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**REVIEW**

- 1367** Biomarkers *vs* imaging in the early detection of hepatocellular carcinoma and prognosis  
*Balaceanu LA*

**ORIGINAL ARTICLE****Basic Study**

- 1383** Study on gene expression patterns and functional pathways of peripheral blood monocytes reveals potential molecular mechanism of surgical treatment for periodontitis  
*Ma JJ, Liu HM, Xu XH, Guo LX, Lin Q*

**Case Control Study**

- 1393** Clinical differentiation of acute appendicitis and right colonic diverticulitis: A case-control study  
*Sasaki Y, Komatsu F, Kashima N, Sato T, Takemoto I, Kijima S, Maeda T, Ishii T, Miyazaki T, Honda Y, Shimada N, Urita Y*

**Retrospective Study**

- 1403** Feasibility of prostatectomy without prostate biopsy in the era of new imaging technology and minimally invasive techniques  
*Xing NZ, Wang MS, Fu Q, Yang FY, Li CL, Li YJ, Han SJ, Xiao ZJ, Ping H*
- 1410** Safety and efficacy of transfemoral intrahepatic portosystemic shunt for portal hypertension: A single-center retrospective study  
*Zhang Y, Liu FQ, Yue ZD, Zhao HW, Wang L, Fan ZH, He FL*

**Observational Study**

- 1421** Impact of gastroesophageal reflux disease on the quality of life of Polish patients  
*Gorczyca R, Pardak P, Pękala A, Filip R*

**SYSTEMATIC REVIEWS**

- 1430** Non-*albicans* *Candida* prosthetic joint infections: A systematic review of treatment  
*Koutserimpas C, Zervakis SG, Maraki S, Alpantaki K, Ioannidis A, Kofteridis DP, Samonis G*

**META-ANALYSIS**

- 1444** Relationship between circulating irisin levels and overweight/obesity: A meta-analysis  
*Jia J, Yu F, Wei WP, Yang P, Zhang R, Sheng Y, Shi YQ*

**CASE REPORT**

- 1456** Cirrhosis complicating Shwachman-Diamond syndrome: A case report  
*Camacho SM, McLoughlin L, Nowicki MJ*

- 1461** Robot-assisted trans-gastric drainage and debridement of walled-off pancreatic necrosis using the EndoWrist stapler for the da Vinci Xi: A case report  
*Morelli L, Furbetta N, Gianardi D, Palmeri M, Di Franco G, Bianchini M, Stefanini G, Guadagni S, Di Candio G*
- 1467** Fulminant liver failure following a marathon: Five case reports and review of literature  
*Figiel W, Morawski M, Grąt M, Kornasiewicz O, Niewiński G, Raszeja-Wyszomirska J, Krasnodębski M, Kowalczyk A, Holówko W, Patkowski W, Zieniewicz K*
- 1475** Gaucher disease in Montenegro - genotype/phenotype correlations: Five cases report  
*Vujosevic S, Medenica S, Vujicic V, Dapcevic M, Bakic N, Yang R, Liu J, Mistry PK*
- 1483** Longitudinal observation of ten family members with idiopathic basal ganglia calcification: A case report  
*Kobayashi S, Utsumi K, Tateno M, Iwamoto T, Murayama T, Sohma H, Ukai W, Hashimoto E, Kawanishi C*
- 1492** Secondary lymphoma develops in the setting of heart failure when treating breast cancer: A case report  
*Han S, An T, Liu WP, Song YQ, Zhu J*
- 1499** Removal of pediatric stage IV neuroblastoma by robot-assisted laparoscopy: A case report and literature review  
*Chen DX, Hou YH, Jiang YN, Shao LW, Wang SJ, Wang XQ*
- 1508** Premonitory urges located in the tongue for tic disorder: Two case reports and review of literature  
*Li Y, Zhang JS, Wen F, Lu XY, Yan CM, Wang F, Cui YH*
- 1515** Female genital tract metastasis of lung adenocarcinoma with EGFR mutations: Report of two cases  
*Yan RL, Wang J, Zhou JY, Chen Z, Zhou JY*
- 1522** Novel heterozygous missense mutation of *SLC12A3* gene in Gitelman syndrome: A case report  
*Wang CL*
- 1529** Thoracotomy of an asymptomatic, functional, posterior mediastinal paraganglioma: A case report  
*Yin YY, Yang B, Ahmed YA, Xin H*

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## Premonitory urges located in the tongue for tic disorder: Two case reports and review of literature

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### Abstract

#### BACKGROUND

Premonitory urges (PUs) was defined as the uncomfortable physical sensations of inner tension that can be relieved by producing movement responses. Nearly 70%-90% patients with Tourette syndrome reported experiences of PUs.

#### CASE SUMMARY

In this paper, we present two cases of young patients with PUs located in their tongue, which is very rare and easily misdiagnosed in clinical work. Both two young patients complained of an itchy tongue and cannot help biting their tongue. These two cases were worth reporting because it was rare that PUs was the initial symptom and located in the tongue. The results indicated that PUs seem to play an important role in the generation of tics.

#### CONCLUSION

Thus, PUs may be the first process, and an essential part, of the formation of tics.

**Key words:** Premonitory urges; Sensory tics; Tic disorders; Tourette syndrome; Case report

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**Core tip:** These two cases were worth reporting because it was rare that premonitory urges (PUs) was the initial symptom and located in the tongue. The results indicated that PUs seem to play an important role in the generation of tics.

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## INTRODUCTION

Sensory tics, which were first described by Bliss<sup>[1]</sup> in 1980, have mainly been described as uncomfortable physical sensations of inner tension that can be relieved by producing movement responses. Over subsequent years, many researchers have used different terminologies to define this symptom<sup>[2-4]</sup>. For instance, Cohen and Leckman called it the "Sensory phenomena" including descriptors such as "Urge", "Somatic Sensation" and "Heightened sensational Impulsivity"<sup>[3]</sup>. Some studies have demonstrated that sensory phenomena mainly include bodily sensations, mental urges and feelings of incompleteness, the need for things to be "just right", and motor or vocal responses to the sensations are required<sup>[5,6]</sup>. However, most recent studies have used the term "premonitory urges (PUs)" to describe this sensory symptom<sup>[7-9]</sup>.

Nearly 70%-90% patients with Tourette syndrome (TS) report experiences of PUs<sup>[10,11]</sup>. It has been proposed that there are three parts to PUs, including sensory urges (focal visceral-sensations or muscular-skeletal), autonomic urges (symptoms such as nausea, sweating, and palpitations) and cognitive urges (feelings of incompleteness)<sup>[5]</sup>. Although PUs were not included in the diagnostic criteria of TS, some published studies have regarded it as the core symptom of TS<sup>[12,13]</sup>.

In this paper, we present two cases of young patients with PUs located in their tongue, which is very rare and easily misdiagnosed in clinical work. Both young patients complained of an itchy tongue and cannot help biting their tongue. These two cases were worth reporting because it was rare that PUs were the initial symptom and were located in the tongue. We also provide a literature review on PUs.

## CASE PRESENTATION

### Chief complaints

**Case 1:** A 9-year-old girl, started to eat less and less for half a year prior to examination. Her main complaint was "Why is my tongue itchy?" The girl described her feeling as "My tongue was so itchy that I cannot help biting my tongue!". Her weight went from 70 pounds to 36.

**Case 2:** A 5-year-old boy, also complained of an itchy tongue and could not stop biting the tip of his tongue (**Figure 1**).

### Personal and family history

**Case 1:** Her medical and family history did not reveal any relevant information.

**Case 2:** His medical and family history did not reveal any relevant information.

### Physical examination

**Case 1:** Her temperature was 36.1, heart rate was 95 bpm, respiratory rate was 16 breaths per minute, blood pressure was 120/85 mmHg with oxygen saturation in room air was 98%. A clinical intraoral examination was conducted but no significant signal was found.

**Case 2:** His temperature was 36.4, heart rate was 98 bpm, respiratory rate was 18 breaths per minute, blood pressure was 120/80 mmHg with oxygen saturation in room air was 95%. We also performed a clinical intraoral examination but no significant indicator was found.

### Laboratory examinations

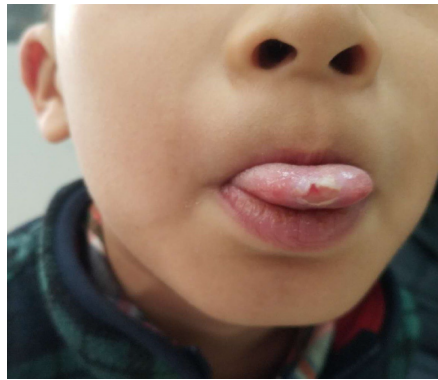
**Case 1:** During the hospitalization, the girl received a series of blood testing, but no significant clinical indicators were found (**Table 1**).

**Case 2:** There was no significant clinical indicators were found (**Table 2**).

### Imaging examinations

**Case 1:** An Magnetic resonance imaging scan of her brain was also obtained but no





**Figure 1** The tip of tongue for case 2. The boy can't help biting his tongue, the tip of the tongue has been bitten through.

significant findings were found (Figure 2).

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## FINAL DIAGNOSIS

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### Case 1

Provisional Tic Disorder (the Diagnostic Criteria of Provisional Tic Disorder see Supplementary Materials). The symptom of an "itchy tongue" was recognized as a PU, and "biting the tongue" was regarded as a symptom of the tic.

### Case 2

The boy was finally diagnosed as having a Provisional Tic Disorder (the Diagnostic Criteria of Provisional Tic Disorder see Supplementary Materials). The symptom "itchy tongue" belonged to one kind of PU.

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## TREATMENT

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### Case 1

For medical treatment, Aripiprazole (2.5 mg per day) was used. Psychological interventions were also performed to relieve the PUs (*e.g.*, habit reversal training). The therapeutic effects and safety of this comprehensive treatment program were assessed during those treatments.

### Case 2

Clonidine transdermal patch on his back for 4 wk (1 pin).

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## OUTCOME AND FOLLOW-UP

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### Case 1

The PUs were mostly relieved, "biting the tongue" has resolved, and her diet returned normal, as well, after four weeks. Finally, by following this treatment program, the girl recovered and was sent back to school after eight weeks.

### Case 2

After 6 wk treatment mentioned above, the boy recovered, and the "itchy tongue" was gone.

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## DISCUSSION

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Although most children with TS report PUs<sup>[10]</sup>, few cases have shown the onset of PUs at such a young age (*i.e.*, Case 2). It should be noted that the reporting of PUs relies on an age-related ability to describe sensory phenomena<sup>[14]</sup>. Rozenman *et al*<sup>[15]</sup> reported that the severity of PUs increased with age. Woods *et al*<sup>[16]</sup> found that there were no correlations between the PUs and the severity of tics in young patients before the age of ten. Based on general clinical observations, Leckman *et al*<sup>[17,18]</sup> reported that young

Table 1 Blood tests of case 1

Items	Abbreviation	Results	Items	Abbreviation	Results
Potassium	K	4.86 mmol/L	White blood cell count	WBC	$5.45 \times 10^9$
Sodium	Na	141.4 mmol/L	Red blood cell count	RBC	$4.26 \times 10^{12}$
Chlorine	Cl	105.7 mmol/L	Hemoglobin	HGB	130 g/L
Bicarbonate	CO <sub>2</sub>	25.3 mmol/L	Hematocrit	HCT	39.80%
Total protein	TP	76.2 g/L	Mean corpuscular volume	MCV	93.4 fl
blood urea nitrogen	Urea	6.41 mmol/L	Mean corpuscular hemoglobin	MCH	30.5 pg
Creatinine	Cre	43.0 $\mu$ mol/L	Mean corpuscular hemoglobin concentration	MCHC	327 g/L
Cholesterol	Chol	4.23 mmol/L	Platelet	PLT	$238 \times 10^9$
Uric acid	UA	352.8 $\mu$ mol/L	Platelet distribution width	PDW	12.0 fl
Glucose	GLU	4.85 mmol/L	Mean platelet volume	MPV	10.8 fl
Calcium	Ca	2.52 mmol/L	Platelet larger cell ratio	PLCR	30.10%
Phosphorus	P	1.60 mmol/L	Plateletcrit	PCT	0.26
Alkaline phosphatase	ALP	298 U/L	Neutrophil ratio	NEUT	$2.50 \times 10^9$
Aspartate aminotransferase	AST	33.3 U/L	Lymphocyte ratio	LYMPH	$2.31 \times 10^9$
Alanine aminotransferase	ALT	26.6 U/L	Monocyte ratio	MONO	$0.30 \times 10^9$
Gamma-GT	GGT	11.6 U/L	Eosinophil ratio	EO	$0.32 \times 10^9$
Total bilirubin	TBIL	9.96 $\mu$ mol/L	Basophil ratio	EASO	$0.02 \times 10^9$
Triglyceride	TG	0.49 mmol/L	Neutrophil count	NEUT%	45.80%
Creatine kinase	CK	134 U/L	Lymphocyte count	LYMPH%	42.40%
Creatine kinase-MB	CK-MB	22 U/L	Monocyte count	MONO%	5.50%
Lactate dehydrogenase	LDH	235 U/L	Eosinophil count	EO%	5.90%
$\alpha$ -hydroxybutyric dehydrogenase	HBDH	195 U/L	Basophil count	BASO%	0.40%
High-density lipoprotein cholesterol	HDL-C	1.54 mmol/L	Antistreptolysin	ASO	75.30%
Low-density lipoprotein cholesterol	LDL-C	2.09 mmol/L	C-reactive protein	CRP	3.37 mg/L
Very low-density lipoprotein	VLDL-C	0.10 mmol/L	Ceruloplasmin	CER	296.0 mg/L

No significant clinical indicators were found in blood tests of case 1.

children with simple tics (*e.g.*, a quick head jerk or blinking) did not always show sensory phenomena or were unaware of these symptoms until they were ten years old. The first case report for the present study supported that being 10 years old may play an important role in tic awareness. However, a low level of PUs has also been reported in younger patients (such as Case 2). For example, Gulisano found a low level of PUs in young patients with TS (mean age, 7.3 years  $\pm$  1.5)<sup>[11]</sup>. We should pay more attention to the "uncomfortable feelings" in young children and take PUs into consideration in clinical work. Developmentally, PUs often appeared three years after the first onset of tics<sup>[12,19]</sup>. However, in the two abovementioned cases, PUs was regarded as the initial symptom, which is rarely reported in clinical work. These cases indicate that PUs might be the first process in tic disorders for some clinical cases. Indeed, in many studies on PUs, patients always report that tics were executed to alleviate distress-related PUs<sup>[13,20]</sup>. Some researchers have proposed that there are two processes related to tics: The PUs (negative reinforcement) and the relief after tic expression (positive reinforcement)<sup>[16,21]</sup>. In the two case reports discussed here, the PUs (itchy tongue) may be an early indicator for tic generation. With the development of PUs, tics (biting the tongue) emerged later.

Moreover, it should be noted that the feelings of PUs might be vague and poorly localized in a certain area, such as the face, neck, shoulder or arms, but rarely in the tongue<sup>[8,22]</sup>. For the assessment of PUs, two validated scales, the University of São Paulo Sensory Phenomena Scale (USP-SPS) and the Premonitory Urge for Tic Scale, have been developed, and both have shown good psychometric properties<sup>[16,23]</sup>. While these two scales have been widely used in clinical research related to tics, they report the feelings before tics and the corresponding severity, but they do not include the locations of PUs. Kwak *et al.*<sup>[6]</sup> also designed a questionnaire to identify PUs in patients with TS, which included the locations of PUs. He found that the anatomical locations, including the face/head, neck, shoulders, arms, hands, throats, feet, and stomach/abdomen, were more often reported in TS with PUs. From the two abovementioned cases, we found that PUs could be easily misdiagnosed in clinical works. It seems that



Table 2 Blood tests of case 2

Items	Abbreviation	Results	Items	Abbreviation	Results
Potassium	K	4.15 mmol/L	White blood cell count	WBC	$5.43 \times 10^9$
Sodium	Na	139.7 mmol/L	Red blood cell count	RBC	$5.26 \times 10^{12}$
Chlorine	Cl	107.7 mmol/L	Hemoglobin	HGB	128 g/L
Bicarbonate	CO <sub>2</sub>	26.8 mmol/L	Hematocrit	HCT	37.80%
Total protein	TP	61.8 g/L	Mean corpuscular volume	MCV	92.4 fl
blood urea nitrogen	Urea	4.12 mmol/L	Mean corpuscular hemoglobin	MCH	31.6 pg
Creatinine	Cre	48.1 $\mu$ mol/L	Mean corpuscular hemoglobin concentration	MCHC	318 g/L
Cholesterol	Chol	2.66 mmol/L	Platelet	PLT	$227 \times 10^9$
Uric acid	UA	268.0 $\mu$ mol/L	Platelet distribution width	PDW	11.7 fl
Glucose	GLU	4.93 mmol/L	Mean platelet volume	MPV	11.1 fl
Calcium	Ca	2.41 mmol/L	Platelet larger cell ratio	PLCR	32.10%
Phosphorus	P	1.74 mmol/L	Plateletcrit	PCT	0.29
Alkaline phosphatase	ALP	336 U/L	Neutrophil ratio	NEUT	$2.34 \times 10^9$
Aspartate aminotransferase	AST	19.3 U/L	Lymphocyte ratio	LYMPH	$2.35 \times 10^9$
Alanine aminotransferase	ALT	9.5 U/L	Monocyte ratio	MONO	$0.28 \times 10^9$
Gamma-GT	GGT	12.5 U/L	Eosinophil ratio	EO	$0.31 \times 10^9$
Total bilirubin	TBIL	13.33 $\mu$ mol/L	Basophil ratio	EASO	$0.02 \times 10^9$
Triglyceride	TG	0.51 mmol/L	Neutrophil count	NEUT%	45.60%
Creatine kinase	CK	79 U/L	Lymphocyte count	LYMPH%	41.40%
Creatine kinase-MB	CK-MB	17 U/L	Monocyte count	MONO%	5.45%
Lactate dehydrogenase	LDH	134 U/L	Eosinophil count	EO%	5.85%
$\alpha$ -hydroxybutyric dehydrogenase	HBDH	123 U/L	Basophil count	BASO%	0.42%
High-density lipoprotein cholesterol	HDL-C	1.53 mmol/L	Antistreptolysin	ASO	74.45%
Low-density lipoprotein cholesterol	LDL-C	0.73 mmol/L	C-reactive protein	CRP	3.21 mg/L
Very low-density lipoprotein	VLDL-C	0.10 mmol/L	Ceruloplasmin	CER	287.0 mg/L

No significant clinical indicators were found in blood tests of case 2.

the clinical information about the locations of PUs was an indispensable piece of information for the assessment of PUs.

Several recent studies on children with TS have reported that the severity of PUs increases with the severity of the tics<sup>[8,24]</sup>. It has been indicated that increased insight into PUs could help patients recognize alleviating factors for tic symptoms and improve their ability to suppress them. For example, higher awareness of PUs could benefit tic suppression<sup>[20]</sup>. Furthermore, as a potential focus for behavioral or psychological therapy, PUs has received increasingly more attention in recent years<sup>[25-27]</sup>. The structured behavioral therapy called Comprehensive Behavioral Intervention for Tics has mainly focused on improving the awareness of tics such as PUs<sup>[28]</sup>. Although the exact role of PUs is unknown, it has been postulated that they could reflect subjective experiences below the tic-production threshold<sup>[12]</sup>. PUs might be a useful predictor for treatment response due to their relationship with tic severity<sup>[29]</sup>. It is believed that the hyperactivity of the insula, as well as the anterior cingulate cortex and the supplementary motor area was involved in the neural mechanism of PUs<sup>[30-32]</sup>. A recent structural neuroimaging study suggested that tic generation was mediated by the insula, which is responsible for the subjective perception of PUs<sup>[33]</sup>. The study indicated that the insula might play an important role in the translation of urges to tics<sup>[34,35]</sup>.

## CONCLUSION

These two case reports are the first to describe PUs as the initial symptoms of TS in clinical work in China. The results indicated that PUs seem to play an important role in the generation of tics. Thus, PUs may be the first process, and an essential part, of the formation of tics. The study of PUs, especially neural mechanisms underlying PUs, would facilitate the understanding of the pathophysiology and pathogenesis of tics.



Figure 2 Magnetic resonance imaging scan for case 1. There was no abnormal signal found on T1, T2 and T2 FLAIR of brain scan for case 1.

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