

A study of the use of Behçet/Behçet's disease or syndrome with or without Adamantiades in the medical literature during the past two decades

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Abstract

Objective: Medical literature shows that some authors tend to use the eponym Behçet/Behçet's disease/syndrome, whereas others use its amalgamated form, that is, Adamantiades-Behçet/Behçet's disease/syndrome. We investigated the changes in the use of these eponyms over the past 2 decades.

Methods: We used the PubMed database to search for publications that contained any of these nomenclatures using the appropriate tools available. Data were obtained for the title evaluated in the database and variations of the title or its distribution by year with respect to the nomenclature found in most related studies.

Results: A total of 4,211 publications were found to use the original eponym compared with 96 publications that included the term Adamantiades from the year 2000 to the present. Considering the title of the articles, there was a declining use of the amalgamated form with Adamantiades from the first decade to the second decade, whereas there was an increasing use of Behçet/Behçet's disease or syndrome. In contrast, the relative percentages of all the reviewed articles that used the eponym disease or syndrome (without the amalgamated form) published in different specialty journals (rheumatology, ophthalmology, dermatology, and others) remained unchanged during the past 2 decades.

Conclusion: The general trend was to use Behçet/Behçet's disease or syndrome alone, although many scientists have contributed to the literature related to this topic during the historical period. The contributions of these scientists are undeniably significant, and honoring their importance is paramount.

Keywords: Behçet syndrome, rheumatic diseases, database

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Introduction

An eponym is a person, place, or thing after whom something is named or thought to be named. Traditionally, it is used to honor a scientist who was credited first with the identification, description, or publication of any disease entity or anatomical structure in the medical field, although there are no definitive rules for producing an eponym with single or multiple names instead of a long descriptive term in different cultures and languages (1, 2). Some diseases may have different eponyms, or the same eponym may refer to different diseases (3, 4). In fact, their usage may lead to some linguistic confusion or problems. In addition, there have also been discussions regarding the use of some eponyms that have been proposed to be abandoned or changed for a variety of reasons that include ethical or historical concerns (5, 6). Subsequently, some researchers have focused on various aspects of eponym use to establish a uniform strategy for editors, authors, and journals (7).

At present, Behçet's disease is used to refer to a variable vessel vasculitis characterized by recurrent aphthous oral and/or genital ulcers with cutaneous, ocular, gastrointestinal, and/or neurological system findings (8, 9). Hulusi Behçet (10) first described recurrent oral/genital lesions and uveitis with hypopyon in 1937 in "Dermatologische Wochenschrift," a new disease complex. In 1947, the findings were described as pathognomonic features of the disease, and the nomination was approved as "Morbus Behçet" by Alfred Guido Miescher (1887-1961), Professor and Chief of the University Dermatology Clinic in Zurich, at the International Congress of Dermatology held in Geneva, Italy (11). In the following years, Feigenbaum reported that Behçet's syndrome was first described by Hippocrates (12). Zouboulis and Keitel (13) also stated that Benediktos Adamantiades reported ocular symptoms, genital ulcers, and arthritis associated with a single disease. As a result, some authors have used different names for Behçet's disease or syndrome, such as Adamantiades-Behçet's disease or syndrome, in their scientific publications (14, 15). Unfortunately, differ-

ent nomenclatures have the potential to cause scientific confusion in the medical literature.

In this study, we analyzed the frequency of the difference in usage of Behçet/Behçet's disease/syndrome or Adamantiades-Behçet/Behçet's disease/syndrome during the past 2 decades to determine whether such changes have been accepted by the scientific community.

Methods

A computerized literature search was performed using the PubMed database for the period from January 2000 to present. In this study, we performed a PubMed search with the following keywords: "Behçet/Behçet disease," "Behçet's/Behçet's disease," "Behçet/Behçet syndrome," "Behçet's/Behçet's syndrome," "Behçet/Behçet disease/syndrome," and these same eponyms with "Adamantiades." We reviewed the publications containing any of these eponyms; their inclusion was not limited by the quality of the study. All article types including addresses, autobiographies, books, documents, case reports, clinical studies, meta-analyses, observational studies, or technical reports written in different languages in the world literature that were found in the PubMed database were included in the study. Data were obtained from the title search in the database. Then, the distribution by year with respect to the nomenclature of the related studies evaluated.

Ethics committee approval was not obtained because the study used data including any title or abstract obtained from the database with open access.

Statistical analysis

Data analysis was performed using the IBM Statistical Package for Social Sciences (IBM SPSS Inc.; Armonk, NY, USA) for Windows version 21.0. The data were presented as frequency (n

and percentage (%). The chi-squared test was performed to determine if there is a statistically significant difference between the expected frequencies and the observed frequencies in 1 or more variables. The Pearson chi-squared test was used if the smallest theoretical frequency was found to be >25. If it was between 5 and 25, the Yates' chi-squared test was used; and if the lowest theoretical frequency was <5, the Fisher exact test was used. The Bonferroni method was used for the differences between columns in multiple variables. A p value of <0.05 was considered statistically significant.

Results

A total of 4,211 publications were reviewed in the PubMed database during the past 2 decades, from 2000 to present. Some of the articles included the nomenclature "Behçet/Behçet disease" or "Behçet's/Behçet's disease" (n=3,863), whereas others included "Behçet/Behçet syndrome" or "Behçet's/Behçet's syndrome" (n=348). The most commonly used nomenclature was "Behçet's/Behçet's disease" (n=3,302) in the reviewed publications (Table 1). When all different specialty journals were evaluated, there were statistically significant differences in nomenclatures (with or without amalgamated form) between decades (p<0.001). In addition, the use of amalgamated form was available in the first 10 years and decreased in the second decade (p<0.001). The distribution for the use of the eponyms "Behçet/Behçet disease" (n=561), "Behçet's/Behçet's disease" (n=3,302), "Behçet/Behçet syndrome" (n=53), and "Behçet's/Behçet's syndrome" (n=295) during the period from 2000 to date in all the journals is shown in Figure 1.

The relative percentages of all the reviewed articles, which used the eponym Behçet(s) dis-

ease or syndrome (without the amalgamated form), published in different specialty journals (rheumatology, ophthalmology, dermatology, and others) remained unchanged during the past 2 decades. Among the all reviewed journals, the rates for rheumatology journals by years were 23.8%, 24.1%, 28.6%, and 25.8%, respectively. When the publications were evaluated according to their specialty, rheumatology journals and others had a tendency to use the eponyms without the amalgamated form (p<0.001), whereas ophthalmology and dermatology journals used the amalgamated form (p<0.001). Furthermore, no statistically significant difference was found between the rheumatology, dermatology, and ophthalmology journals in terms of the use of amalgamated eponyms.

No statistically significant difference was found between the first and second decades in the rheumatology, dermatology, ophthalmology, and other journals in terms of the use of "Behçet/Behçet disease," "Behçet/Behçet syndrome," and "Behçet's/Behçet's syndrome." However, a statistically significant increase was found in the use of "Behçet's disease" in the rheumatology journals (387 vs. 491, p=0.03) (Table 1).

The search for articles using "Adamantiades" published during the past 2 decades revealed a total of 96 articles. We found a decrease in the use of "Adamantiades" in the title or abstract of the articles from 70 articles over the first decade to 26 articles in the second decade (Figure 2). In the rheumatology journals, "Adamantiades-Behçet/Behçet disease" was used once during 2010-2014. "Adamantiades-Behçet's/Behçet's disease" was used 4 times during 2000-2004, 4 times during

Main Points

- Behçet's disease, which refers to vasculitis characterized by recurrent oral/genital lesions and uveitis, was first described by Hulusi Behçet in 1937.
- In the early 2000s, some authors claimed that the first description of this disease was given by Dr. Benedictos Adamantiades.
- In the past 2 decades, 4,211 publications were found to use the eponym compared with 96 publications that used the term Adamantiades.
- The trends show an increasing use of Behçet/Behçet's disease or syndrome and declining use of the amalgamated form with Adamantiades.

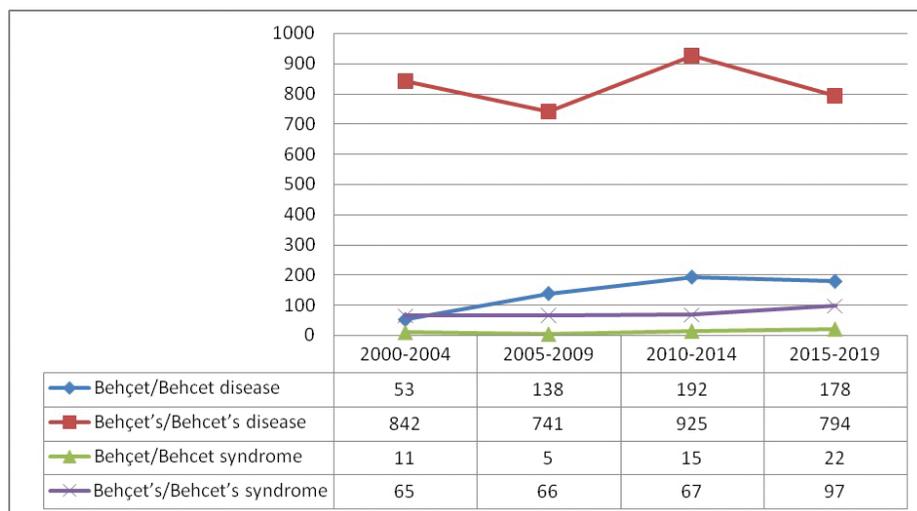
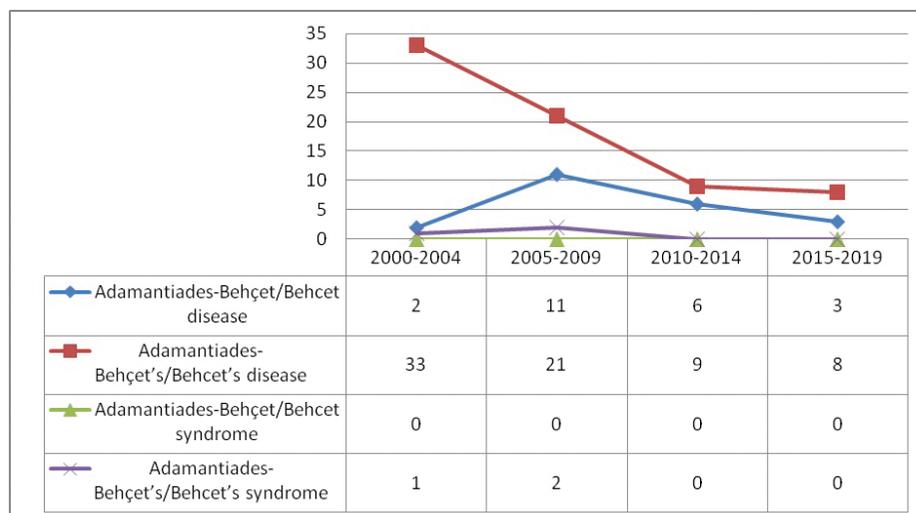


Figure 1. Distribution of different nomenclatures associated with "Behçet/Behçet's disease or syndrome" by years.

Table 1. Different nomenclatures related to "Behçet/Behçet's disease or syndrome" according to the years and different specialty journals.

	Behçet/ Behcet disease	Behçet's/ Behcet's disease	Behçet/ Behcet syndrome	Behçet's/ Behcet's syndrome	Total
2000-2004	53	842	11	65	971
Rheumatology, n (%)	5 (9.4)	208 (24.7)	1 (9.1)	18 (27.7)	232 (23.8)
Dermatology, n (%)	7 (13.2)	59 (7)	-	2 (3.1)	68 (7)
Ophthalmology, n (%)	19 (35.8)	68 (8.1)	-	2 (3.1)	89 (9.2)
Other, n (%)	22 (41.6)	507 (60.2)	10 (90.9)	43 (66.1)	582 (60)
2005-2009	138	741	5	66	950
Rheumatology, n (%)	14 (10.1)	179 (24.2)	1 (25)	35 (53.0)	229 (24.1)
Dermatology, n (%)	12 (8.7)	52 (7)	-	1 (1.5)	65 (6.8)
Ophthalmology, n (%)	27 (19.6)	60 (8.1)	-	-	87 (9.2)
Other, n (%)	85 (61.6)	450 (60.7)	4 (75)	30 (45.5)	569 (59.9)
2010-2014	192	925	15	67	1,199
Rheumatology, n (%)	23 (12)	278 (30)	3 (20)	38 (56.7)	342 (28.6)
Dermatology, n (%)	9 (4.7)	63 (6.8)	-	1 (1.5)	73 (6.1)
Ophthalmology, n (%)	39 (20.3)	60 (6.5)	-	1 (1.5)	100 (8.3)
Other, n (%)	121 (63)	524 (56.7)	12 (80)	27 (40.3)	684 (57)
2015-2019	178	794	22	97	1,091
Rheumatology, n (%)	25 (14)	213 (26.8)	4 (18.2)	40 (41.2)	282 (25.8)
Dermatology, n (%)	16 (9)	42 (5.3)	1 (4.5)	1 (1)	60 (5.5)
Ophthalmology, n (%)	24 (13.5)	57 (7.2)	1 (4.5)	4 (4.1)	86 (7.9)
Other, n (%)	113 (63.5)	482 (60.7)	16 (72.8)	52 (53.6)	663 (60.8)

**Figure 2.** "Adamantiades" alone and in its amalgamated form with "Behçet/Behçet's disease or syndrome."

2005-2009, once during 2010-2014, and twice during 2014-2019. "Adamantiades-Behçet/Behcet syndrome" has not been used in the past 2 decades. In the dermatology journals,

the use of any form (with disease/syndrome) decreased during the 2015-2019 period compared with the 2000-2004 period. "Adamantiades-Behçet's/Behcet's disease" was used

the most in ophthalmology journals during 2005-2009; however, during 2015-2019, its use decreased to just once. Moreover, there were no significant differences in the use of any amalgamated form in each of the evaluated journals (rheumatology, dermatology, and ophthalmology) between the first and second decades within the amalgamated nomenclatures group. The distribution for "Adamantiades-Behçet/Behçet's disease or syndrome" according to the years and different specialty journals is shown in Table 2.

According to the distribution by country, depending on the affiliations of corresponding authors of the reviewed publications, 44.8% of the publications with "Adamantiades" were from Germany, 18.8% from Greece, 8.3% from the United States of America, 5.2% from Japan, and the remaining 22.9% from other countries. Considering the distribution of the publications included in the study, there was a significant difference between the studies using the eponym "Behçet/Behçet's Behçet(s) disease" or "Behçet(s)syndrome" and its amalgamated form with "Adamantiades" in the world literature during the past 2 decades ($p < 0.001$).

Discussion

It is widely accepted that Hulusi Behçet identified the presence of mouth and genital ulcers as the hallmarks of the disease (10, 11). Adamantiades, however, was only able to identify the ocular findings associated with arthritis and the mucocutaneous symptoms of the disease (16). In fact, historically, some researchers, including Gilbert (17), Shigeta (18), Fuchs (19), and Planner and Remenovskiy (20), have also made similar contributions in recognizing the clinical features of the disease before Hulusi Behçet. In the early 2000s, however, some authors claimed that the first description of this disease was made by Dr. Benedictos Adamantiades (13, 21-23). It was suggested that Adamantiades first reported the disease in 1930 to the Athens Medical Association and then published that case report in the French journal "Annales d'Oculistique" in 1931 (16, 24).

In 2001, Cheng (25) reported in his article titled "Some historical notes on Behçet's disease" that Hippocrates had described the disease for the first time in the fifth century, which he called "silk route disease" owing to its frequent occurrence between the Northern latitudes of 30° and 45°. Tirilomis (22) suggested that the "silk route disease" should be referred to as "Adamantiades-Behçet's disease" because of the contributions

Table 2. Different nomenclatures related to "Adamantiades-Behçet/Behçet's disease or syndrome" according to the years and journals.

	Adamantiades-Behçet/ Behçet disease	Adamantiades-Behçet's/ Behçet's disease	Adamantiades-Behçet/ Behçet syndrome	Adamantiades-Behçet's/ Behçet's syndrome	Total
2000-2004	2	33	-	1	36
Rheumatology, n (%)	-	4 (12.1)	-	-	4 (11.1)
Dermatology, n (%)	-	6 (18.1)	-	-	6 (16.7)
Ophthalmology, n (%)	-	1 (3.1)	-	-	1 (2.8)
Other, n (%)	2 (100.0)	22 (66.7)	-	1 (100.0)	25 (69.4)
2005-2009	11	21	-	2	34
Rheumatology, n (%)	-	4 (19.0)	-	-	4 (11.8)
Dermatology, n (%)	4 (26.4)	4 (19.0)	-	-	8 (23.5)
Ophthalmology, n (%)	2 (18.2)	7 (33.3)	-	-	9 (26.5)
Other, n (%)	5 (45.4)	6 (28.7)	-	2 (100.0)	13 (38.2)
2010-2014	6	9	-	-	15
Rheumatology, n (%)	1 (16.7)	1 (11.1)	-	-	2 (20.3)
Dermatology, n (%)	1 (16.7)	1 (11.1)	-	-	2 (13.3)
Ophthalmology, n (%)	2 (33.3)	1 (11.1)	-	-	3 (20.0)
Other, n (%)	2 (33.3)	6 (66.7)	-	-	8 (53.4)
2015-2019	3	8	-	-	11
Rheumatology, n (%)	-	2 (25.0)	-	-	2 (18.2)
Dermatology, n (%)	-	1 (12.5)	-	-	1 (9.1)
Ophthalmology, n (%)	2 (66.6)	3 (37.5)	-	-	5 (45.4)
Other, n (%)	1 (33.3)	2 (25.0)	-	-	3 (27.3)

of both of these scientists. Cheng (26) later suggested the eponym "Hippocrates-Adamantiades-Behçet disease" in his reply text titled "Behçet Disease, Adamantiades-Behçet Disease, or Hippocrates-Adamantiades-Behçet Disease?" in 2002. In 2002, Zouboulis (13), in his article titled "A historical review of early descriptions of Adamantiades-Behçet's disease," called the disease "Adamantiades-Behçet's disease" to honor both the early describers of the disease in modern times. Furthermore, in one of the chapters in the book "A Historical Review of Adamantiades-Behçet Disease," the same name was used (27). In addition, in a chapter written by Zouboulis (28) in the textbook titled "Dermatology in General Medicine," this subject was later included in the agenda.

The use of the disease name without its amalgamated form "Adamantiades" has been widely accepted. The disease is named "Behçet disease" in the International Classification of Diseases, 10th Revision list by the World Health Organization, and the eponym Behçet's disease was also used and classified

as a variable vessel vasculitis at the 2012 Revised International Chapel Hill Consensus Conference Nomenclature of Vasculitides (8, 29). In addition, the title of the 2018 update of the European League Against Rheumatism (EULAR) recommendation was changed to "EULAR Recommendations for the Management of Behçet's Syndrome" (30). Some of the experts preferred that the disease be considered a "syndrome," whereas others disagreed and voted that it be considered as a "disease" or "neutral" (30). Nonetheless, it has been stated that the issue should be discussed further in the future (30).

To the best of our knowledge, this is the first study investigating the use of the eponym "Behçet/Behçet's disease" or "syndrome" with/without its amalgamated form "Adamantiades" in the literature. The findings from this study clearly revealed that the numbers of articles using the eponym "Behçet/Behçet's disease" or "syndrome" without its amalgamated form "Adamantiades" were far more in number than those using the amalgamated form "Adamantiades" in the past 2 decades.

Undoubtedly, there are naming violations of diseases that may be considered unfair to other scientists who have scientifically contributed to the better understanding of these diseases. The results of our PubMed search showed that the use of "Adamantiades" in the titles of articles has decreased over the past 2 decades, whereas there was an increasing tendency to use "Behçet/Behçet's disease or syndrome" (Figures 1 and 2). The limitation of this retrospective study is that we searched only PubMed database from the past 2 decades (2000-2019) and did not include oral presentations and printed brochures.

In conclusion, the general trend is to use Behçet/Behçet's disease or syndrome, although the symptoms of this disease or syndrome have been repeatedly described during the historical process, and many scientists have contributed to the literature regarding this topic. We believe that the contributions of these scientists have added immense value to the medical literature, and they should be honored.

Ethics Committee Approval: Ethics committee approval was not received.

Informed Consent: Informed consent was not obtained due to the nature of this study.

Peer-review: Externally peer-reviewed.

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