from bipolar disorders e.g. in an earlier onset of illness, a higher presence of psychotic features, a higher rate of hyperthyemia, smaller rate of suicide attempts and less response to lithium prophylaxis (Yazici et al. 2002). Further data showed significant differences in a lesser social disability, a lesser anxious worries, and lesser hostility (Perugi et al. 2007). To our knowledge there is no consistent theory explaining the pathomechanism of unipolar mania.

CASE REPORT

We report on a 46-year-old female patient with bipolar disorder, which has been diagnosed for the first time in 1990. Since that time our patient has been in periodical treatment. Her psychiatric history included multiple episodes of mania including psychotic symptoms with subsequent hospitalisations in the past. CT and MRI scan of the brain revealed no pathology, repeated drug screenings never showed positive results, family history concerning mood or psychotic disorders were negative. Between 1990 and 2013 we registered 62 admissions because of mania with aggressive behaviour. Medical treatment included fluphenazine, haloperidol, valproate, carbamazepine, lithium, risperidone, chlorprothixen, haloperidol, zuclophentixol, olanzapine and various benzodiazepines. From the beginning of treatment full remission could never be achieved, no matter how long she stayed at hospital. At least the symptoms declined to a mild form of hypomania. But after some years medication hardly affected the mania. During the protracted hospitalisation physical restraints were repeatedly necessary. The patient’s acceptance of illness was very limited, our treatment proposal was declined most of the times. Even the use of depot medication did not improve the situation. In 2007, after 17 years of psychiatric treatment history and 2.5 years of treatment with lithium TSH was depressed to 0.03 uU/ml (reference value: 0.23-4.00 uU/ml) for the first time. In the beginning T3 and T4 were within normal limits, but considerably increased in repetitive blood samples over the coming years. An associated increase of serum antithyroid antibody concentrations beyond reference value was never detected. A radionucleotide scan of the thyroid showed a multinodular goiter with suspicious autonomy which is an indication for a strumectomy. Lithium was stopped at this time and thiamazole was started, first with 5 mg per day, later increased to 20 mg per day. Due to the bad medical-adherence a strumectomy did not seem possible, since after such an operation substitution of thyroid hormone for life time would have been inevitable. And we could not assume our patient would follow our recommendations. Thus, the only possible treatment could be a radio-iodide-therapy. But despite all our efforts, we could not persuade her to accept this therapy. She even stopped the medication every time she left hospital. So she continued to be in a state of hyperthyroidism throughout the years.

Interestingly, we never saw our patient in periods of depression. Even the patient’s general practitioner and the social worker, who visited the patient at home at a regular basis, never noticed depressive symptoms and hardly states of balanced affective condition. During the past four years she spent more time at our hospital than outside. Nearly all the time she has shown pronounced symptoms declined to a mild form of hypomania. But matter how long she stayed at hospital. At least the treatment full remission could never be achieved, no matter how long she stayed at hospital. At least the symptoms declined to a mild form of hypomania. But after some years medication hardly affected the mania. During the protracted hospitalisation physical restraints were repeatedly necessary. The patient’s acceptance of illness was very limited, our treatment proposal was declined most of the times. Even the use of depot medication did not improve the situation. In 2007, after 17 years of psychiatric treatment history and 2.5 years of treatment with lithium TSH was depressed to 0.03 uU/ml (reference value: 0.23-4.00 uU/ml) for the first time. In the beginning T3 and T4 were within normal limits, but considerably increased in repetitive blood samples over the coming years. An associated increase of serum antithyroid antibody concentrations beyond reference value was never detected. A radionucleotide scan of the thyroid showed a multinodular goiter with suspicious autonomy which is an indication for a strumectomy. Lithium was stopped at this time and thiamazole was started, first with 5 mg per day, later increased to 20 mg per day. Due to the bad medical-adherence a strumectomy did not seem possible, since after such an operation substitution of thyroid hormone for life time would have been inevitable. And we could not assume our patient would follow our recommendations. Thus, the only possible treatment could be a radio-iodide-therapy. But despite all our efforts, we could not persuade her to accept this therapy. She even stopped the medication every time she left hospital. So she continued to be in a state of hyperthyroidism throughout the years.

Interestingly, we never saw our patient in periods of depression. Even the patient’s general practitioner and the social worker, who visited the patient at home at a regular basis, never noticed depressive symptoms and hardly states of balanced affective condition. During the past four years she spent more time at our hospital than outside. Nearly all the time she has shown pronounced mania mostly with psychotic symptoms. Only twice we could detect mixed states with manic and depressive symptoms, but each time only lasting some hours before switching again to pure mania without any medical intervention. The best we could achieve up to now were intermittent periods of hypomania alternating with severe mania.
DISCUSSION

Patients with bipolar disorders usually spend more time suffering from depression than mania or hypomania. Recurrent manic episodes without depressive states are mentioned in ICD 10 as a residual category under F31.89 ‘Other bipolar affective disorders’. Single manic episode is listed too in ICD 10 under F30.x, but recurrent episodes of depression, mania or hypomania are excluded here. In DSM V hypomanic episodes without prior major depressive episode is listed under 296.89. But recurrent manic episodes without major depression is not included. Searching the literature on unipolar mania we could find several papers describing the difference between bipolar disorder and unipolar mania (Harish et al. 2005, Perugi et al. 2008). But we could not find an exact differentiation between the terms chronic mania, recurrent mania and persistent mania.

Our patient started with a form of unipolar manic disorder, which we would describe as recurrent. Her states changed between mania with psychotic symptoms to almost normal affective states with mild hypomanic signs under medication, but never to complete affective normality regardless of treatment duration. The initial diagnosis of bipolar disorder from 1990 was probably wrong. But we cannot be sure, whether she used to have depressive episodes in between the manic states before 1990. Over the years she developed from recurrent mania towards a persistent manic episode. This course of illness is not described in ICD 10 or DSM V. How can we explain the nearly nonstop manic state of our patient, which is hardly influenced by medication? The only physical abnormality we could find related to multinodular goiter and latent or manifest hypothyroidism. The effect of thyroid impairment influencing affective states is beyond dispute, even if the exact mechanisms as well as quality and quantity of the impact on mood is not yet clear and discussed controversially. In one case report recurrent mania was linked with recurrent hyperthyroidism (Corn & Checkley 1983). But in our case the thyroid impairment could at the most explain the aggravation of the course of illness during the last years, e.g. the transformation of recurrent to persistent mania. Still a change of diagnosis into an organic manic disorder due to ICD 10 or a bipolar and related disorder due to another medical condition in DSM V is not justified, since the unipolar disorder was preexisting. Unfortunately, the needed treatment of the multinodular goiter could not be arranged for our patient due to lack of insight and adherence of our patient and the legal regulations. We found no indications for other syndromes that could possibly have caused the described symptoms like encephalitis or drug abuse.

We were surprised, that despite high doses of antimanic, antipsychotic and sedative medication the symptoms could be hardly influenced. Of course, high-dose sedation showed short term effects on drive and sleep, but only for a few hours. Interestingly, a review by Yazici et al. (Yazici et al. 2002) found patients suffering from recurrent unipolar mania to be less responsive to lithium in comparison to patients with bipolar disorders. Others have described the difficulties regarding resistance to treatment in chronic mania (Grover et al. 2012).

CONCLUSIONS

To our understanding and in accordance with established doctrine sooner or later the manic state should have turned into depression or at least into a balanced affective state. If only ash remains after fire, when does this fire go out? Further studies of this topic should clear this unsolved issue.

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Conflict of interest:
Independently of this paper Kurosch Yazdi has commercial associations with Janssen Cilag and Lundbeck as lecturer. Barbara Pischinger has commercial associations with Lundbeck as lecturer. Jan Rosenleitner has no commercial associations.

References


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