

## Barré–Lieou syndrome and the problem of the obsolete eponym

C A FOSTER, P JABBOUR\*

### Abstract

**Background:** Eponym lists in major sources can give an aura of legitimacy to discredited diagnoses, as exemplified by the case of Barré–Lieou syndrome, a ‘rare’ vestibular disorder.

**Methods:** A literature review for information on the posterior cervical syndrome of Barré–Lieou.

**Results:** Barré–Lieou syndrome includes very common symptoms – tinnitus, dizziness, and head or neck pain – attributed to ischaemia caused by cervical sympathetic nerve compression. Its original description brings together many unrelated disorders, and its causative mechanism has been discredited. However, it appears credulously in a number of eponym lists, and references to the syndrome are steadily increasing on the internet in general and on alternative medicine and legal profession websites in particular.

**Conclusion:** By inclusion in eponym lists, without a disclaimer, a syndrome can be given legitimacy before the general public. A syndrome, such as Barré–Lieou syndrome, that is useless to the medical profession can unfortunately prove to be very useful for litigants and disability claimants.

**Key words:** Autonomic Nervous System Diseases; Eponyms; Spinal Osteophytosis; Vascular Headaches; Vertebrobasilar Insufficiency; Vestibulocochlear Nerve Diseases

### Introduction

Eponyms persist because they are useful. Naming a complex syndrome allows it to be discussed in an efficient manner, avoiding the need to provide details repeatedly in writing and conversation. Such eponyms also honour discoverers with eponymous fame and immortality.<sup>1</sup> Eponyms were necessary in the past, when decades could pass between syndrome recognition and an understanding of its pathology. Today’s more rapid determination of aetiology favours the use of pathologic descriptive names or acronyms, such as AIDS (acquired immunodeficiency syndrome) and CADASIL (cerebral autosomal dominant arteriopathy with sub cortical infarcts and leukoencephalopathy). This has been lamented by some. Eponyms act as reminders of the profession’s background and so provide an element of historical respect.<sup>2</sup> This leads many editors to include eponym sections in their medical texts. However, these lists may inadvertently serve to prolong the life of eponyms that have been previously discarded for lack of merit. This is of particular danger when no clear refutation of the syndrome has been made in the literature. The original papers may be obscured by antiquity, foreign language publication, or appearance in inaccessible

formats such as hospital bulletins, leaving an eponym that has become orphaned from its historical background and is then vulnerable to rediscovery and misuse.

The posterior cervical syndrome of Barré–Lieou, an eponym used in otolaryngology, orthopaedics, neurology and neurosurgery, illustrates this problem. It is a ‘rare’ syndrome combining the very common symptoms of dizziness, head or neck pain, and tinnitus. It was based on an incorrect theory of disease, but no refutation was published. No scientific review of Barré–Lieou syndrome has appeared in the English literature in the last 50 years, but it has been resurrected in modern eponym lists, lending legitimacy to the diagnosis. While no longer useful in traditional medicine, it is extremely useful in the medicolegal, disability and alternative medicine communities.

### Materials and methods

A literature search was performed using Cumulated Index Medicus and Medline, using the key words ‘Barré–Lieou’, ‘Barré’, ‘sympathetic nervous system’ and ‘vertebral artery compression’, for the period 1925 to 2005. Articles by Barré, Lieou or Neri

From the Departments of Otolaryngology/Head and Neck Surgery, and \*Neurosurgery, University of Colorado Health Sciences Center, Denver, Colorado, USA.

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referring to sympathetic compression, and articles in English referencing the syndrome, sympathetic dysfunction or vertebral artery compression as a cause of vertigo, were also reviewed. Internet searches (using the Google search engine) were performed from February 2004 through to August 2005 using the search term 'Barré-Lieou'.

### Results and analysis

The posterior cervical syndrome of Barré-Lieou was described in three French papers, two published in 1926 by Jean-Alexandre Barré (1880–1967)<sup>3,4</sup> and a doctoral thesis by his Chinese protégé Yong-Choen Lieou in 1928.<sup>5</sup> Barré was a neurologist at the University of Strasbourg with a prolific publication record, who had achieved eponymous fame for a 1916 paper describing Guillain-Barré syndrome. Lesions of the sympathetic nerves along the carotid were known to cause an 'anterior cervical syndrome' (Horner's syndrome). Barré hoped to define a corresponding 'posterior cervical syndrome' affecting sympathetic nerves along the vertebral arteries. However, he was unable to identify any consistent neurologic signs in his cases. He thus defined his disorder as opposite to Horner's: posterior rather than anterior in location, and subjective rather than objective in its findings. No detailed case reports or data were presented. Symptoms included vertigo when tipping the head, headache, tinnitus, reading difficulty, vocal weakness and hysterical conversion reaction. He theorized that sympathetic nerves regulated intracerebral blood flow and that cervical spine disease could irritate or compress them, resulting in transient intracerebral ischaemia. In a 1952 published lecture,<sup>6</sup> Barré expanded the causation to include sympathetic compression in the thorax and by the skin, musculature and periosteum in the neck.

Lieou's 97-page dissertation<sup>5</sup> presented 10 cases with disparate symptoms, all including pain in the head or neck, divided into three subtypes. Six patients had the vertiginous form and four were assigned either to a facial form (with symptoms of trigeminal neuralgia or cluster headaches) or a pharyngeal form (with hoarseness or dysphagia). The typical pain could occur in any part of the head, ear or neck and ranged from mere heaviness to profound, hammering pain. The vertigo varied from brief, paroxysmal positional vertigo to episodes lasting hours. The tinnitus ranged from pulsatile to continuous, often with hearing loss. Although Barré reported an absence of objective findings as characteristic, Lieou's work contradicted Barré by describing a number of objective abnormalities in these patients.

Both cited a 1924 paper<sup>7</sup> by Vincenzo Neri, an Italian neurologist, to support their theory. In a case report of a neck wound associated with migraine, vertigo and depression, Neri hypothesized an impairment of cerebral circulation, and referred to Horner's work on the anterior cervical sympathetics. Because of Neri's case precedence, the disorder is sometimes referred to as Neri-Barré-Lieou syndrome.

Nineteen articles were published in European journals prior to the first English language paper. In 1954, a review by Gayral and Neuwirth appeared,<sup>8</sup> and most subsequent English references cited this paper. It contained a brief presentation of three cases, with symptoms expanded to include coughing, corneal ulcers, generalized pain and various psychiatric disorders. Vestibular symptoms ranged from simple unsteadiness to severe vertigo. The cause was again ascribed to ischaemia from cervical compression of the vertebral sympathetics. An additional 57 scientific articles were published between 1954 and 2005. Eleven were in English and appeared in orthopaedic and chiropractic journals; none appeared in journals of otolaryngology, neurosurgery or neurology. A recent historical note questioned the validity of the diagnosis<sup>9</sup> but did not review the original articles in detail.

In 1983, Barré-Lieou syndrome was mentioned as an eponym in a major otolaryngology textbook,<sup>10</sup> with symptoms (including occipital headache, vertigo, tinnitus, vasomotor disorders and facial spasm, or 'cervical migraine') attributed to irritation of the sympathetic plexus causing vertebrobasilar insufficiency. It was subsequently listed in another otolaryngology textbook<sup>11</sup> as a syndrome combining neck pain radiating to the occiput, vertigo, tinnitus and 'clouded vision'. Barré-Lieou syndrome is listed in the National Institutes of Health Office of Rare Diseases database<sup>12</sup> as combining headache, tinnitus, vertigo and impaired concentration. It also appears on several internet eponym lists. No scepticism regarding the diagnosis or causation is contained in these references. Articles published in the last few decades which refer to Barré-Lieou syndrome apply the name to vertebrobasilar insufficiency due to extrinsic vertebral artery compression rather than to sympathetic irritation, although Barré never proposed the former aetiology in his publications.

In February 2004, an internet search on 'Barre Lieou' revealed 345 'hits'. The following year (August 2005), these had more than doubled, reaching 861. These hits were predominantly websites for prolotherapy, chiropractic and alternative medicine, and medical information. Manipulative or surgical treatments for the neck were often recommended. The syndrome was featured in a Colorado legal bulletin as an important diagnosis for disability and accident cases.<sup>13</sup> When patients bearing this diagnosis were evaluated at the University of Colorado vertigo clinic, diagnoses ranging from malingering to migraine-associated vestibulopathy were made, and none were found to have vertebrobasilar insufficiency.

### Discussion

Barré-Lieou syndrome is not a useful eponym, for two reasons.

Firstly, its symptoms are not unique, being common to many vestibular disorders, all of which could be included in its spectrum. Cervical arthritis, tinnitus and headache are extremely common and

so a number of people will have this combination purely by chance. Vertigo, tinnitus and headache associate in several different vestibular disorders, as noted by Ménière 60 years before Barré.

Secondly, the cause of the syndrome has been discredited. Barré attributed it to compression or irritation of the vertebral sympathetic nerves, causing vasoconstriction and secondary ischaemia. In the 1920s, the function of the posterior cervical sympathetics was not known, although the vasoconstrictive role of the sympathetic nervous system in general was understood. It is now believed that the posterior sympathetics act to dampen sudden increases in blood pressure, protecting the brain capillary bed from large pressure fluctuations that could otherwise violate the integrity of the blood–brain barrier.<sup>14,15</sup> Denervation has no apparent ill effects in normotensive individuals, and stimulation does not cause vasoconstriction.<sup>16,17</sup> In the laboratory, stimulation modestly reduces cerebral blood flow, primarily in the setting of acute hypertension.<sup>18</sup> Thus, Barré and Lieou's theory that sympathetic compression could cause symptomatic ischaemia has been disproven.

While eponyms are valued for honoring eminent discoverers, they have serious shortcomings. Unlike a pathologic description, a person's name does not provide a helpful clue to the disease aetiology or even to the organ affected.<sup>19</sup> The names used for a syndrome may vary over time or between countries.<sup>20</sup> The syndrome itself may be ambiguous or lacking in utility.<sup>21</sup> Eponyms may also confer honour on the wrong individual or on undesirable personages (for example, a Nazi scientist experimenting on concentration camp inmates).<sup>22</sup> The posterior cervical syndrome of Barré–Lieou illustrates another shortcoming of eponyms: a tendency to re-emerge from deserved obscurity. The lack of English translations and difficult access to both the original reports and most subsequent articles hinders objective review of this syndrome, a problem common to many older syndromes.

- **The posterior cervical syndrome of Barré–Lieou, described in 1926, is included in major eponym lists as a rare vestibular disorder, and citations of it are increasing**
- **The pathophysiology and utility of the syndrome as a diagnosis have not previously undergone detailed scientific review**
- **In this paper, the syndrome is reviewed and found to be invalid as a medical diagnosis, but it is noted to be increasingly used by alternative medicine practitioners and the legal profession**
- **Inclusion in a credulous eponym list can give the appearance of legitimacy to a discarded diagnosis**

The dearth of citations in the current literature reflect an understanding by the medical profession that it is no longer valid, but the absence of a refuting

article and the syndrome's listing as an eponym in credible sources lends support to its legitimacy. We found that internet references to the syndrome have in recent years been doubling annually on sites not associated with traditional medicine. The absence of objective findings within the definition of Barré–Lieou syndrome makes it impossible to exclude the diagnosis, and the syndrome offers a way to tie neck injuries to a plethora of head and neck symptoms. This makes it a 'dream syndrome' in the context of medicolegal and disability consultation, since neck injuries are a major cause for such referrals.

## Conclusion

Barré–Lieou syndrome is not valid, yet it is treated as if legitimate in major sources that can be consulted by the public. Eponym lists, by their nature, publicize obscure syndromes. Some may be obscure because they are rare, but list editors and readers need to be aware that obscurity can also indicate that the eponym has been discarded for lack of merit. When an eponym is credibly listed, it encourages re-use by people for a variety of reasons, some of which may be antithetical to patient well-being and the progress of medicine. In such lists, a thorough review of the eponym prior to listing, and notation that some names may be of historical interest only, would help prevent these undesirable consequences.

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## References

- 1 Cooley DA. In pursuit of an eponym. *Texas Heart Inst J* 2004;**31**:117
- 2 Lee P, Hunter TB, Taljanovic M. Musculoskeletal colloquialisms: how did we come up with these names? *RadioGraphics* 2004;**24**:1009–27
- 3 Barré JA. On a posterior cervical sympathetic syndrome and its frequent cause: cervical arthritis [in French]. *Rev Neurol (Paris)* 1926;**33**:1246–8
- 4 Barré JA. Critical studies on the phenomenon of sympathetic irritation and paralysis, circulatory dysfunction due to the sympathetic system, sympathetic pain sensitivity, the posterior sympathetic syndrome, the value of pharmacological tests and the importance of sympathetic dysfunction in hysteria [in French]. *Rev Neurol (Paris)* 1926;**33**:1046–9
- 5 Lieou YC. Posterior cervical sympathetic syndrome and chronic arthritis of the cervical vertebral column (clinical and radiological study) [in French]. Strasbourg, University of Strasbourg, 1928
- 6 Barré JA. Neuro-otologic and ophthalmologic dysfunction of cervical origin [in French]. *Rev de Otoneuroophthal* 1952;**24**:18–21
- 7 Neri V. Cervical sympathetic cerebral syndrome [in Italian]. *Bologna Soc Med* 1924;**96**:382
- 8 Gayral L, Neuwirth E. Oto-neuro-ophthalmologic manifestations of cervical origin. Posterior cervical sympathetic syndrome of Barré–Lieou. *N Y State J Med* 1954;**54**:1920–6
- 9 Pearce JMS. Historical note. Barré Lieou "syndrome". *J Neurol Neurosurg Psychiatry* 2004;**75**:319

- 10 Lee KJ, Lee ME. Syndromes and eponyms. In: Lee KJ, ed. *Essential Otolaryngology Head and Neck Surgery*. Stanford CA: Appleton & Lange, 1999;197
- 11 Jones K, Pillsbury HC. Eponyms in otolaryngology. In: Bailey BJ, Calhoun K, eds. *Head & Neck Surgery-Otolaryngology*. Philadelphia PA: Lippincott Williams & Wilkins, 2001;2479
- 12 National Institutes of Health. <http://ord.aspensys.com/asp/diseases/diseases.asp> [17 August 2005]
- 13 Boulder County Bar Association Newsletter. [http://www.boulder-bar.org/pdf/bcba\\_2\\_01.pdf](http://www.boulder-bar.org/pdf/bcba_2_01.pdf) [17 August 2005]
- 14 Faraci FM, Heisted DD. Regulation of large cerebral arteries and cerebral microvascular pressure. *Circ Res* 1990;**66**:8–17
- 15 Sadoshima S, Fujishima M. Hypertension and the auto-regulation of cerebral blood flow III. Trophic influences on vascular wall. In: Phillis JW, ed. *The Regulation of Cerebral Blood Flow*. Boca Raton FL: CRC Press, 1993;335
- 16 Sadoshima S, Heisted DD. Regional cerebral blood flow during hypotension in normotensive and stroke-prone spontaneously hypertensive rats: effect of sympathetic denervation. *Stroke* 1983;**14**:575–9
- 17 Mueller SM, Heisted DD, Marcus ML. Total and regional cerebral blood flow during hypotension, hypertension and hypocapnia. Effect of sympathetic denervation in dogs. *Circ Res* 1977;**41**:350–6
- 18 Tuor UI. Acute hypertension and sympathetic stimulation: local heterogenous changes in cerebral blood flow. *Am J Physiol Heart Circ Physiol* 1992;**263**:H511–51e
- 19 Aetna Intellihealth Harvard Commentaries. <http://www.intelihealth.com/IH/ihtPrint/EM/35320/35328/> [13 April 2006]
- 20 British Medical Association. [www.bma.org.uk/ap.nsf/Content/LIBSyndromes](http://www.bma.org.uk/ap.nsf/Content/LIBSyndromes) [13 April 2006]
- 21 Mirilas P, Colborn GL, Skandalakis LJ, Skandalakis PN, Zoras O, Skandalakis JE. Benign anatomical mistakes: “ampulla of Vater” and “papilla of Vater”. *Am Surg* 2005;**71**:269–74
- 22 Shevell M. Hallervorden and history. *N Engl J Med* 2003;**348**:3–4

## Address for correspondence:

Dr Carol A Foster,  
Department of Otolaryngology/Head & Neck Surgery,  
University of Colorado Health Science Center,  
PO Box 6510,  
Mail Stop F736,  
Aurora CO 80045, USA.

Fax: 720 848 2857

E-mail: carol.foster@uchsc.edu

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