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Psychogenic Palatal Tremor May Be Underrecognized: Reappraisal of a Large Series of Cases

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Abstract

Background—Palatal tremor is characterized by rhythmic movements of the soft palate and can be essential or symptomatic. Some patients can have palatal movements as a special skill or due to palatal tics. Psychogenic palatal tremor is recognized but rarely reported in the literature.

Methods—We retrospectively evaluated all patients with palatal tremor seen in our center over a period of 10 years.

Results—Of 17 patients with palatal tremor, we identified 10 patients with isolated palatal tremor. In 70% of those the diagnosis of psychogenic palatal tremor could be made. Of the remainder, 2 had palatal tics and 1 essential palatal tremor.

Conclusions—We suggest that psychogenic palatal tremor may be underrecognized and propose that targeted clinical examination of positive signs for psychogenic movement disorders in these patients is essential. The correct identification of such patients has important clinical and scientific implications.

Keywords

essential palatal tremor; palatal myoclonus; psychogenic; tic; symptomatic

Palatal tremor (PT) (or palatal myoclonus) is a movement disorder characterized by rhythmic movements of the soft palate at 0.5 to 3 Hz.[1] PT is classically classified as essential (EPT)[1] when PT (with or without ear clicks) is the only feature and all imaging and laboratory investigations are normal, and as symptomatic (SPT) when PT is due to a structural or degenerative cause[1,2]; eg, lesion in Guillain-Mollaret triangle, glial fibrillary acidic protein (GFAP)[3,4] or polymerase-γ (POLG) mutations,[5] neuroferritinopathy,[6] or as part of progressive ataxia and PT (PAPT).[7] The tensor veli palatini innervated by the

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trigeminal nerve is mostly involved in EPT, whereas in SPT it is the levator veli palatini innervated mainly by the vagus nerve.[2,8-11]

Recently, a new classification has been proposed[12] in which EPT should be redesignated as "isolated," indicating the lack of any further signs, and include primary isolated PT (the classical EPT) and secondary isolated PT (PT as a special skill, palatal tic, and psychogenic PT). This report and our observation that some of our longstanding patients who initially presented with apparent isolated PT were in fact psychogenic, prompted us, in this study, to retrospectively revisit all PT patients seen in our center between 2001 and 2011.

Patients and Methods

We searched our database with the term "palatal," "palatal myoclonus," and "palatal tremor" for a period of 10 years (2001-2011). Twenty patients with PT were identified, of whom 3 were excluded because of insufficient clinical data. Of the excluded patients 1 had SPT and 2 isolated PT. We collected details on age, disease onset and duration, precipitating factors, treatment, evolution, concomitant conditions, clinical examination, psychiatric assessment, and further investigations. All patients were examined and followed by the same examiner (K.P.B.), and had at least 6 months of follow-up (range, 0.5-27 years).We specifically noted signs consistent with psychogenic movement disorders (PMDs)[13] and applied the criteria for PMDs to define the degree of diagnostic certainty.[14,15] Statistics were performed using PASW Statistics, version 19. Data are shown as mean ± standard deviation. Nonparametric variables were compared with the 2-sided Wilcoxon-Mann-Whitney test and $P < .05$ was considered significant.

Results

Of the 17 patients included, 7 had additional signs at first presentation and were therefore classified as SPT, and 10 had no additional signs at first presentation and were classified as having isolated PT. The mean age at onset for SPT was significantly older than for isolated PT (53.6 ± 5.2 vs 37.2 ± 7.4 years, respectively) ($P = .003$), in line with published data.[1,12] Of the 7 SPT patients, 5 were diagnosed as PAPT (2 negative for GFAP mutations), 1 developed PT subacutely after a left hemispheric stroke, and 1 had diaphragmatic myoclonus with coherent movements of the palate. Three had normal and 4 had abnormal brain MRI (1 olivary hypertrophy, 1 left hemispheric stroke, 1 cerebellar atrophy, and 1 olivary hypertrophy and cerebellar atrophy), whereas all EPT patients had extensive investigations that were normal (see Supporting Information).

From the 10 cases with isolated EPT at their first visit, 6 were diagnosed as primary EPT, 2 as palatal tics, and 2 as psychogenic PT. The patients were followed and the diagnosis was revised in 5 of 6 primary EPT cases to psychogenic PT. The duration from first visit to revision of diagnosis ranged from 2 to 18 years (Table 1). All patients with isolated EPT were examined for positive signs of PMDs according to published criteria (Table 1).[13,14] Of the 10 EPT cases, 7 (70%) had documented positive signs of PMD (Table 1), 6 of those 7 were female. The mean age at onset in those was 35.4 ± 6.4 years and the mean disease duration was 13.6 ± 11.2 years (range, 2-29 years). In all patients with psychogenic PT,

there was a physical precipitant (Table 1). The latency from trigger to PT onset is shown in Table 1. All patients reported ear clicking, mostly bilaterally, and in 3 cases this resolved later.

On examination of the 7 psychogenic PT cases, there was PT that was documented to be incongruous, variable, entrainable, and distractible (Table 1). In 3 of 7 there was an electromyography (EMG) confirmation of the variability and the irregularity of the rhythm, and in 1 additional patient of distractibility while recording. In 2 of 7 patients there were further neighboring muscles involved on follow-up, but this was variable and inconsistent in further follow-ups. In 6 of 7 patients there were multiple somatizations recorded, in 4 of 7 there was a psychiatric evaluation suggesting an underlying psychiatric condition, and in 1 case each, psychotherapy and antidepressants improved the symptoms (Table 1). Based on these data, 3 patients would be classified as clinically definite, 3 as clinically established. and 1 as probable psychogenic PT, according to published criteria.[13] Further supportive signs of PMDs[14] were abrupt onset and static course of the symptoms (n = 7), multiple somatizations (n = 6), spontaneous remissions (n = 1), self-inflicted injury (n = 1), and other movements consistent with PMDs[15,16] (n = 3) (Table 1). Of 7 psychogenic PT patients, 2 improved with botulinum neurotoxin (BoNT) injections (duration of treatment: 5 years and 10 years, respectively) (Table 1). Of 7 patients, 1 improved after the first time injected, but subsequent injections did not help. She was then started on amitriptyline 10 mg with benefit (Video Segment 2). One (case 3) has never received any treatment and is stable, with a moderate aggravation of her symptoms in the form of facial spasms. Of 7 patients, 3 (cases 2, 4, and 6) have not received BoNT but tried oral medications without success (Table 1). Interestingly, over the years, these 3 patients developed multiple complex PMDs or/and further psychogenic neurological symptoms (Table 1).

The 2 patients with palatal tics had other motor tics and a tic-disorder in the family history, excellent response to BoNT injections, and did not demonstrate signs of a PMD (Table 1). Of 10 EPT patients, 1 (case 8) was diagnosed as primary EPT, in whom all signs suggestive of PMDs were tested and found to be consistently negative. Illustrative cases may be found in the online Supporting Information.

Discussion

We report here that 70% of the patients with isolated PT seen in our center over a period of 10 years, were likely to be of psychogenic etiology based on published criteria for diagnosis of PMDs.[13] This is the largest series of patients with psychogenic PT reported in the literature. In line with published literature on PMDs the majority of the patients were female[16,17]; there was a precipitating factor (predominantly a minor viral respiratory infection)[12,18]; PT was often accompanied by bilateral ear clicking[12]; and there was either an additional PMD or other somatizations.[13] Consistent with other PMD, BoNT helped even at long-term follow-up.[19] Some of our patients showed involvement of further neighboring muscles in a variable and inconsistent way over the years, which could also be a sign of incongruity for EPT.[1,12]

Patients with psychogenic PT are rarely documented in the literature and therefore psychogenic PT is thought to be uncommon.[12,20-24] However, signs of psychogenicity according to published criteria[13] are not frequently tested in these patients: in an extensive review of 103 cases with EPT, signs of PMDs such as distractibility and entrainment were commented on in only 11 patients.[12] Supportive of our data, 5 out of those 11 had positive signs for PMDs. Thus, we propose here that psychogenic PT may be underrecognized and misdiagnosed as primary EPT when targeted examination for signs of psychogenicity according to published criteria is not done. In our case series this is illustrated by the fact that the majority of these patients were diagnosed by us as primary EPT for many years, before direct examination for these signs led to revision of the diagnosis.

Misdiagnosis of these patients has implications for their management and their long-term outcome. Delay in correct diagnosis and failure to provide suitable treatments are predictors of poor prognosis in PMDs.[25-28] This is perhaps reflected in our series by the fact that 3 cases deteriorated significantly over the years, presenting with more complex PMDs that rendered them severely disabled. Accurate identification of these patients would also enable their exclusion from studies on the largely unknown pathophysiology and evolution of primary EPT, and inclusion in future studies of treatment for PMDs.

Limitations of this study are that there is no biomarker available for the diagnosis of PMDs, and we acknowledge the difficulties in clinically diagnosing psychogenic PT, particularly via application of clinical diagnostic criteria for PMDs. However, 8 in 10 of the patients in the isolated EPT group are under active follow-up, and therefore were accessible for current assessment. The high percentage of psychogenic PT found in our population may not be representative for the prevalence of psychogenic PT in the community, given the method of patient ascertainment.

Finally, we would like to suggest that although the proposed new classification for PT[12] has some merit, there are some important caveats. Classification of psychogenic PT as a form of “secondary PT” is confusing. Secondary movement disorders are typically those in which an identifiable secondary event occurs on the background of previously normal brain function. Other PMDs such as psychogenic dystonia are not classified within the “secondary” category. Therefore, we suggest that psychogenic PT should not be classified under secondary EPT and that PT should instead be divided into 3 categories: EPT (“primary”), SPT, and psychogenic PT.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Table 1

Clinical characteristics of patients with isolated PT

PN	G	Age at onset (yr)	Duration (yr)	Followup (yr)	Time from first visit to diagnosis (yr)	Precipitant (latency)	Other somatizations and evolution	Psychiatric history and evaluation	Incogruity/variability/entrainment/distractibility	Diagnostic classification[a]	Treatment
Psychogenic PT											
1	F	30	2	0.5	0	Viral labyrinthitis (3 weeks)	PT stable, pressure on left side of head	History of sexual and physical abuse; positive psychiatric family history	+/+/+/+	Clinically definite	Symptoms better after psychotherapy-BoNT: improvement after 3 days; further BoNT treatments no clear benefit; 2 weeks treatment with 10 mg amitryptiline, clear improvement of PT
2	F	34	12	9	6	Tinnitus (same time)	PT stable, over the years other muscles involved, eg, larynx; 10-yr history dysphagia, 1-yr history sleep disturbance	Anorexia in her teens, later diagnosed with depression	+/+/+/+	Clinically established	Pregabalin, levetiracetam, clonazepam: no benefit; diazepam: better; BoNT refused
3	F	33	29	27	18	Flu-like illness (2 days)	PT stable, over the years other muscles involved but variability of those in each follow-up, new onset facial spasms	No obvious psychiatric disorder	+/+/+/+	Clinically established	BoNT not tried
4	F	40	9	7	3	Sore throat, right-sided otitis (same time)	3 yr after onset psychogenic head and neck jerks, rocking movements of the trunk, head bobbing	Dissociative motor disorder; mother depression; history of sexual abuse	+/+/+/+	Clinically established	Clonazepam, no benefit, BoNT refused; cognitive behavioral therapy declined
5	F	47	15	4	2	Flu (1 week)	Stable, no additional symptoms	No obvious psychiatric disorder	+/+/+/+	Probable	BoNT improvement
6	F	28	5	4	0	Endoscopy for nausea (2 weeks)	Nausea, tachycardia, fatigue,pain, headaches; 3 yr later generalized weakness; wheelchair; visual disturbance	Self-injury in the past; paternal aunt had depression and comitted suicide	+/+/+/+	Clinically definite	BoNT refused
7	M	36	28	23	14	Vomiting, vertex headache (same time)	PT stable; feeling of lump on scalp which he rubs causing hair loss; long periods of	Depression diagnosed 10 yr after onset	+/+/+/+	Clinically definite	Benzehxol, baclofen, diltiazem, levodopa, clonazepam: no benefit; fluoxetine, paroxetine, BoNT: improvement

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PN	G	Age at onset (yr)	Duration (yr)	Followup (yr)	Time from first visit to diagnosis (yr)	Precipitant (latency)	Other somatizations and evolution	Psychiatric history and evaluation	Incogruity/variability/entrainment/distractibility	Diagnostic classification[a]	Treatment
							symptom remission with relapse				
							symptom remission with relapse				
Primary EPT											
8	F	33	2,5	1	NA	Mild throat infection 2.5 months before	Generalized weakness-breathing problems, abdominal pain with no organic cause; uses wheelchair outside	No obvious psychiatric disorder	−/−/−/−	NA	BoNT: improvement
Palatal tics											
9	M	40	2	2	0	No	Motor tics	No obvious psychiatric disorder	−/−/−/−	NA	BoNT: improvement
10	F	40	7	4	0	No	Motor tics	No obvious psychiatric disorder	−/−/−/−	NA	BoNT: improvement

[a]According to criteria for psychogenic movement disorders.[14,15]EPT, essential palatal tremor; PT, palatal tremor; PN, patient number; G, gender; F, female, M, male; BoNT, botulinum toxin; NA, not applicable.