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Congenital Aglossia and the Report by Antoine de Jussieu: A Critical and Historical Review

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Abstract

Objective: Antoine de Jussieu, renowned French botanist and physician, laid the groundwork for future research on the topic of congenital aglossia when his study, "Observation About How a Girl Born Without the Tongue Was Able To Perform All Functions That Depend On That Organ," was presented to the 22-member assembly of the Royal Academy of Sciences of Paris on April 22, 1718. This study reviews the literature on de Jussieu's report.

Study design: The original report by de Jussieu was read and translated into English, after which 94 studies about congenital aglossia history and botany were selected.

Results: Of the 95 studies selected, one refers to de Jussieu's report (the groundwork), Annex I; 85 discuss congenital aglossia; five are about history and four about botany. Of the 85 directly associated with congenital aglossia, 36 do not mention de Jussieu, and 49 cite his observations differently.

Conclusion: Because of the 1718 report, de Jussieu became a major figure in the history of congenital aglossia and the author most often quoted in studies about it. However, there were historical facts that needed to be clarified. Some of the issues addressed in this review included the replacement of the letter *A* with *M* as the initial of de Jussieu's first name; the omission of the word *née* in the original title; the replacement of *née sans langue* with *privée de langue* in the title; the change of the girl's age from 15 to nine years; the suggestion that the patient had limb defects; the change of year from 1718 to 1717 and 1719; the incorrect spelling of de Jussieu's name and title of his report.

Keywords: Congenital aglossia; Historical review; Antoine de Jussieu

Introduction

Three factors convinced the authors of this manuscript of the importance of publishing a full and reliable English version of the original manuscript [1], originally written in French, which was extracted from the minutes of the assembly during which de Jussieu made his report. The first factor relates to the rich details contained in his observations, which likely make it the most complete medical report to this date on a case of congenital aglossia (CA), even though it was produced at a time when medicine did not rely on diagnostic tests. At that time, physicians had only their cognition and their senses- sight, hearing, smell and taste- to guide their diagnoses, and de Jussieu examined the patient only two times, once at night by candlelight, and once in daylight. The second factor has to do with the differences in the notation of the citations of de Jussieu in bibliographies, where the letter *A*, as in *Antoine*, was often replaced with *M*, as in *Monsieur* [2-20], which corresponds to Mr. in English, and which was found before the name *de Jussieu* on page 12 of the minutes that announced his lecture. This finding is confirmed by the fact that all the names of other assembly members were preceded by an M, when referring to only one member, or *MM*, the abbreviation of the plural form of *Monsieur*, to announce a group of lecturers. It is still standard for male names, and the abbreviation *Mme*, for *Madame*, before female names. Finally, the third factor can be found in the fact that in addition to errors in interpretation, made even by some respectable authors [2,4,14], other lapses have been found: (a) incorrect spellings, such as *De Jéssieu* [5], *Jusquieu* [21] and *Jessieu* [22]; (b) in-text citations but with no indication of source or inclusion of reference in a bibliography [21,23-33]; (c) omission of the initial letter of the author's first name and of the title of the report in bibliographies, as well as misspellings of the author's name that listed, for example, only *Jessieu*, or *De Jéssieu* (1718, Hist. De l'Acad. des Sciences, 612) [22,34-37]; (d) omission of the word *née* ("born") found in the original title [2-20]; (e) replacement of the words *née sans langue* ("born without the tongue"), found in the original title, with *privée de langue* ("deprived of the

tongue"), without mentioning the source or including a reference in a bibliography [25]; (f) inclusion of the information that the patient had limb defects [25]; (g) inclusion of another inaccurate reference to the fact that the girl had limb defects in a report made ten years later by another author [28] who did not cite his source; and (h) the use of the year 1719 instead of the year 1718 [2-4,6,10,11,14,16-20] ; and (i) incorrect citation dates that include 1717-1719 [3].

An intriguing finding was the omission of the word *née* ("born"), found in the original title. Almost all references, even those that correctly spell the name de Jussieu A, omit the French word *née* in the original title, which was only kept by three texts by two authors [38-40]. The word née (the past participle or adjective in the French language) used in the title has the same meaning as *congenital*. This detail is important because it identifies *de Jussieu* as the first author to have identified a case of CA. His intention was to establish a difference between his case and that of the boy from Saumur, who had lost his tongue due to gangrene resulting from smallpox.

The authors are unaware of any versions other than the original text by de Jussieu; however, Rosenthal [2] reported that the article was republished in several European periodicals, the last time as late as 1770. Meyer, in his article, "Ueber die angeborenen Fehler der Zunge und die dadurch bedingte Hinderung des Saugens" (In English, "On congenital anomalies of the tongue and the consequent difficulties of suction and swallowing") [23], stated that the Portuguese physician Dr. Somarive published a description of the same case in a special pamphlet.

The references made by most authors seem to have been based on secondary citations, and not on direct readings or analyses of the original text by de Jussieu, or the European publications mentioned by Rosenthal [2] and Meyer [23]. The omissions and misspellings of the author's first name and the title of his original report, the omission of the word *née* in the title, the use of the years 1717 and 1719 instead of 1718, the replacement *of née sans langue* ("born without the tongue") with *privée de langue* ("deprived of the tongue") and the presumption that the Portuguese girl had limb defects may have resulted from the errors found in those European publications. Meyer [23] includes a reference to de Jussieu in the body of his article and states that a Portuguese physician named Somarive had published a report of the same case in a special newsletter. However, he does not mention the title of that publication or any references that may facilitate the search for such an important source. He also does not clarify whether Dr. Somarive had examined the Portuguese patient described by de Jussieu, or only reproduced the report with additional comments. Moreover, Rosenthal [2] did not offer the bibliographic pathway to find those reissues in several European periodicals.

Please see the supplemental file for documentation of the original 1718 article and references to it.

Considerations about the de Jussieu Family

From 1686, when Antoine Laurent de Jussieu was born, to 1853, when Adrien-Henri de Jussieu died, France was the birthplace of five notable botanists, all from one of the branches of the large de Jussieu family [41,42]. These botanists contributed to the rise of botanical science, made important journeys in Europe and Latin America to study the flora and fauna of those still virgin regions, and contributed significantly to the enrichment of taxonomy. They were so highly regarded that the name *de Jussieu* is currently associated with the classification of several plant species, such as the *Simaruba jussiaei*, the quassia plant named after Antoine by Linnæus [41,42]. Further, the six Brazilian biomes- Amazon, Caatinga, Cerrado, Atlantic Forest, Pampa and Pantanal- are rich in plants of the *Sapindaceae Jussieu* and *Araceae Jussieu* families [43-46]. Bernard de Jussieu, the second in chronological order, was the first to classify whales as mammals [42] and Joseph de Jussieu, the youngest, participated as a botanist in the scientific expedition called *La Condomine*, which was sent to Peru and other South American countries by France to measure a meridian arc. After the investigations were completed, Joseph de Jussieu remained in South America practicing medicine.

France still honors the de Jussieu family today, and their name was given to a campus of Université Pierre-et-Marie-Currie, to streets and subway lines in Paris, Marseille, and Nice, and to the planetary asteroid # 9470 named after de Jussieu, as well as to several schools, hotels and other places.

The group discussed above was composed of three brothers (Antoine, Bernard and Joseph), a nephew (Antoine Laurent) and a son and grandnephew (Adrien-Henri, Laurent's son). Of the five relations, four studied medicine, but only Antoine, the oldest, and Joseph, the youngest, actually practiced it. Antoine dedicated special attention to the poor [41], which places him among the precursors of social medicine and contributes to the perpetuation of his name in the history of medicine. Moreover, evidence of his commitment to social medicine is found in the fact that his patient with CA was a very poor girl from a small village in Alentejo, a province in Portugal.

Absence in the Medical Literature

De Jussieu did not publish his observations because there was no medical literature at the time when he wrote them. The first attempt to edit a medical journal was made by Nicolas de Blegny [47,48], who was the editor of the journal "Nouvelles Découvertes sur Toutes les Parties de la Médicine (in English, "New Findings in All Areas of Medicine"), which was released monthly from 1679 to 1712. The same journal was edited also in Switzerland and translated into Latin under the title "Zodiacus Medico Gallicus," from 1680 to 1685. The second medical journal, also French, was edited by the priest J.P. de la Roque, under the title *Journal de Savants* ("Scientists' Journal"), and was published from 1681 to 1685. Finally, a third medical journal was edited by Claude Brynet, under the suggestive title of *Progress de la Médicine* ("Medical Advances"). The latter was the medical journal with the longest life, as it was published from 1695 to 1709. None of these journals were in publication when de Jussieu examined the case of CA.

Consideration about the report made by de Jussieu

The observations made by de Jussieu were organized according to the structural norms of present-day reports. He begins with a short introduction that justifies his intention to report the case for the scientific community and continues with information about the patient, including her age, origin and social status, as well as how he gained access to her. He then reports on the detailed oral examination, and describes a protuberance rising 4 mm on the floor of the mouth, and concludes that the protuberance is part of the tongue. He also evaluates the way the girl, a 15-year-old Portuguese adolescent, performs the five functions of the tongue (mastication, taste, deglutition, articulation, spit), and notes how fundamental the genioglossus, mylohyoid and geniohyoid muscles are for her speech performance. He describes the tongue functions one by one, and makes considerations about the areas of taste perception, physiology,

nutrition, speech, and hearing, and provides a detailed description of her pronunciation of some phonemes, as well as the resources that she used to overcome difficulties. He clarifies the difference between swallowing solids and liquids, and the use of her fingers to help the distribution of solids during mastication. This description is one of the few detailing such use of the fingers. He provides a detailed explanation of how she spit. He makes comments about the strategy that the mother adopted to feed the child, which consisted introducing the nipple into the infant's mouth and pressing her breast to make the milk flow into the infant's pharynx. He also describes the details of the girl's mandibular dentition, reporting that her teeth were not distributed in a double row and were not inclined lingually, as those of the boy in the report made by the Saumur surgeon, and classifies such distribution as a notable fact.

The enlarged uvula is referred to as displaying hypertrophy, which allowed it to function in a vicarious role that reduced the opening of the throat and modulated the sounds produced through the nose. De Jussieu continues with a detailed description of all organs that compose what today are known as the speech organs. He mentions the successful experiments conducted by Ambroise Paré, who used an instrument in the form of a gutter to help patients without the tongue to communicate better. Finally, he concludes with a discussion and makes the surprisingly up-to-date conclusion that "the parts that form the mouth are so necessary for speaking that they may compensate for the absence of the tongue" [1]. This conclusion is consistent with current knowledge and has been discussed by Twisleton [49] in his book, *The Tongue Not Essential to Speech*, based on reports that African confessors were able to speak after their tongues had been amputated.

Review of the Literature

The review of the literature revealed surprising and intriguing facts. Of the 95 studies selected, 85 discussed CA; five were about history and four about botany. The first one, obviously, was the original of de Jussieu's report. Of the 85 directly associated with CA, 36 did not mention de Jussieu [50-85], and 49 mentioned him in different ways (Supplementary file- Figure 1) [2-40,86-95]. Of the 49 that mention him, 27 included his work in a list of references, [2-20,34-40], ten referred to other authors that quoted him [86-95], and 12 mentioned him only in the body of the text [21,23-33]. Of the 27 that included his work in the list of references, 19 used the letter *M* as the initial of his first name [2-20], 6 correctly used the letter *A* (texts 37, 38 and 40 were published by the same author) [35-39,49] and two did not mention either *A* or *M* in the title [22,34].

Twenty-two authors omitted the word *née* ("born") in the original title [2-20,35-37]. Only three texts [38-40] kept the original title of the document provided by the *Royal Académie de Sciences de France*, as found in the hand-written text in French, the focus of this study. In fact, these texts were produced by only two authors, as the texts [39,40] were published by the same author.

Of the 10 studies that mentioned de Jussieu in the text only, without information about the source of that citation, two stated that the patient had limb defects [25,28]. One of them [25] replaced the terms *née sans langue* ("born without the tongue"), found in the original title, with *privée de langue* ("deprived of the tongue"). The word *née* (past participle or adjective in the French language) used in the title has the same meaning as *congenital*. This is important because it identifies de Jussieu as the first author to have identified a case of CA. His intention was to establish a difference between his case and that of the boy from Saumur, who had lost his tongue due to gangrene resulting from smallpox.

Other intriguing oddities were found, such as misspellings of his name in texts that used *De Jéssieu*, [5] *Jusquieu* [21] or *Jessieu* [22], instead of *de Jussieu*, mistakes in the title [3,15,19], and listings without his first name and title, as in Jussieu, Hist. de l'acad. des Sciences – 612 [22,34], as well as changes in the girl's age, from 15 to 9 years [34,88] , and inclusion of the years 1717 [3] and 1719 [2,4,6,10,11,14,16-20] as the date of the original report (Supplementary file- Annex II).

Discussion

Numerous discoveries and discussions have taken place in the 287 years that separate de Jussieu from our time, always in the search of explanations for CA and its association with other anomalies. Although the actual etiology has not been clarified, the action of drugs during pregnancy, gestational hyperthermia, and genetic factors have been pointed as possible causes, but conclusions are all still speculative.

In the last three centuries, some authors, such as Rosenthal [2], Fulford [87], Gardiner [34], Hall [4], and Gorlin [14] have attempted to determine the occurrence of cases, define causes and stablish a coherent classification. Despite all their efforts, toward cohesion, paradigms of classification still need improvement. Although the classification system devised by Hall seems to be coherent, it has not settled the question of how to classify CA. Our literature review also revealed that some surprising historical facts should be clarified, and some issues addressed: the replacement of the letter *A* with *M* as the initial of the French physician's first name; the omission of the word née in the original title; the replacement of *née sans langue* with *privée de langue* in the title; the suggestion that the patient examined by de Jussieu had limb defects; and the incorrect spelling of his name and title. Details found in texts all suggest that the source or sources of information of the authors reviewed might have been texts other than the original manuscript by de Jussieu; perhaps those found in the European publications of the 1770s mentioned by Rosenthal [2]. To answer these questions, such publications should be found and studied, particularly the newsletter published by the Portuguese physician named Somarive mentioned by Meyer [23]. Did Dr. Somarive see the patient examined by de Jussieu before or after the French physician made a report of the case? Did the patient described by de Jussieu actually have limb defects, as reported by Grislain and Deffez [25,28]? Did de Jussieu overlook (unlikely) such abnormalities, whereas Dr. Somarive noted them and later described them, after de Jussieu had made his report? Is the newsletter published by Dr. Somarive, together with the publications by his contemporary authors, the source reviewed by those that have misrepresented so many facts in their reports? The authors of this study expect to answer these and other questions in the future, as studies already underway disclose new findings about the publications by Somarive and the other authors quoted by Rosenthal. These findings will be the topic of a future report.

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