From "Shaking Palsy" to "Parkinson's Disease" and Charcot's *Lateral Extension* type PD.

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Abstract

In light of contemporary knowledge of Parkinson's disease, we review a classic case with photographic evidence presented by Charcot in which his patient, Bachère, was instrumental in changing the name of the disease from "shaking palsy". We also examine 4 cases of *extension type* Parkinson's disease (a description no longer in use) especially that of Léon as described in 2 articles (1889 and 1892) in *Nouvelle Iconographie de la Salpêtrière*. Although Charcot diagnosed Bachère's condition as a type of Parkinson's disease, and historically it has been accepted as such, we believe he may have been mistaken.

Key words: shaking palsy, Parkinson's disease, Bachère, Léon, *extension type* Parkinson's disease

Introduction

The following paper, based on a Japanese language version (BRAIN and NERVE, August 2019) by MK, TO and RK has been adapted by MWM. The translators, from Japanese to English, and French to English, are both acknowledged at the end of the paper.

James Parkinson (1755-1824) published *An Essay on the Shaking Palsy* in 1817¹. He described three of his own patients and three cases observed on the street. Aside from his failure to note muscular rigidity or sensory-perceptual impairment and to identity relative lesions in cervical spinal cord, he described the disease accurately, and his description has survived for over 200 years. Unfortunately, his Essay did not gain widespread, contemporary acceptance, its print run was small and today there are no more than 8 known, first edition copies in existence. However, before we proceed it is important to note that the term *paralysis agitans* or "shaking palsy" was first used by Marshall Hall (1790-1857) in his Principles of Diagnosis (1834)².

Jean-Martin Charcot (1825-1893) read Parkinson's Essay in an effort to better understand multiple sclerosis and hand tremor. His copy, obtained with difficulty from the University of Manchester, impressed him very much, and in one of his Tuesday Lessons on 12 June 1888³ he introduced a patient called Bachère and suggested renaming paralysie agitante (shaking palsy) to la maladie de Parkinson in honor of the pioneering neurologist⁴. He was also concerned with the inaccuracy of the original description as demonstrated by paralysis-free patients with mild tremor such as Bachère, and by patients' aversion to the term "palsy." However Charcot arrived at his decision, the Bachère case is important in that it marks the end of "shaking palsy" and the beginning of "Parkinson's disease".

Between January 2016 and December 2018 *BRAIN and NERVE* published a "Cover of the Month" sourcing articles in French from *Nouvelle Iconographie de la Salpêtrière* (NI): first published in 1888. Each article featured a case with distinct neurological symptoms plus original, photographic material

Of the 36 cover photographs two concerned Parkinson's disease (PD). One is a case report from the second issue (NI 1889)^{5,6} by Adolphe Dutil (1862-?). This describes a female patient with abnormal extended posture as "a case of shaking palsy–a hemiplegic type with abnormal posture (extension) of the head and trunk". The other is a review of three cases (two male, one female) from the fifth issue (NI 1892)^{7,8}. Two of these cases are described as showing the "peculiar postures observed in Parkinson's disease". The reports were published around the same time as Charcot's 20th *Tuesday Lessons* in which he introduced Bachère. The disease was thus defined as "paralysie agitante" in the earlier case report and as "la maladie de Parkinson" three years later and makes Bachère's case, metaphorically speaking, a watershed in PD symptomology.

Joseph Jules Dejerine (1849-1917) published his major work, *Sémiologie des Affections du Système Nerveux*⁹ in 1914. Interestingly, its index does not refer to "paralysie agitante" but to "la maladie de Parkinson". It also includes two photographs of Bachère, showing his facial expressions and abnormal posture. Taken from *The Tuesday Lessons*, these photographs have succinct footnotes by Dejerine on Bachère's case. Furthermore, "Parkinson's disease in *extension*" was clearly noted in a classical textbook in English: *A Manual of Diseases of the Nervous System* (1900) ¹⁰ by William Gowers (1845-1915). In our review, we discuss not only Bachère's case but also Goetz'¹¹ English translations of Charcot's *Tuesday Lessons* and review four cases with possible connection to Léon, presented by Eugène Béchet (1862-1939).

I. Bachère¹²

In 1888, when Charcot presented his case, Bachère was about 30 years old. Early symptoms had appeared at the age of 20 and he suffered three rheumatic attacks between 12Y and 20Y leading to facial masking. He also had deep forehead lines that Charcot interpreted as rigidity of the frontalis and occipital muscles or the "muscles of surprise". Other facial muscles were also frozen making him expressionless. However, he showed no tremor and retained a normal grip. Unique aspects of his case included a posture with bilateral extension of upper and lower limbs.

Many readers will be familiar with Bachère's face without knowing his name. He is possibly the best known case of PD since both Charcot (Fig. 1) and his pupil, Paul Richer¹³ (1849-1933, Fig. 2), made sketches of him that are still used to illustrate PD facies. However, his features may not be typical of PD. Goetz¹¹, for example, cites Jankovic¹⁴ to say that the look of surprise is seen in patients with progressive supranuclear palsy (PSP). Moreover, Goetz suggests that Dutil's female patient^{5,6} did have PSP as indicated by her facial features. Dutil describes this patient as being one of "shaking palsy–a hemiplegic type with abnormal posture (extension) of the head and trunk".

The face is masked, the forehead wrinkled, the eyebrows raised, the eyes immobile.... This facies, associated with the extended posture of the head and trunk, gives the patient a singularly majestic air.

[https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3234454/]

Next, let us consider how Dejerine viewed Bachère's case. As noted above he included him, plus two photographs, in his textbook⁹. Bachère was not named. However, the footnote to the photograph says that Charcot had presented him in one of his *Tuesday Lessons* (Fig. 3). Interestingly, while Dejerine published his textbook after he became a professor at Pitié-Salpêtrière Hospital his observation of Bachère must have occurred at least 20 years earlier while working at the Bicêtre Hospital: indeed, the photograph carries the caption: "Bicêtre 1893." Furthermore, Charcot concluded his own remarks on Bachère as follows:

The physiognomy is expressionless; the brow crossed by transversal furrows, the eyes motionless, the eyelids blinking very infrequently. This facies, combined with the upward thrust of the head and the trunk, gives the patient a very particular look of majesty (after Goetz' translation¹¹).

In our view when Bachère was presented in 1888 he was already scheduled for transfer to Bicêtre, where Dejerine was working. Furthermore, Charcot's prognosis was incorrect and Bachère was still alive five years later. Given current therapeutic guidelines on treatment including L-DOPA and the latest treatments (described in the Japanese version of this paper in (Brain and Nerve, August 2019) the contrast between now and then as to prognosis could not be greater.

The most reliable account of Dejerine's career, we believe, is that provided by Édouard Gauckler¹⁵ (1858-1924) in 1922. According to Gauckler, Dejerine worked at the Bicêtre Hospital from 1887 to 1894. If so, then he was there when Charcot presented Bachère in 1888. Subsequently, Dejerine moved to the Jacquart Unit of the Pitié-Salpêtrière Hospital in 1894 and became a professor in the neuropathology clinic in 1911. In his textbook, Dejerine described Bachère as a "36-year-old patient with Parkinson's disease–*extension type*–with onset at age 21 after severe trauma (with) marked propulsion" (Fig. 4). The description, "onset after severe trauma" occurs nowhere else, and if Dejerine was correct, then differential diagnoses for Bachère should include post-traumatic conditions such as subdural hematoma.

We should now like to examine Bachère's case with regard to the diagnostic criteria of The International Parkinson and Movement Disorder Society (MDS). Interestingly, it does not meet these because of its atypical symptomology. With regard to the patient's idiosyncratic facial features, his significant extended posture with bradykinesia of the head, and the young age of onset, Goetz suggests the possibility of PSP, post-encephalitic Parkinson's syndrome or the rigid form of Huntington's disease¹¹. We agree with this and also emphasize the post-traumatic onset reported by Dejerine. Whatever the precise etiology, Bachère is likely to have had a parkinsonian syndrome rather than PD. If this is the case it is time to revisit Charcot's classic diagnosistic switch from shaking palsy to PD.

II. Léon

Aside from the usual photograph of Bachère, we recently came across that shown in Fig. 5. An enlarged version of the same was shown, together with historical works by Parkinson, in the exhibition hall during the Alzheimer's and Parkinson's Diseases (AD/PD) Congress in Vancouver in 2017. There is also a version on Wikipedia.

When writing "Cover of the month" comments, we first digitized all the NI photographs and projected them onto a screen to make our selections. It was only after deciding which photographs to use that we read the source articles. These clinical photographs were accompanied by excellent symptom details, and many made a deep impression. We wanted the cover images to be impressive, have aesthetic value, and to provide specific information.

The photo of Léon, standing up from a chair and starting to walk, evoked deep feelings in all of us. Although a still image, it appears to animate his short-step gait. We also believed that it represented a typical case of PD. However, this was not the case. Fig. 5 was used on the cover of the October 2016 issue of *BRAIN and NERVE* and our comments¹¹ included remarks made after Béchet⁷ who presented Léon, Case III, as follows:

Ayant eu l'occasion depuis quelques mois, d'observer à la Salpêtrière, un assez grand nombre de sujets atteints de maladie de Parkinson, nous en avons rencontré quelques-uns dont l'aspect s'éloigne notablement du type classique, et nous avons pensé que la description de leur attitude n'était pas sans présenter quelque intérêt. Deux d'entre eux, M. Beh.^{*1} et Mme Ot.^{*2}, nous ont paru mériter de prendre place à côté des cas décrits par M. le professeur Charcot sous le nom de type d'extension dont deux exemples remarquables ont été rapportés dans ce journal par M. P. Richer en 1888^{*3}, et par M. Dutil en 1889^{*4}.

(Having had the opportunity for some months now to observe a significant number at the Salpêtrière suffering from Parkinson's disease, we have encountered some whose appearance differs noticeably from the classic type, and we concluded that there would be

some interest in describing their posture. Two of them, M. Beh and Mme Ot., seemed to us to be worth placing alongside the cases described by Professor Charcot in the category of *extension type*, two remarkable examples of which were reported upon in this journal by P. Richer in 1888 and Dutil in 1889).

OBS. III^{*5}. Rab. Léon, 37 ans, boucher, est venu le 21 juin, consultation du mardi, . . . Il s'avance le corps penché en avant, la tête immobile sur les épaules, tout d'une pièce ; la démarche précipitée se fait de la façon suivante : le malade semble prendre appui presque uniquement sur le pied gauche ; il incline fortement le tronc de ce côté, le membre inférieur droit en extension soulevé à un centimetre à peine du sol, est porté, directement en avant et retombe lourdement, puis la jambe gauche est à son tour portée en avant comme dans la marche physiologique ; la cadence du pas se précipite, le corps s'incline de plus en plus en avant, et le malade est menacé de tomber ; la rétropulsion existe également, mais à un degré moindre.

[http://jubilotheque.upmc.fr/fonds-nvicono/CS_000005_005/document.pdf?name=CS_0000 05_005_pdf.pdf]

*1: Case I; *2: Case II; *3: 12; *4: 4; *5: Léon

(Obs. III. Rab., Léon, aged 37, a butcher, came on 21 June to the Tuesday consultation, where Professor Charcot presented his case with those of two other patients affected by the same condition but manifesting different external characteristics. He moves with his body leaning forwards, his head fixed on his shoulders in a single block; accelerated motion occurs as follows: the patient appears to put his weight solely on the left foot; he leans his trunk sharply to that side, while the extended right leg, which is raised barely a centimeter from the ground, is brought straight forward, coming down heavily; then in turn the left leg is brought forward as in normal walking; the pace speeds up, the body leans further and further forwards, and the patient is in danger of falling. Retropulsion can also occur, but to a lesser degree).

A close inspection of this photo shows that Léon has a straight neck without dropping his head. To us, this posture resembled that of a patient with the *extension type* condition presented by Dejerine (Fig. 4: in fact this shows Bachère as we explain below). Although not explicitly stated by Béchet, we considered the possibility that Léon also had the so-called *extension type* condition and we included this in our "Cover of the month" comments for the October 2016 issue of *BRAIN and NERVE*⁸.

Additional research established that Léon was originally described by Béchet as having atypical PD⁶ and we believe he had so-called PD in *extension*⁷.

The original, NI, photographic negatives of Léon were prepared by Albert Londe (1858-1917). And NI began in 1888 on Charcot's initiative with three core members: Londe, Richer and Georges Gilles de la Tourette (1857-1904). The preface to the first issue declared that its purpose was to make use of numerous illustrated reports accumulated at Pitié-Salpêtrière Hospital.

Going back a little in the history of photography, the first commercially available process began with Louis Daguerre (1787-1851) in which a single image was directly fixed onto a metal plate and the sitter, in the case of a portrait photograph, would have his head immobilized by a clamp. However, subsequent developments made exposure times shorter, prints reproducible and equipment portable, and by the 1880s, sequential photography was possible and began to influence the medical sciences. The earliest applications include microscopic photography by Alfred François Donné (1801-1878), documentation of mental patients by Hugh Welch Diamond (1809-1886), and the study of facial expressions by Duchenne de Boulogne (1806-1875) who also started taking photographs of patients admitted to The Pitié-Salpêtrière Hospital.

After becoming clinical director of Pitié-Salpêtrière Hospital, Charcot created new units, including those for electrotherapy, microscopy and photography. The principal aim of the photographic unit was to document patients with epilepsy and hysteria, and resident photographers were needed in readiness to document attacks. Fortunately a new intern, Paul-Marie-Léon Regnard (1850-1927) was familiar with photography and played a central role in setting up the unit. Furthermore, its existence bore fruit in the form of a three-volume book, *Iconographie Photographique de la Salpêtrière* (1876-1877, 1878, 1879-1880), edited by Désiré-Magloire Bourneville (1840-1909) and Regnard. After their departure Londe filled the post of photographic engineer. Unlike Regnard, he was not a physician but an amateur photographer, and following Charcot tried to capture every relevant detail. To this end he improved facilities such as the photographic studio and dark room and invented a camera capable of taking sequential photos. He also wrote many books on photography. It is safe to say that Londe fulfilled Charcot's demand for accurate visualization of patients and greatly contributed to the launch of NI. Publication of the journal discontinued in 1918, a year after Londe's death – possibly a coincidence, but we believe his contribution was essential to its success.

III. Parkinson's disease in extension

When writing "Cover of the month" we reviewed two NI articles concerning four PD cases^{5, 7} three of which were described as PD in *extension*. The remaining case, concerning Léon, was also possibly of the *extension type*. Charcot also described Bachère's case as PD in *extension*.

Dejerine viewed the same condition as follows9:

A côté des attitudes dues à la contracture musculaire, je mentionnerai aussi celle des malades atteints de maladie de Parkinson. Au repos, ces malades se tiennent penchés en avant, raides, comme soudés, la tète relevée, les bras en flexion et collés au tronc (fig. 215 et 526) ; lorsqu'ils se déplacent, ils marchent courbés en avant (fig. 215, 214 et 325), d'un mouvement progressivement accéléré, semblant courir après leur centre de gravité ; ils sont toujours dans un équilibre instable, et se déplacent dans le sens de la moindre impulsion qu'on leur donne — anté- et rétro-pulsion, latéro-pulsion. D'autre fois, mais beaucoup plus rarement, le tronc et les membres, au lieu d'être en flexion, sont en extension — type d'extension de Charcot (fig. 527) ^{*6}.

*6: Fig. 4

(Alongside postures due to muscular contracture, I will also mention the posture of patients suffering from Parkinson's disease. When at rest, these patients lean forwards, stiffly and as if solidified, heads raised, arms flexed and clasped against the trunk (figs. 216 and 526); in motion, they walk bent forwards (figs. 214, 215 and 325) moving progressively faster as if they were chasing their own centers of gravity. They are always in a state of unstable equilibrium, and move in whatever direction they are pushed, however gently – antepulsion, retropulsion, lateropulsion. On other occasions, but much more rarely, instead of being flexed, the trunk and limbs are extended – Charcot *extension type*).

Both Dejerine and Gowers¹⁰ accepted Charcot's view of PD in *extension*. And it is easy to understand how both the facies and postural characteristics of PD as understood by its early observers should differ from our own viewpoint.

Fig. 4 is a photograph from Dejerine's textbook⁹ and shows a patient with PD in *extension*. This contrasts with another photograph, two pages before, of a typical parkinsonian, forward-flexed posture (Fig. 6). The difference is obvious. However, close examination of the face in Fig. 4., shows that it is Bachère.

Now we need to return to the descriptions of Charcot's PD in extension in Béchet's article⁷.

M. A. Beh 52 ans, avocat, ancien magistrat, a suivi pendant quelques semaines le traitement par le fauteuil trépidant, la Salpêtrière, . . .

Antécédents héréditaires, — Les renseignements que j'ai pu recueillir sur les antécédents héréditaires de M. Beh soit par luimême, soit par les personnes de sa famille qui l'accompagnaient sont fort peu précis; il paraîtrait qu'aucun de ses ascendants n'a eu de maladie nerveuse ; plusieurs auraient présenté des accidents rhumatismaux.

Antécédents personnels, — Il affirme n'avoir jamais fait de maladie sérieuse pendant sa jeunesse.

M. B... est continuellement en mouvement . . . ; si on le prie de s'arrêter pour l'observer dans la station debout, il ne peut y rester que quelques courts instants . . . L'aspect général est le même que pendant la marche, les membres inférieurs restent accolés Tun à Tautre, les cuisses et le tronc forment une ligne oblique dirigée en arrière jusqu'au niveau de la troisième vertèbre dorsale, à ce point, la colonne vertébrale se porte en avant, brusquement, ce qui accentue la voussure des épaules ; une ligne verticale abaissée de la troisième dorsale tomberait loin en arrière des talons^{*7}.

Le membre supérieur gauche est en extension complète le long du tronc, l'épaule gauche est un peu abaissée, Tavant-bras est étendu sur le bras et la main sur Tavant-bras;

[https://archive.org/stream/McGillLibrary-osl_bechet_1892-16352/osl_bechet_1892_djvu.txt]

*7: Fig. 7

(Obs. I – M. A. Beh., aged 52, a former magistrate, was treated for several weeks with the vibratory chair at La Salpêtrière. There is absolutely no evidence of antecedents in his family, and he claims always to have enjoyed good health.

Mr. Beh. never stops moving: if one asks him to stand still so that he can be observed in a standing position, he cannot hold it for more than a few moments, but his general appearance is the same as when he is walking. The lower limbs are held close together, the thighs and trunk form a single line sloping backwards up to the third up to the third dorsal vertebra; from that point on the spine leans sharply forwards, emphasizing the hunching of the shoulders. A vertical line drawn from that point would reach the heels well behind the heels).

According to a contemporary review¹⁶ (1899) this PD in *extension* had at least two subtypes: one with extended, rigid limbs (Bachère's type) and the other with flexed arms, tremor and extended neck and trunk. Charcot himself thought that the condition had two subtypes.

Whatever it was and however many subtypes it may have possessed, PD in *extension* is rarely talked about nowadays. And a PubMed search with the terms "*type extension*" in French and "*extension type*" in English returns no hits. Therefore, what "PD in *extension*" really meant by way of a diagnostic criterion and whether it meets any of the modern PD criteria is an unsolved mystery.

Concluding remarks

The year 2019 marks 202 years since Parkinson first used the term "shaking palsy." And in 2017 *Movement Disorders* celebrated the 200th anniversary by publishing in September a special issue: *Parkinson's Disease: Past, Present and Future*¹⁷. This issue, coauthored by Goetz and others, features excellent reviews on the past, present, and future of PD with a cover design that shows the title page of Parkinson's *An Essay on the Shaking Palsy* and a photograph of Léon (Fig. 8). As discussed above, Léon was one of three cases presented in the article titled "Note on some peculiar postures observed in Parkinson's disease", and was originally considered an

atypical case. In our opinion it was, based on the original terminology, a case of *extension type* PD.

In 2011, two of the authors of this paper (MK, MWM), while writing about Brodmann area 12^{18,19}, sounded the alarm over not using primary sources. And while it may seem unnecessary to current clinical practice to review historical data, we believe like Charcot, who went to the trouble of obtaining Parkinson's original monograph, that reviewing original data is essential to good practice.

Addendum

This review was compiled with revisions and additions from explanatory comments^{6,8} and a postscript²⁰ to "Cover of the month" photographs published in issues of *BRAIN and NERVE*.

Acknowledgements

We should like to thank Dr. Daisuke Yanase for his assistance in translating the original paper from Japanese to English, and Professor C.J.N. Mann for translating the original French texts into English. The research was supported by Grants-in-Aid for Scientific Research (also known as KAKENHI) for Innovative Areas (JP 25119006; JP 18H05525).

[Legends]

Fig. 1 Bachère's portrait drawn by Charcot

Note the signature "JMC" (Jean-Martin Charcot).

Reproduced from Blin MM, Charcot, Colin H: Vingtième Leçon. Un cas de maladie de Parkinson sans tremblement homme de 32 ans. Policlinique du Mardi 12 Juin 1888. Cours de M. Charcot. Leçons du Mardi à la Salpêtrière, Professeur Charcot. Policlinique 1887-1888.

Fig. 2 Bachère's portrait drawn by Richer

Dated 22 June 1888 (The 20th lecture of *The Tuesday Lessons* was delivered on 12 June 1888).

Reproduced from Richer P: Habitude extérieure et faciès dans la paralysie agitante. Nouv Iconogr Salpêtrière 1: 213-216, 1888.

Fig. 3 Bachère's photographic portrait in Dejerine's Sémiologie des Affections du Système Nerveux

Dejerine's footnote says: "A 36-year-old patient with Parkinson's disease-the same patient as in Fig. 327- with severe contracture of the frontalis. He experienced onset at age 20, one year after severe trauma. Photograph taken at The Bicêtre Hospital in 1893. For the patient's clinical conditions and drawings refer to Charcot's *The Tuesday Lessons* (1887-1888, t.1, p.437)".

Reproduced from Dejerine J: Sémiologie des Affections du Système Nerveux. Masson, Paris, 1914, p. 683.

Fig. 4 Posture of Parkinson's disease in extension presented by Dejerine

Note the resemblance to Léon's posture. The subject of this photograph is actually Bachère. Dejerine's footnote says: "A 36-year-old patient with Parkinson's disease – *extension type* – with onset at age 21 after severe trauma, who has marked propulsion. His facies is shown in Fig. 323 (Fig. 3. of our review) (Bicêtre 1893)".

Reproduced from Dejerine J: Sémiologie des Affections du Système Nerveux. Masson, Paris, 1914, p. 688.

Fig. 5 Léon

Reproduced from Béchet E: Note sur quelques attitudes rares observées dans la maladie de Parkinson. Nouv Iconogr Salpêtrière 5: 223-228, 1892.

Fig. 6 Typical posture of Parkinson's disease in Dejerine's Sémiologie des Affections du Système Nerveux.

Dejerine's footnote says: "Severely flexed posture of a 64-year-old patient with Parkinson's disease (Bicêtre 1891)".

Reproduced from Dejerine J: Sémiologie des Affections du Système Nerveux. Masson, Paris, 1914, p. 686.

Fig. 7 Richer's drawing of Parkinson's disease in extension

Based on the photograph of Case 1 in Béchet's article⁶ it shows abnormal posture, backwardleaning trunk and extended arms.

Reproduced from Béchet E: Note sur quelques attitudes rares observées dans la maladie de Parkinson. Nouv Iconogr Salpêtrière 5: 223-228, 1892.

Fig. 8 Cover of the September 2017 issue of Movement Disorders

This design consists of the title page of Parkinson's original paper and a photograph of Léon.

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