

# Psychogenic Dystonia in Tunisian Children

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## ABSTRACT

Psychogenic dystonia in children is rare and often difficult to distinguish from organic dystonia. It is usually related to a psychological or psychiatric underlying cause. From January 2004 to November 2009, 5 children with psychogenic dystonia among 200 with dystonia were followed up in our department. Elements of history, physical examination, videotaping and management were analyzed. Mean age was 14.9 years, mean age of onset was 13 years and mean follow up period was 6 months. The dystonia onset was abrupt in 3 patients and progression resulted rapidly into fixed dystonia in 4 patients. Pain was observed in all patients. Paroxysmal dystonia was observed in one patient. An underlying psychiatric disorder was found in all patients. All patients improved with psychotherapy and anxiolytic or antidepressant drugs. Only one patient showed relapse after each familial conflicts. The small size of our series reflects this disorder is rare (1 case/year). Pain was a prominent feature in all patients. Children have acute onset, short duration of disease and improved under psychological therapy and drugs. Psychogenic dystonia in children is usually misdiagnosed. It is necessary to analyze clinical features and outcome of this disorder to reach a clear diagnosis and adequate management, which requires multifaceted approach, including psychological, physical and pharmacological therapies.

**Keywords:** Dystonia; Psychiatric Disorders; Childhood

## 1. Introduction

Psychogenic movement disorders (MD) are movement disorders that cannot be explained by organic lesions and commonly associated with emotional “functional” signs [1]. Fahn and Williams proposed a classification system for psychogenic dystonia [1]. However psychogenic dystonia remains difficult to diagnose and particularly in children. We describe 5 Tunisian children with psychogenic dystonia and compare our findings with literature.

## 2. Methods

From January 2005 to November 2009, 5 out of 200 cases of dystonia, followed up at the Department of Child and Adolescent Neurology, were diagnosed with psychogenic dystonia. All 5 patients were admitted in our department from emergency neurological unit.

All of them underwent neurological examination, videotaping and psychological test follow up. Laboratory studies including copper test, peripheral blood stream and organic acid chromatography were made in all the patients. All patients underwent Brain MRI. The diagnosis of documented psychogenic dystonia was made according to the criteria of Fahn and Williams (1988). Treatment was decided according to the underlying psychiatric disorder.

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We performed a web search by using the NIH pubmed database with the following keywords “psychogenic dystonia in children” and found few studies reported on psychogenic dystonia in children [1-12].

## 3. Case Descriptions (Table 1)

### 3.1. Case 1

K. H is a 14 year-old-girl, born to non consanguineous parents. Her father had schizophrenia. At the age of 14, she had sudden onset of cervical movement disorder. Neurological examination showed a fixed and painful left torticollis with no other neurological signs. She was free of symptoms when she left alone. Psychological tests showed depression. Laboratory studies including copper tests, peripheral blood stream and organic acids chromatography were normal. Brain MRI was normal. The diagnosis of documented psychogenic dystonia was made. She underwent psychotherapy with complete remission over 24 hours.

### 3.2. Case 2

Z. F is an 18 year-old-boy, born to non consanguineous parents with no family history of neurological or psychiatric disorders. At the age of 14, he demonstrated a progressive, fixed and painful trunk deformity (lateral-deviation). He was not able to walk but he could feed himself and

**Table 1. Main clinical particularities, outcome and treatment of our patients.**

Patients	1	2	3	4	5
Sexe	F	M	F	M	F
Age (years)	14	18	14	11	17 $\frac{1}{2}$
Age of onset (years)	14	14	10	10	17
Family history	Schizophrenia	N	N	N	N
Onset distribution	Cervical	Trunk	Left hand	Left foot	Cervical
Onset	Acute	Progressive	Acute	Progressive	Acute
Type of dystonia	Fixed	Fixed	Paroxystic	Fixed	Fixed
Pain	Y	Y	Y	Y	Y
Outcome distribution	Focal dystonia	Generalized dystonia	Left hemidystonia	Left hemidystonia	Segmental dystonia
Neuropsychological test	Depression	Depression	Conversive disorders	Conversive disorders	Conversive disorders
Investigations	Normal	Normal	Normal	Normal	Normal
Treatment	psychotherapy	Anti-depressive drugs psychotherapy	Anxiolytic psychotherapy	Anxiolytic psychotherapy	Anxiolytic psychotherapy
Outcome duration	24 hours	12 months	ND	6 months	6 months
Relapse	N	N	ND	N	Y

M: male; F: female; Y: yes; N: no; ND: not determined.

write. Neurological examination showed isolated, fixed and painful generalized dystonia (trunk, upper and lower limbs). He was free of symptoms when he was unobserved. Psychological tests showed depression. Laboratory studies including copper tests, peripheral blood stream and organic acids chromatography were normal. Brain MRI was normal. The diagnosis of documented psychogenic dystonia was made. He underwent psychotherapy and anti depressive drugs (clomipramine 10 mg/day) with complete remission over a period of 12 months.

### 3.3. Case 3

A. T is a 14 year-old-girl, born to non consanguineous parents with no family history of neurological or psychiatric disorders. At the age of 10, she demonstrated an acute and paroxystic painful deformity of left hand. She was able to walk, feed herself and write. Neurological examination showed isolated, fixed and painful left hemidystonia. This movement disorder was accentuated when her parents were present. She was with free of symptoms when she was unobserved. Psychological tests showed conversive traits. Laboratory studies including copper tests, peripheral blood stream and organic acids chromatography were normal. Brain MRI was normal. The diagnosis of documented psychogenic dystonia was made. She underwent psychotherapy and anxiolytic drugs (meprobamate 400 mg/day) with complete remission.

### 3.4. Case 4

L. M is an 11 year-old-boy, born to non consanguineous parents with no family history of neurological or psychiatric disorders. At the age of 10, he demonstrated a discrete equinus of the left foot. This deformity spread progressively to left upper limb. He was not able to walk but he could feed himself and write. Neurological examination showed isolated, fixed and painful left hemidystonia. He was free of symptoms when he was unobserved. Psychological tests showed conversive disorders. Laboratory studies including copper tests, peripheral blood stream and organic acids chromatography were normal. Brain MRI was normal. The diagnosis of documented psychogenic dystonia was made. He underwent psychotherapy and anxiolytic drugs (meprobamate 400 mg/day) with complete remission over a period of 6 months.

### 3.5. Case 5

A. E is a 17<sup>1/2</sup> year-old-girl, born to non consanguineous parents with no family history of neurological or psychiatric disorders. At the age of 14, she had peripheral facial palsy. At the age of 17 years, she demonstrated an acute facial and cervical movement disorder. Neurological examination showed a fixed and painful facial dystonia and left torticollis (**Figure 1**) with no other neurological signs. She was free of symptoms when she was unobserved. Psychological tests showed conversive traits. Laboratory



**Figure 1. Fixed facial dystonia and left torticollis.**

studies including copper test, peripheral blood stream and organic acids chromatography were normal. Brain MRI was normal. After placebo treatment (intramuscular injection of physiological serum) and suggestive symptoms disappeared. The diagnosis of documented psychogenic dystonia was made. She underwent psychotherapy and anxiolytic drugs (meprobamate 400 mg/day) with complete recovery over a period of 6 months, dystonia relapsed after each familial conflict.

#### 4. Discussion

We report the result of a clinical based study on 5 Tunisian patients with psychogenic dystonia. During a follow up period of 5 years, our center gathered 200 cases of dystonia. Only 5 (3 girls and 2 boys) among them had a documented psychogenic dystonia (2.5% of all dystonic cases) (95% CI).

Psychogenic dystonia is rare particularly in children. It was first described in 1988 by Fahn and colleagues, who reported 39 patients, 17 of whom were categorized as having a documented dystonia [1]. In this series, only 7 of them had early onset (18 years) [1]. Ferrara *et al.* reported on 54 patients with psychogenic movement disorders mostly with dystonic features (median age 14.2 years) [2]. According to Ferrara's results, the median age of onset of our patients was 13 years (range 10 - 17) and the median age at diagnosis was 14.9 years (range 11 - 18).

All our patients had variable dystonic features (onset, topography and progression of dystonic features). Most common site of dystonic posture onset in psychogenic dystonia is the foot [2-4]. Our patients had variable dystonic onset site: foot (case 4), hand (case 3), cervical dystonia (cases 1, 5) and axial (case 2). In 4 of our cases, dystonia progressed over time: from the foot to the ipsilateral hand, becoming hemidystonia in 2 patients and generalized in 2.

Important clues suggesting the psychogenic of a movement disorder are: movement usually, with sudden onset, fixed and painful [3-8]. All our patients had some of these pictures: abrupt onset (case 1, 3, and 5), fixed posture (case 1, 2, 4, and 5) and painful (case 1 - 5).

Psychiatric disturbances are commonly associated to psychogenic dystonia. In children, conversive traits are frequent (more than 33%) predominately in girl [2-4,7]. These symptoms represent an attention seeking behaviour (secondary gain) [11]. Others psychiatric disturbances were associated to psychogenic dystonia such as personality disorders and depression [1]. All our patients had psychiatric disorders: conversion (3 patients) and depression (2 patients).

The prognosis of psychogenic dystonia is most favourable in patients when the diagnosis and a multidisciplinary approach are made early [11,12]. Our patients received psychological assessments, treatment with complete recovery and only one patient had relapse (case 5). Long multidisciplinary approach is compulsory in psychogenic dystonia to avoid relapses and to all the best possible prognosis [12].

#### 5. Conclusion

Psychogenic dystonia is rare in children and is clinically similar to adults. It should be evoked when dystonia is painful and fixed with associated psychiatric disorders. Treatment should include behavioral, psychological, physical and pharmacologic therapies to have best prognosis.

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