Case Report

Sudden onset Cough like spasms – a case report

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ABSTRACT

Sudden cough like spasms may be a manifestation of movement or tic disorders. We present herewith the case of 9 year old with cough like spasms that responded to Clonidine, Haloperidol and Risperidone. The unique features of the case and its treatment and diagnosis are discussed.

Key words: tics, movement disorders, spasms, cough.

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INTRODUCTION

Movement Disorder (MD) most commonly found in children is Tic Disorder (TD) [1] but when compared to other medical illnesses of childhood it forms a very small percentage. Thus it becomes challenging to establish the correct clinical diagnosis and management when TD mimic other more common medical illnesses (like can be confused with cough of Respiratory Tract Obstruction/Infection as in the present case). The clinician's difficulty further increases if there exists a clear stressful psychological event before the onset of MD, because literature review suggests a high prevalence of medically unexplained psychogenic origin somatic complaints in primary care settings [2-3]. This thus necessitates a careful study of multiple factors so as to differentiate between Psychogenic Movements Resembling Tics (PMRTs) and Tic Disorder (TD) per se. PMRTs are movement disorder characterized by psychogenic origin with no organic cause and refractoriness to medical management [4-5], while TD is characterized by a neuro-biological basis of origin with good response to medical management [6]. Confusion arises because many TD have an underlying psychogenic cause, both TD and PMRTs share common features and both can coexist in the same patient [7-9]. Thus Physicians, Pediatricians and ENT specialists who are the first doctors to encounter such cases in clinics and hospitals in India need to have full awareness and vigilance in respect to this illness & the closest differential diagnosis.

Traditionally literature has emphasized the importance of clinical observation and phenomenology in differentiating various movement disorders [10]. Some observable differences among PMRTs and TD patients with respect to their clinical history, clinical examination and response to treatment are as follows. In clinical history PMRTs in contrast to TD have a female preponderance, sudden onset, a comparatively older age of presentation with lack of family history of similar illness. Tics on the other hand are more common among children/adolescents when compared to adults [11] and amongst the recent onset tics in children the most common ones are those having a primary origin [12]. On clinical examination most patients with PMRTs in comparison to TD are not able to voluntarily suppress the movements, even transiently. While these same patients may partially or completely stop the movement if distracted or

provided with a suggestion [13]. Tics on the other hand decrease or stop completely when the patient is given a goal directed behavior requiring focused attention and they increase during leisure times [14-15]. Also evident on clinical examination in most patients with PMRTs is the presence of other psychogenic movement disorders or psychogenic seizures. From treatment perspective, PMRTs do not respond well to Dopamine Receptor Antagonists while the Tics do [16].

Literature analysis for recent-onset tics in children is difficult because of discrepancy in the criteria set and defining features. There has always been a dilemma regarding the place of recent onset tics in respect to chronic tic disorders. Some believe in categorical classification taking both as two different disorders [17-20], while others advocate a spectrum based classification with both lying on a continuum [21-24]. DSM - 5 defines tics of primary origin with onset less than a year ago and before 18 years of age as Provisional Tic Disorder (PTD) [25-26]. DSM - 5 also abandons the term psychogenic cough and replaced it with somatic cough as recent evidence shows cerebral correlates for previously thought pure psychogenic disorders [25-28].

Caveat also exists in literature when we talk about the causative factors for PTD. Although unconfirmed but the interaction among psychological, neuro-biological, genetic and environmental factors are the currently believed etiology for PTD. Their interaction is assumed to cause neurotransmitter dysregulation in cortico-striato-thalamo-cortical circuits. Dopaminergic overactivity in basal ganglia causes subcortical disinhibition and thus impairment in automatic movement control which clinically manifest as motor and/or vocal tics [29-30]. Although these tics are manifestation of the assumed neurotransmitter dysregulation in brain, the majority data on PTD suggest a good prognosis with improvement within a few months. Literature evidence supports a high spontaneous remission rate in PTD [31-32]. Provisional Tics (previously called Transient Tics) are most frequently simple motor tics while phonic transient tics are extremely uncommon [33].

In general, tics may have extremely benign to extremely severe phenomenology. Those that are commonly encountered in clinical practice are the severe ones which are socially and functionally incapacitating for the patient [34]. Thus prompt diagnosis & urgent management is of utmost importance. But certain tics, ex. cough like spasms are notoriously difficult to be correctly diagnosed and are usually missed by doctors in routine day to day practice until late after multiple unnecessary investigations and failed treatments. In this research paper we have thrown light on one of such difficult to diagnose case and have discussed the various related aspects.

CASE REPORT

A 9-year-old male child was referred to psychiatry department of our hospital in view of sudden onset, almost continuous, anti-tussive refractory, non productive coughing. Detailed evaluation revealed that the child suffered fall over head 10 days ago with unconsciousness for 2 minutes. Thereafter the child was playful and thus was not taken immediately for any medical evaluation. One day later the child began to complain about uneasiness and thus was taken to a physician who prescribed him multivitamins and a dextrose drip. After the child was taken back home, soon within next 6 hours he began with sudden onset, recurrent cough like spasms and thus was taken back to the same physician who referred him to a pediatrician. Pediatrician prescribed him anti-tussives, nebulization and bronchodilator medications over which the child perceived minimal improvement. Thus on follow up visit to the pediatrician a CT Scan was done which showed clinically insignificant mild cerebral edema. The child was referred to a neurophysician who again prescribed medications (anti-tussives) with minimal improvement.

As the cough like spasms were seen almost continuous, it was causing the child pain and discomfort. It is when the child was referred for a higher center, the parents decided to visit Kiran Super Multispeciality Hospital. The child was evaluated by the Psychiatrist, ENT Specialist, Pediatrician and admitted under psychiatry department in view of marked distress and requirement of a detailed work up. There was no history of use of substance or dopamine depleting medications and complete neurological examination was in normal limits, except for cough like spasms. There were no abnormal involuntary movements of other body parts with no personal or family history of any movement disorder or other psychiatric disorders in past. The child did not report of premonitory sensations but was seen to be able to voluntarily

stop the spasms for a few seconds, after which the coughing spasms would come with a greater force. The spasms were seen to decrease when he would focus on playing games on mobile phone and increase when he sits doing nothing. They were also seen to increase when staff people or any relative were seen around him, while completely stop when fast asleep. Two differential diagnosis, namely PMRT and TD were made for the child's condition. Thus the child was given a trial of Tablet Haloperidol 2.5 mg/day, Tablet Clonidine 0.15mg/day and Tablet Trihexiphenidyl 2mg/day. Total improvement with no cough like spasms within one day of starting D_2 Receptor Antagonist & Alpha-2 Receptor Antagonist helped us to confirm PTD as final diagnosis. But the child developed Extrapyramidal Side Effect (EPS) involving Acute Dystonia of neck and eyeballs for which he was given one intramuscular injection of promethazine 25mg and was shifted from oral Haloperidol 2.5mg/day to oral Risperidone 1mg/day. Response on treatment was maintained with no cough like spasms and thus after one day of observation the child was discharged on oral medications. Thereafter on monthly follow-ups the improvement is maintained till date (2 months following discharge from hospital).

DISCUSSION

The child described in this case report had a few days (less than one year) history of cough like spasms with onset before age of 18 years and no identified organic cause on multiple investigations. The diagnosis of PTD & decision to treat the patient with D_2 Receptor Antagonists was taken on the basis of clinical history, clinical symptomatology, minimal improvement on anti-tussives and profound distress by recurrent cough like spasms. The case clearly met the DSM- 5 diagnostic criteria for PTD.

Through this case report we bring to notice the diagnostic dilemma faced by clinicians while managing an 11-year-old male patient with sudden onset recurrent cough like spasms. Clinician's dawdle in having a differential of movement disorder in the current case invited unnecessary investigations and delay in appropriate timely management causing a marked increment in distress and suffering to the patient and his relatives. We emphasize that clinical suspicion could be crucial for timely diagnosis.

The clinical case commentary highlights the fact that the presence of psychological issues does not rule out TD and that these may also be the contributing factors in its pathogenesis. Sensitivity of these patients to their perceptions about their internal self and the external world underscores the contribution of psychological factors. Caution should always be practiced while confirming the causality so that appropriate referral & treatment can be done. Clinical history, symptomatology and improvement on D_2 Receptor Antagonists helped us to ascertain the diagnosis of PTD. Appropriate and prompt diagnosis of these cases is critical as the treatment paradigm depends on it.

Not always the tics in PTD are benign, rather can be significantly disruptive causing severe distress to the patient. They can generate excessive and disproportionate worrying thoughts and emotions among the close ones of the patient. Thus some PTD cases can't be left untreated for spontaneous remission. They may need urgent attention and pharmacological intervention.

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