

Challenges of Misdiagnosis and Suboptimal Treatment of Persistent Idiopathic Facial Pain and Atypical Odontalgia: A Retrospective Multi-Centric Cross-Sectional Investigation

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Purpose: To explore the challenges faced in the diagnosis and treatment of atypical odontalgia (AO) and other persistent idiopathic facial pain (PIFP).

Patients and Methods: This retrospective multi-centric cross-sectional study utilized clinical information (eg, clinical manifestations, history of consultations and treatments prior to correct diagnoses) from patients' medical records. Their economic parameters were also extracted from medical insurance databases. Each variable collected was statistically analyzed. Differences of variables between AO and other PIFP were statistically tested.

Results: A total of 394 patients were included in this study. On average, the diagnostic delay was 34.8±14.2 months, a median of 7 consultations were performed prior to a correct diagnosis, and 5 ineffective prescriptions were issued. Patients with AO suffered from longer diagnostic delays than patients with other PIFP (38.4±14.3 months vs 31.6±13.4 months) and were more likely to receive invasive dental treatments (73.3% vs 4.3%). The median total cost of treatment for a single patient before correct diagnosis was 8506.3 yuan. Patients with AO spent more than patients with other PIFP (10,146.5 Yuan vs 3864.0 Yuan).

Conclusion: Patients with PIFP were frequently misdiagnosed and the length of diagnostic delay was observed to be long. Ineffective medications, unwarranted procedures, and unnecessary economic burden were imposed on the patients. Patients with AO are in a worse situation than patients with other PIFP. As significantly urgent and unsatisfactory results of misdiagnosis of PIFP have been reported in our study, more attention should be paid on the research and education in this field.

Keywords: persistent idiopathic facial pain, atypical odontalgia, misdiagnosis, suboptimal treatment, economic burden

Introduction

Persistent idiopathic facial pain (PIFP), previously termed atypical facial pain, was defined in the third version of the International Classification of Headache Disorders (ICHD-3) as persistent facial and/or oral pain with varying presentations, recurring daily for more than two hours per day over the course of three months, in the absence of a clinical neurological deficit.¹ Clinical presentation of PIFP is variable, the diagnostic criteria for PIFP are loose and ambiguous, and the pathological mechanism is not clear.²⁻⁵ As a result, the diagnoses of PIFP are usually regarded as “waste baskets” or exclusive diagnoses.⁶ Some patients are diagnosed

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with PIFP only after a prior misdiagnosis, and insufficient and/or incorrect treatment methods that failed to provide satisfactory pain relief.⁷⁻⁹

Atypical odontalgia (AO), a subtype of PIFP, was defined in ICHD-3 as a condition in which a continuous pain is experienced in one or more teeth, or in a tooth socket after tooth extraction, in the absence of a dental cause.¹ Because of its similarities in diagnostic criteria with other facial pain conditions and the lack of understanding of its pathological mechanism, AO is frequently misdiagnosed and improperly treated.¹⁰ Consequently, irreversible and ineffective dental treatments may be applied and cause worsening morbidity.^{11,12}

Therefore, patients with AO and other PIFP continue to suffer from the decline in quality of life associated with chronic pain and possible complications of ineffective treatments.¹³⁻¹⁵ Moreover, repeated diagnoses and treatments might impose serious health-economic burden on patients, which could have been avoided with early accurate diagnosis.

A recently published research, focused on the diagnostic challenges of PIFP, ingeniously reported the detailed circumstances of diagnostic delay of PIFP and found that intraoral PIFP was more easily misdiagnosed than its extraoral sibling.⁸ Their report calculated the challenges faced in the diagnoses of PIFP, and emphasized the importance of extensive education and training of professionals regarding this entity.⁸ However, the study simply reported on intraoral PIFP, which is not limited to teeth and the tooth socket as AO, from which biased results might have been generated. Meanwhile, physical and financial burden caused by the misdiagnosis were not considered. Furthermore, the monocentric research was performed with a limited number of participants.

We conducted this study in order to further clarify the condition of diagnostic delay of PIFP and to complement previous studies. The detailed aims of this study include the following aspects: to investigate the general condition of diagnostic delay of PIFP, especially whether the subset AO has distinctive characteristics compared to other PIFP; to investigate the condition of ineffective treatments given to and related economic burdens imposed on patients during the diagnostic delays; to get more reliable results by conducting investigations in a multicentric cross-sectional perspective. In addition, this study was conducted amongst the Chinese population. To the best of our knowledge, the diagnostic challenges of AO and other PIFP have not been studied in the Chinese population yet.

Methods

Study Design, Setting and Participants

This retrospective cross-sectional study was performed in compliance with the Declaration of Helsinki and reported in accordance with the STROBE (Strengthening The Reporting of Observational studies in Epidemiology) statement. This study was approved by the IRB (Institutional Review Board) of Beijing Red Cross Peace Orthopedic Hospital. Due to the retrospective nature of this study, informed consents from patients were waived by the IRB. Patient data collected in this study were maintained with confidentiality.

A total of four hospitals including Beijing Tiantan Hospital, Beijing Fengtai Hospital, Beijing Red Cross Peace Orthopedic Hospital and Beijing Puhua International Hospital in Beijing, China, served as participant centers. Patients who were diagnosed as PIFP or AO in the participating centers from January 2017 to December 2019 were identified and regarded as candidates to be included in this study. Their medical documents were retrieved for review. Patients whose medical information was incompletely or inaccurately recorded were excluded. The participating centers applied consistent criteria to diagnose PIFP or AO: (1) Facial pain and/or oral pain more than 2 hours a day for more than 3 months. (2) Pain not following the distribution of peripheral nerves. (3) Patients with disorders related to teeth, oral mucosa, salivary glands, temporal-mandibular joint and maxillary sinus were excluded. (4) Other underlying causes were excluded by proper examinations indicated by the patients' clinical manifestation. The diagnoses of PIFP in these centers were established by experienced and well trained clinicians specializing in pain management.

Data Acquisition

Data of included patients were acquired through two approaches. Firstly, their medical records were retrieved and demographic and clinical conditions were obtained. Variables such as age, gender, onset of symptom, position, intensity and feature of pain, history of dental procedures prior to the onset of pain, date of first medical consultation for the pain, number of previous consultations, detailed nature of each consultation (date of consultations, consulted faculties, diagnoses, prescriptions, therapeutic interventions or procedures) were extracted from the patient's medical records. The intensity of pain was routinely graded using visual analog scale (VAS) score, with

0 representing no pain and 10 representing the worst pain imaginable, in all participating centers. Secondly, the economic parameters were extracted from the databases of the medical insurance department of the health administration. The cost of each previous consultation, including total cost, cost of medications, cost of therapeutic interventions or procedures were enquired and recorded separately.

Statistics Analysis

IBM SPSS Statistics Version 23 was used for statistical analyses. Each collected variable was statistically analyzed. For measurement data, if the variables followed normal distribution, means and standard deviations were calculated and Studentt-tests were used for intergroup comparison; otherwise, quartiles were calculated and Mann–Whitney U-test or Kruskal–Wallis H-tests were used for intergroup comparisons. For categorical data, frequencies and percentages were calculated and chi-square tests were used for intergroup comparisons. Multiple linear regression was used to analyze the factors of diagnostic delay. $P < 0.05$ was set for statistical significance.

Results

Participants and Descriptive Data

A total of 394 patients were included in the setting period, among whom 187 (47.5%) were diagnosed as AO and 207 (52.5%) were diagnosed as other AO PIFP. The summary of descriptive data is shown in [Table 1](#).

Diagnostic Delay

Nearly all patients (381, 96.7%) started consulting medical professionals for pain within 1 month after onset. Patients with AO received correct diagnosis on an average of 38.4 ± 14.3 months after the onset of pain. This length was longer than the average diagnostic delay of 31.6 ± 13.4 months in patients with other PIFP. The detailed summary is shown in [Table 1](#).

Previous history of dental procedure, intraoral position of pain, number of consultations, number of prescriptions, number of invasive treatments and patients with AO were found to be linearly correlated with longer delay in univariate analyses. However, only patients with AO were found to have independent factors for longer delay in multiple linear regression. The factors related to diagnostic delay are shown in [Table 2](#).

Overall, a median of 7 consultations were performed preceding a correct diagnosis. The median number of

consultations was higher in patients with AO with other PIFP ($P < 0.001$). For patients with AO, dentists (1047, 61.0%) were the most commonly consulted medical professionals, followed by neurologists (156, 9.1%), general practitioners (153, 8.9%), pain management specialists (145, 8.4%) and neurosurgeons (134, 7.8%). For patients with other PIFP, neurologists (757, 44.3%) were most commonly consulted, followed by general practitioners (267, 15.6%), pain management specialists (221, 12.9%), dentists (197, 11.5%) and neurosurgeons (161, 9.4%).

All patients received at least one diagnosis apart from PIFP, and all patients with AO had the history of being diagnosed as dental pain, such as periodontitis, pulpitis, tooth sensitivity, cracked tooth, failed root canal treatment, postoperative pain, and occlusal trauma. In 74.3% of the included patients, diagnoses included neuralgia (217, 55.1%), neuritis (163, 41.4%), burning mouth syndrome (91, 23.1%), iatrogenic nerve trauma (61, 15.5%), myofascial pain (57, 14.5%), and osteomyelitis (34, 9.1%) were also received.

Therapeutic Conditions

The median number of prescriptions for the treatment of pain was 5. Generally, patients with AO received more prescriptions ($P < 0.001$). All patients received nonsteroidal anti-inflammatory drugs and 134 (34.0%) patients received antibiotics for treatment. Decongestants, steroids, muscle relaxants, antidepressants, antiepileptics, antihistamines and antianxiety medications were also prescribed to 293 (74.4%), 242 (61.4%), 171 (43.4%), 134 (34.0%), 93 (23.6%), 84 (21.3%) and 41 (10.4%) patients, respectively. In addition, most of the patients with AO (173, 92.5%) were prescribed mouthwashes.

For the control of pain, 229 (58.1%) patients were found to have undergone invasive treatments. We detected significant discrepancy in invasive treatments between AO and other PIFP, as AO patients were more likely to have received these manipulations ($P < 0.001$). Consequently, AO patients were more likely to have received dental procedures ($P < 0.001$), however other invasive treatments such as nerve block, microvascular decompression (MVD), percutaneous balloon compression (PBC), radiofrequency and radiotherapy were performed on a similar proportion of patients in both groups ($P = 0.395$).

Characteristics of Health Economics

The median total cost of medical treatment for a single patient before the correct diagnosis was 8506.3 yuan. Specifically, patients with AO spent more than their other

Table 1 Summary of Descriptive Data and Conditions of Diagnostic Delay

	AO (n = 187)	Other PIFP (n = 207)	P	All Patients
Age (mean ± SD, years-old)	48.7±10.6	53.9±13.4	<0.001*	51.4±12.4
Gender (Female Proportion)	67.9%	63.8%	0.397	65.7%
Previous history of dental procedure [n (%)]	149 (81.0%)	8 (3.9%)	<0.001*	157 (39.8%)
VAS at onset [median (IQR)]	5 (7–8)	5 (7–9)	0.179	5 (7–8)
Intraoral position at onset [n (%)]	187 (100%)	37 (17.9%)	<0.001*	224 (56.9%)
First consultation within 1 month after onset [n (%)]	184 (98.4%)	197 (95.2%)	0.092	381 (96.7%)
Diagnostic delay (mean ± SD, months)	38.4±14.3	31.6±13.4	<0.001*	34.8±14.2
Number of consultations [median (IQR)]	9 (7–11)	5 (3–7)	<0.001*	7 (4–9)
Number of prescriptions [median (IQR)]	7 (5–8)	4 (2–5)	<0.001*	5 (3–7)
Patients who received invasive treatments [n (%)]	156 (83.4%)	78 (37.7%)	<0.001*	229 (58.1%)
Dental procedures [n (%)]	137 (73.3%)	9 (4.3%)	<0.001*	146 (37.1%)
Other invasive treatments [n (%)]	59 (31.6%)	74 (35.7%)	0.395	133 (33.8%)
Number of invasive treatments per patient [median (IQR)]	2 (1–3)	0 (0–1)	<0.001*	1 (0–2)
Dental procedures [median (IQR)]	2 (0–3)	0 (0–0)	<0.001*	0 (0–2)
Other invasive treatments [median (IQR)]	0 (0–1)	0 (0–1)	0.594	0 (0–1)

Note: *The difference reached statistical significance.

Abbreviations: AO, atypical odontalgia; IQR, interquartile range; PIFP, persistent idiopathic facial pain; SD, standard deviation; VAS, visual analog scale.

Table 2 Multiple Linear Regression Concerning Diagnostic Delay and Its Factors

	Univariate Analysis		Multiple Analysis	
	t	P	t	P
Age	-1.792	0.074	-0.806	0.421
Gender	-1.907	0.057	-1.754	0.080
Previous history of dental procedure	3.648	<0.001*	0.011	0.991
VAS	-0.943	0.346	/	/
Unilateral or bilateral	-0.100	0.920	/	/
Intraoral or extraoral	2.597	0.010*	-1.148	0.252
First consultation within 1 month after onset	-0.416	0.678	/	/
Number of consultations	3.023	0.003*	0.363	0.717
Number of prescriptions	2.222	0.027*	-0.320	0.749
Number of invasive interventions	2.192	0.029*	-0.882	0.378
AO or other PIFP	-4.829	<0.001*	-2.704	0.007*

Note: *The result reached statistical significance.

Abbreviations: AO, atypical odontalgia; PIFP, persistent idiopathic facial pain; VAS: visual analog scale.

PIFP counterparts (10,146.5 yuan vs 3864.0 yuan, $P<0.001$). Furthermore, it cost patients with AO more for both medications (3845.0 yuan vs 2855.0 yuan, $P<0.001$) and invasive treatments (5991.2 yuan vs 0.0 yuan, $P<0.001$). The summary of economic cost regarding the diagnosis and treatment is shown in [Figure 1](#).

Discussion

In this study, it was observed that patients with PIFP suffered from a median diagnostic delay of 34.8 months, during which the pain was not adequately controlled. This result was similar to that of Hassona et al, in which

a previous research advocated that the delayed diagnosis of PIFP was a common clinical scenario.⁸ Since PIFP is generally regarded as an exclusive diagnosis, it is highly possible that correct diagnosis is only established after repeated misdiagnosis and ineffective treatment measures.⁶ Despite the importance of improving knowledge and awareness of health care professionals about chronic orofacial pain conditions indicated in previous reports, the challenges of diagnosis of PIFP still requires further investigation. To our knowledge, this study includes the largest number of participants, is the first to investigate the economic burden caused by delayed

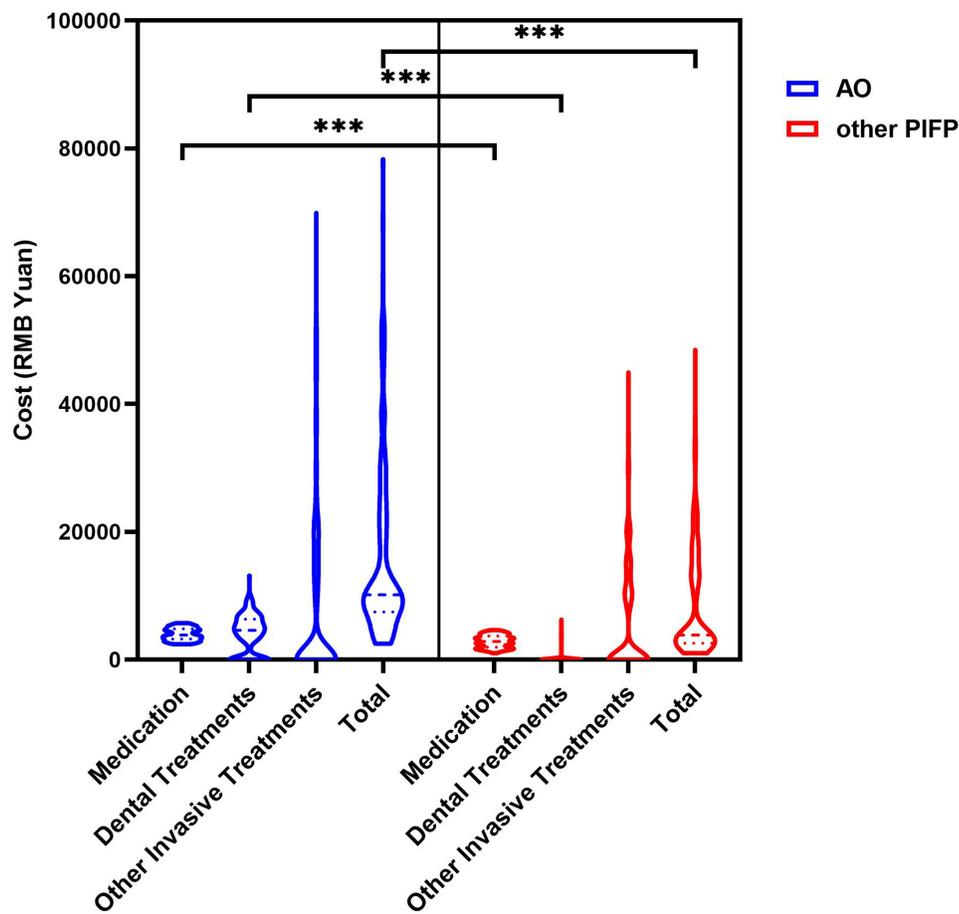


Figure 1 Medical costs before correct diagnoses of PIFP or AO.

Note: ***P value<0.001

Abbreviations: AO, atypical odontalgia; PIFP, persistent idiopathic facial pain; RMB, Renminbi.

diagnosis of PIFP, and is the first multi-centric study in this topic.

Our study found that patients with AO suffered from significantly longer delays than patients with other PIFP, which was similar to what was found by the previous study of Hassona et al.⁸ Other variables were found not factors for diagnostic delay. Although the sources of differences between AO and other PIFP require further investigation, it has been established that patients with AO face more challenges in getting their pain under control, and their conditions of delay calls for more attention. Meanwhile, in our study, we observed longer diagnostic delays than previously reported 19.3 ± 11.1 months. Although the sources for the difference in length of delay between the two studies are not easy to analyze and clarify, the significantly longer delay found in this study might indicate that it is imperative for Chinese clinicians to deepen their understanding of PIFP. Meanwhile, both studies found that patients with PIFP

face long diagnostic delays and improvements in the diagnosis of PIFP is crucial.

Long-lasting pain does not only reduce the quality of life of patients, but also negatively impacts patients' daily activities, social interaction, sleep pattern, emotional conditions and psychological conditions.^{6,8,13,14} What was worse, patients also suffered from ineffective treatments and even invasive procedures. During the average delay of 34.8 months, a median of 7 consultations were done and a median of 5 prescriptions were issued. The average number of prescriptions received per patient was rather high. Furthermore, we observed that a higher fraction of patients with AO had to undergo ineffective dental procedures, which was similar to the report of Hassona et al. Therefore, it is important for dentists to have a better understanding of the clinical features and diagnostic criteria of AO. Apart from dental procedures, we also observed that nearly one third of all participants had undergone other redundant invasive interventions. These

interventions were ineffective, invasive and expensive, also complications might be induced; therefore, awareness and caution regarding proper evaluation of these surgical indications is paramount. We observed significant linear correlation between the number of consultations, prescriptions and interventions, and the length of diagnostic delay. The establishment of correct diagnoses of PIFP or AO in an early stage after onset could have prevented patients from receiving these inappropriate managements. Although a standard treatment protocol is not available and the effectiveness of medications lack support from high-level evidences, a multidisciplinary approach which is comparable to other chronic headaches and encompasses comorbidities is recommended by some researchers.^{6,16}

This might be the first study of its kind that investigates the economic burden caused by the diagnostic delay of AO or PIFP. We found that the cost of a correct diagnosis of PIFP was 8506.3 yuan, and patients with AO spent much more, with a median of 10,146.5 yuan. It was difficult to evaluate the economic impact of a misdiagnosis without comparison. And the financial burden on the patient could not be easily described. However, a report on macroeconomics indicated that the per capita disposable income in China, in the year of 2018, was 28,228 yuan. This reflected a grim reality that the diagnostic delay cost patients more than a quarter of their annual disposable income in China. Furthermore, as the median cost for invasive treatment was 4591.0 yuan, it was apparent that if patients received invasive interventions, they would suffer even heavier economic burden.

There is no doubt that accurate diagnosis is key to proper treatment and prevention of ineffective treatments as well as socioeconomic burden.^{6,8} However, there are many problems that bring practical difficulties in timely and accurate diagnosis of patients with PIFP. Firstly, the clinical presentations of PIFP are nonspecific, symptoms of PIFP overlap with other pain syndromes, the diagnostic criteria are not precise enough and precise diagnostic tools are lacking.^{1,6,10,11,17,18} Secondly, the epidemiological characteristics of PIFP still lack adequate research and published data are also controversial. AO is described as a painful condition after tooth extraction in ICHD-3, however Miura et al reported 43.3% of their patients developed AO without such a procedure.^{1,10,19} As the prevalence of AO after endodontic treatment reportedly ranges from 2.1% to 6% and a total of 20 million endodontic procedures and 55 million tooth extractions are performed in the

United States per year, PIFP and AO could not be a reported "rare condition".^{6,11,20,21} Thirdly, despite several theories and hypotheses were proposed by previous studies, the etiology and pathological mechanisms are not well understood.^{2,9,22-29} Fourthly, there are no standardized treatment protocols established and the effectiveness of medication lacked support from high-level evidence.^{2,6,10} Evidently, addressing these problems will result in a deeper understanding of the issues and more precise reports, which in turn will improve the prevalence of diagnosis of AO and PIFP.

Apart from increasing investigations in the above aspects, education should also be emphasized. It is suggested that proper information, counseling and patient education, together with reassurance, are essential for the management of PIFP.⁸ Meanwhile, it was evident that clinicians were somewhat undereducated about this condition. We found that patients with AO were independent factors for longer delay. As dentists are commonly consulted by patients with AO, education about AO is highly recommended to this faculty of medicine. Needless to say, all related medical professionals should also be provided with adequate education about PIFP.

This study has some limitations. Firstly, this study only focuses on the diagnostic delay and economics burden of AO and PIFP. It discloses an urgent need for extended research and education on PIFP. However, our study does not provide direct knowledge to help deepen the understanding of PIFP or improve the quality of diagnosis or treatment. Secondly, because of the retrospective collection of data, potential bias may have been induced by possibly inaccurate records and the limited number of variables. A literature review suggests numerous other potential factors determining a general delay of diagnosis: pain behavior, a propensity to anxiety, social class, marital status, experience of negative dental experiences and medical history. However, we were not able to analyze these potential confounding factors due to the retrospective nature of this study. A prospective study on this topic with these aforementioned factors collected and analyzed will be performed in the future. Thirdly, as this retrospective study was conducted at four different study sites without consistent, standardized, step-by-step study guidance, sampling bias may have occurred. Fourthly, although this study includes a relatively larger population than previous studies, the scale is still not large enough, and biased results may have been generated from the study. Lastly, our study was only carried out in Beijing. It may not have been a good

representation of the entire country of China. These concerns should be addressed in future studies. However, this study did disclose the urgency and dissatisfaction of the misdiagnosis of PIFP, and more attention should be paid to proper research and education in this field.

Conclusion

Patients with PIFP are frequently misdiagnosed and the length of diagnostic delay is observed to be long. As a result of this delay, ineffective medications are prescribed and unnecessary procedures are performed. Patients with AO are in a worse situation than patients with other PIFP. Also, economic burdens are imposed by the delay of correct diagnosis. Since urgency and dissatisfaction of misdiagnosis on PIFP have been disclosed, more attention should be paid on research and education in this field.

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Disclosure

All of the authors report no conflicts of interest in this work.

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