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## Structural brain changes in migraine and cluster headache

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## **An early 18<sup>th</sup> century case description of cluster headache**

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## Abstract

We present a previously unreported early 18th-century description of cluster headache by the English antiquary Abraham de la Pryme (1671–1704) initially attributed to hydrophobia (rabies). We will also give a short overview of other descriptions of cluster and cluster-like headache in historical literature.

## Case description

In 1702, Abraham de la Pryme published on “A Remarkable Case of the Hydrophobia” in the Royal Society’s *Philosophical Transactions*.<sup>1</sup> The report is about his brother’s dog and her whelps which had all succumbed to rabies in 1695. One of his brother’s servants, an apprentice of about 14 years of age, was seized with headache shortly after caring for the dogs, together with other servants. He developed a fever and characteristic features of rabies, including and behavioural alterations before ultimately passing away. De la Pryme also mentions another servant who was afflicted by headache shortly after the death of the whelps:

“They being thus dead, were soon forgot, until that about 3 Weeks after, my Brother’s Servant, a most strong laborious Man, that had frequently put his Fingers into the Whelp’s Mouth, began to be troubled now and then with an exceeding acute Pain in the Head, sometimes once, sometimes twice a Day, so very vehement that he was forc’d to hold his Head with both his Hands, to hinder it from riving in two, which Fits commonly held him about an Hour at a time; in which his Throat would contract, as he said, and his Pulse tremble, and his Eyes behold every thing of a fiery red Colour. Thus was he tormented for a whole Week together, but being of a strong Constitution, and returning to his Labour in every Interval, he sweat and wrought it off, without any Physic.”

Abraham de la Pryme, an English antiquary born January 15, 1671 in Hatfield, South Yorkshire, and of Flemish and French Huguenot ancestry, kept a diary from the age of 12 until his death in 1704, *Ephemeris vitae, or a diary of my own life, containing an account likewise, of the most observa-*

*ble and remarkable things that I have taken notice of from my youth up hitherto.* He studied at St. John's College in Cambridge from 1690 and in addition to the usual classical and philosophical studies, he concentrated on natural history, chemistry and magic until he received his degree of Bachelor of Arts in 1694.<sup>2</sup> At Cambridge, he was a contemporary of Sir Isaac Newton, whom he described as “a very learned man” and as “an excellent mathematician, philosopher, divine, etc.” He was the author of the first history of the city of Hull and several scientific papers of his were published in the Royal Society's *Philosophical Transactions*. This ultimately led to his election as fellow of the Royal Society. His diary was published in 1870.<sup>3</sup>

De la Pryme ascribed the servants' disease symptoms to rabies because of the close relationship they had had to the dogs. Moreover, as de la Pryme wrote about his experiences only seven years later, recall bias may have contributed as well. Rabies can indeed cause a wide range of neurological symptoms, including quite frequently headache. Rabies-associated headache tends to be more of a continuous character and often precedes the acute neurological syndrome.<sup>4</sup> De la Pryme notes that the first time the second-mentioned servant had his headache attacks was about three weeks after the death of the whelps. The incubation period of rabies most commonly is between 20 and 60 days with a range from 10 days to a year.<sup>4</sup> He did not mention other specific symptoms than recurrent headache attacks. Intermittent “contraction of the throat” during the headache attacks is unlikely to be due to hydrophobia. De la Pryme stressed that the servant was cured spontaneously without receiving any medication. This makes it most unlikely that the man had suffered from rabies and that the headache attacks had been a consequence of this disease. Even to date, rabies remains a fatal disease in many once clinical symptoms have become manifest or is associated with long-lasting neurological features.<sup>5-7</sup> The chances of a man surviving rabies in the late 17<sup>th</sup> century, without any lingering symptoms, are therefore negligible.

De la Pryme's attribution of the headache to rabies can still be considered as progressive, however. In the 17<sup>th</sup> century, superstition was still manifest in a large part of the population, with de la Pryme being a prime example, especially at younger age. It is therefore remarkable that he thought that an infectious disease caused the “devilish headache” he had observed.

The pattern of the headache attacks, once or twice a day and their duration of an hour without lasting symptoms are suggestive of cluster headache. Although a cluster period of one week is relatively short, episodic cluster headache bouts can, at the lower end of the scale, last for periods as short as a week;<sup>8</sup> even minibouts (bouts shorter than of one week) have been described.<sup>9</sup>

Unfortunately, de la Pryme, who had hardly any medical training, did not mention the localization of the headaches, and used non-medical prosaic and metaphoric descriptions. “The riving in two” might be interpreted as hemicrania, although it could also indicate the severity of the pain. The passage “his Eyes behold every thing of a fiery red Colour” most likely points to de la Pryme’s observation of reddening of the eyes (a probable accompanying cranial autonomic symptom), but theoretically could also describe the very rare symptom of erythropsia (abnormality of vision in which all objects appear in a red tint). Clear (other) secondary symptoms, such as lacrimation, ptosis, miosis and rhinorrhea, remain unnoticed, unreported or absent. Despite the lack of a clear description of secondary symptoms, we believe De la Pryme witnessed a one-week long episode of cluster headache and that this is the oldest description of cluster headache in English literature.

## Discussion

Cluster headache and paroxysmal hemicrania were conceptualized in the 20th century as separate entities. Bing, Harris and Horton<sup>10-14</sup> contributed to the concept of cluster headache in the first half of that century, while the term *paroxysmal hemicrania* was introduced by Sjaastad and Dale only a few decades ago.<sup>15,16</sup> Together with the very infrequent short-lasting unilateral neuralgiform headache with conjunctival injection and tearing (SUNCT), these three headache types are often collectively referred to as trigeminal autonomic cephalalgias.

Historical reports of possible trigeminal autonomic cephalalgias are, probably due to the rarity of these disorders, scarce. Descriptions suggestive of cluster headache date back to the 17<sup>th</sup> century, when the Dutch physician Nicolaes Tulp (the main physician seen in Rembrandt’s paint “The Anatomy Lesson”) published his *Observationes medicae* in 1641. He described the headache history of an Amsterdam merchant of Flemish origin, Isaack van Halmale (1577/1578–1641), who, in the beginning of the summer, “was afflicted with a very severe headache, occurring and disappear-

ing daily on fixed hours”, with headache attacks rarely lasting longer than two hours. Although the details are insufficient to meet today’s criteria for episodic cluster headache of the International Headache Society (IHS),<sup>17</sup> cluster headache is a highly probable diagnosis.<sup>18</sup>

The only other known 17<sup>th</sup> century description of a possible trigeminal autonomic cephalalgia was published by the English doctor and founding member of the Royal Society Thomas Willis. He was one of the first to distinguish continuous from intermittent headaches and described in his *De anima brutorum* (1672) “a venerable matron” who was afflicted with severe recurrent daily headaches, occurring every afternoon around 4 o’clock, for about five weeks.<sup>19</sup>

Whereas Tulp’s observation is currently regarded as the earliest case of cluster headache, the first description of a patient meeting today’s IHS criteria for episodic cluster headache is by the Dutch physician Gerard van Swieten, court physician of Mary Therese of Austria.<sup>20</sup> He discussed in his *Commentaria in Hermannii Boerhaave aphorismos de cognoscendis et curandis morbis* (1745) the medical cases of his teacher, the famous professor Herman Boerhaave at Leiden University. He described a middle aged man, who suffered from a peculiar left-sided headache that occurred every day at the same hour. This headache lasted a few hours and was accompanied by reddening and tearing of the left eye.

Besides these three cases, a dozen of general descriptions and case histories in both medical and non-medical literature, like diaries, have been reported in the past decades that are suggestive of periodical, hemicranial headaches that could be diagnosed as cluster headache or paroxysmal hemicrania. A 35-year old German female patient in Johann Christoph Ulrich Oppermann’s *Dissertatio medica inauguralis de hemicrania horologica* (1747) suffered from excruciating daily headache attacks that lasted fifteen minutes. This case of clockwise headache (“hemicrania horologica”) is currently regarded as the earliest account of (chronic) paroxysmal hemicrania by some,<sup>21</sup> but not by all.<sup>22,23</sup> Additional possible pre-20<sup>th</sup>-century cases and descriptions of cyclical headaches suggestive of cluster headache were published, among others, by the Italian professor Giovanni Battista Morgagni (1761),<sup>24</sup> the English physicians Robert Whytt (1764) and Marshall Hall (1836),<sup>24</sup> the German neurologists Moritz Heinrich Romberg (1840),<sup>25</sup> and Albert Eulen-

berg (1871),<sup>26</sup> and self-reported by the German physician Johann Valentin Müller (1813)<sup>27</sup> and the English vicar Robert Francis Kilvert (1840-1879).<sup>28</sup>

Tulp and Van Swieten both originally published their case histories in Latin, the language which was most common in medical literature until the 19th century; Tulp translated his *Observationes medicae* into Dutch as soon he discovered it had been translated inadequately by others. Willis, the first Englishman publishing about periodical headaches, wrote in Latin as well. In English medical literature, the oldest description of a hemicranial headache suggestive of cluster headache so far was published by the Edinburgh professor Robert Whytt, who, like Van Swieten, was a pupil of Boerhaave's. He wrote about periodical headaches in his manuscript *On the nature, causes, and cure of those disorders which are commonly called nervous, hypochondriac or hysteric* published in 1764.<sup>24</sup>

It probably is a matter of time until other cases of cluster headache, perhaps even older than those described by Tulp, Willis and de la Pryme, will be found in historical medical or non-medical literature. The overwhelming nature of disorders now known as trigeminal autonomic cephalalgias may have inspired more historical authors to describe headache patients suffering from these disorders. New innovations like Google Book Search, a service of Google that allows searching the full text of old digitized books, may help us in this search. It has already been suggested that, as the use of Latin in medicine became obsolete after the 1820s, the limited knowledge of classic Latin and Greek languages possessed by today's doctors hinders a systematic search of old medical texts, which is needed for recovering historical descriptions of headache syndromes.<sup>20</sup> However, the case by de la Pryme underlines, that not only the medical observations of historical physicians should be critically reviewed for descriptions of headache, but that other texts and diaries, even in currently used languages, might still provide us with information on the existence and nature of trigeminal autonomic cephalalgias in earlier centuries.

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