



Functional movement disorders of the face: A historical review and case series



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ARTICLE INFO

Keywords:

Functional Neurological Disorders
Facial
Dystonia
Spasm
Psychogenic
Conversion Disorder
Functional Movement Disorders

ABSTRACT

Functional facial dystonia or spasm has, in recent years, been recognised as a relatively common form of functional movement disorder. We describe historical ‘forgotten’ neurological literature indicating that this was a well described phenomenon in the late 19th and early 20th century but subsequently faded from awareness. We add data from our own series of 41 patients with functional facial dystonia to explore the clinical features and associated comorbidities of patients with this movement disorder.

1. Introduction

The last decade has seen increasing awareness of the fact that functional movement disorder may affect the muscles of the face. An initial report on four patients by Tan and Jankovic [1], was followed by scattered reports [2,3] prior to a seminal case series of 63 patients by Fasano et al. [4,5]. Subsequent small series [6] have all added to the characterisation of functional facial movement disorders which were defined by Fasano and Tinazzi as a dystonia with fixed unilateral facial contraction, usually involving the lower lip and often with ipsilateral orbicularis oculis and jaw involvement. They are often of maximal severity at onset and display inconsistencies on clinical examination, such as resolution with distraction and changes in side and pattern during or between examination or spontaneous remissions [5].

The history of the field has not been one of linear accumulating knowledge. ‘Hysteria’ was a core part of neurological textbooks in the 19th century but it gradually lost its popularity as a subject of neurological study over the course of the 20th century [7,8]. Consequently, a considerable number of useful but older clinical descriptions have been forgotten. Much of our apparently new knowledge in this field revisits clinical experience that had been documented in the past.

Fasano et al. noted that patients diagnosed with atypical facial movement disorders in studies going back to 1986 also probably fulfilled the diagnostic criteria for functional movement disorders [4]. From our own reading of the older literature, however, it was clear that

this clinical entity had already been recognised much further back in the 19th century.

In this article, we re-examine the historical literature on functional facial movement disorders and compare it, and recent work, with a new case series of 41 patients with functional facial movement disorders to extend the historical and clinical perspective of this clinical presentation.

2. Methods

For the historical review, we carried out a systematic search of a collection of neurological textbooks and books on hysteria and allied conditions published prior to 1920. All book titles are available for public download from www.archive.org. Each book was searched for the terms ‘facial’, ‘blepharospasm’ and ‘ocular’. In this section, we sometimes use the term hysterical when it was the term used in these publications. In addition, we searched for descriptions of patients with atypical facial dystonia or movement disorder from 1960 until 2017 who had features in keeping with a functional facial movement disorder. For both the historical literature and recent search we explored references when relevant. Our case series is derived from 41 patients with functional facial movement disorders seen consecutively in general neurology and specialist ‘functional’ clinics in the Department of Clinical Neurosciences, Edinburgh by one of the authors (JS) over a period from 2008 to 2013.

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<https://doi.org/10.1016/j.jns.2018.09.031>

Received 3 July 2018; Received in revised form 19 September 2018; Accepted 25 September 2018

Available online 26 September 2018

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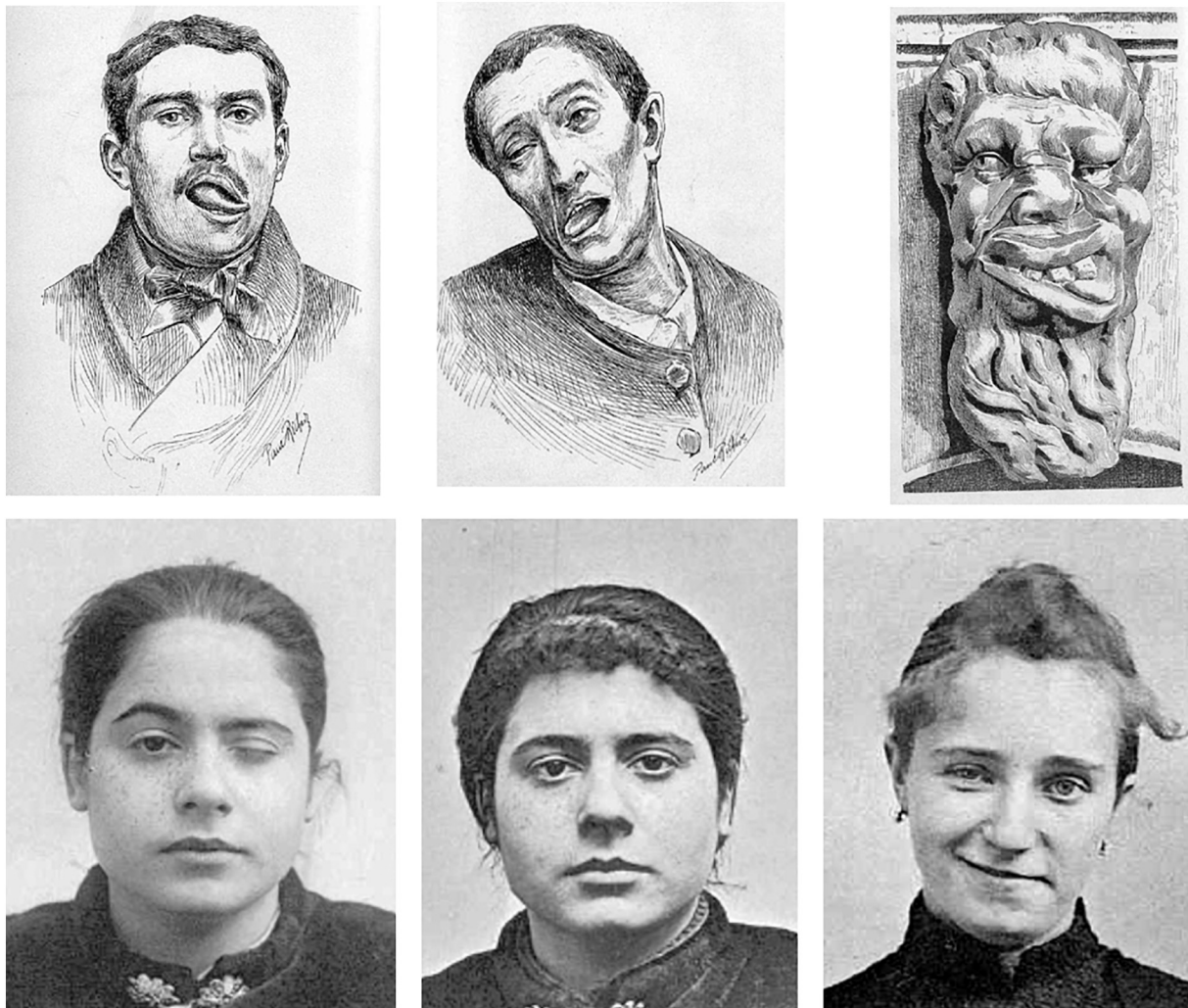


Fig. 1. Drawings and photographs from *Nouvelle Iconographie de Salpêtrière*, the in-house neurology journal of the Salpêtrière, founded by Jean-Martin Charcot. Upper images are from Volume 1 and show two patients with ‘l’hémispasme glosso-labié hystérique’ compared to a Venetian ‘mascaron’ [10]. Lower left and centre is a patient of Tourette with unilateral spasm treated with hypnosis [11] and lower right a patient of Bogroff in Odessa [13].

3. Review of literature 1880s to 1960s

We cannot find an earlier reference than 1887 when Charcot described unilateral hysterical facial spasm in the well-known patient ‘Le Log’ as follows [9]: “...left labial commissure is raised and mouth is partly open. At first thought to be paralysis of right inferior facial...on further examination it is due to spasm of the muscles on the left side of the face”. The following year Charcot launched his new journal, *Nouvelle Iconographie de Salpêtrière*, which incorporated the latest photographic techniques. In the first issue is an article by Charcot and Paul Richer with drawings demonstrating this phenomenon, as well as showing tongue deviation to the affected side. The article’s main purpose was to point out the similarity of the appearances to various types of mascaron, similar to a gargoyle on a church and used to frighten away evil spirits (Fig. 1) [10].

The next volume of the journal in 1889 carried a long article by Tourette which included a description of “*blépharospasme hystérique*” with photographs of a patient with a typical ‘depressed’ eyebrow on the affected side, and showing subsequent recovery with the aid of hypnosis (Fig. 1) [11]. In 1891 another article from Odessa carried another photograph of a patient with ‘hysterical’ hemifacial spasm (Fig. 1).

Gowers at around the same time [12] also described hysteria affecting the face:

“In hysteria there is either tonic contracture, especially in the orbicularis,

or attacks of quivering movement, which do not resemble true facial spasm... The effect of the preponderant contraction in the orbicularis and zygomatic muscles is a curiously mixed emotional aspect, a sort of whimpering smile”. Gowers, whose chapter on hysteria in that book has rarely been surpassed, also commented that these spasms were “usually lessened by rest, physical and mental... always increased by emotion, and by movement of the face, whether in speaking or chewing and ...by light and by cold.” He noted that “The influence of light is intelligible, since the orbicularis palpebrarum is almost always involved, and a strong light produces reflex contraction in this muscle under normal circumstances”. Gowers also refers to the presence of ‘wrong way’ tongue deviation, in which the tongue deviates to the side of facial spasm/apparent weakness, and is the opposite of what would be expected were the patient to have a pontomedullary lesion.

Babinski and Froment summarise earlier descriptions under the term ‘glosso-labial hemispasm’, under which it can often be found in subsequent textbooks [14]: “In glosso-labial hemispasm, described by Charcot, Brissaud and P. Marie, the spasm, as its name indicates, may be limited to the tongue and lips but sometimes affects simultaneously the orbicularis palpebrarum, platysma and neck muscles. The hook-like appearance of the tongue and the intermittent spasms of the contracted muscles give it an almost pathognomonic appearance.” They also reinforce the potential for confusion that could arise regarding whether there was paralysis or not “when there is facial asymmetry it will be found to be due not to

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