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Plica neuropathica – unraveling the mystery of *Plica polonica*

Plica neuropathica – odkrywanie tajemnicy kołtuna polskiego (*plica polonica*)

Summary

Background: Plica as a disease entity appeared first in the 13th century. Its aetiology is still unknown and not fully understood. Being, in fact, an irreversible condition of tangling and felting the hair, its origin was interpreted in various ways, from religious ones, through lack of hygiene to mental illness. Although *p. polonica* originates from Eastern and Central Europe, and *p. neuropathica*, typical of Indian people, was first described in England, many characteristics connect these two conditions. Aim and Objectives: The study aimed to review the plica species over the centuries and to compare *p. neuropathica* and *p. polonica*. Materials and Methods: PubMed and Google Scholar databases were searched for medical articles, books and case studies on plica published up 1884 to 2020. We conducted a systematic and critical review of the literature to compare *p. polonica* and *p. neuropathica*. Results: Plica polonica and Plica neuropathica are the same disease entity. Conclusion: The variety of names and interpretations of the disease has made plica a condition still to be fully explained.

Keywords: *plica neuropathica*, *plica polonica*, Polish plait, medicine in the 19th c., dermatology in the 20th c., psychiatry in the 20th c., India, Europe

Słowa kluczowe: *plica neuropathica*, *plica polonica*, kołtun polski, medycyna XIX w., dermatologia XX w., psychiatria XX w., Indie, Europa

Introduction

Plica neuropathica is a term that has appeared in medical publishing since the 19th century.¹ The older name, occurring since the Middle Ages, is *plica polonica*. This disease aroused the interest of royal doctors, charlatans, and society. The term *p. neuropathica* has not emerged until 1884. *P. polonica* and *p. neuropathica* is an irreversible condition of tangled hair. Hair forms into a dense, rigid mass, which tends to mat and felt consistent. This is accompanied by dirt, tallow and often unpleasant smell. The reasons for this condition are found in the lack of hygiene, mechanical damage to the hair, parasitic infections of the scalp and hair, and the use of inappropriate cleaning products.²

For centuries, *plica* common among representatives of some populations was a symbol of body and spirit disease, and an expression of superstition and religious beliefs. It was most widely observed in Central and Eastern Europe, especially in rural areas.³ It was attributed to the Slav people as an endemic disease. Over the centuries, its clinical picture has changed. Initially, it was considered an infectious and congenital disease, associated with possession, or constituting a counterweight to other conditions.⁴ It was often accompanied by other somatic symptoms such as bone and joint pain, dizziness and headache. Moreover, its presence was associated with changes in the emotional and psychological state of people to which it occurred.⁵ Also, *plica* was often linked with witchcraft and quack activities.⁶

Over the centuries, it has gained many names, depending on the place of occurrence, associations it evoked and the attributes that

¹ J.J. Pruszyński, J. Putz, D. Cianciara, *Plica neuropathica – a short history and description of a particular case*, “Hygeia Public Health” 2013, vol. 48, pp. 481–485.

² S. Gupta, R. Kumar, A. Vijay, S.K. Jain, *Plica Polonica in a Patient on Chemotherapy: A Case Report with Review of Literature*, “International Journal of Trichology” 2017, vol. 9, pp. 124–126; A. Ohry, N. Ohana, *On Plica Polonica and the forgotten Joseph Romain Louis Kerckhoffs (1789–1867)*, “Progress in Health Sciences” 2018, vol. 8, n. 1, pp. 208–212; M. Marczevska, *Kiedy choroba była gościem – o językowym obrazie kotłuna w przekazach ludowych*, [in:] P. Zbróga (ed.), *Współczesna polszczyzna w badaniach językoznawczych. Od języka w działaniu do leksyki*, Kielce 2011, pp. 87–108.

³ J.J. Pruszyński, J. Putz, D. Cianciara, op.cit.; S. Anisha, K. Sukhlot, G.K. Sunil, P. Sandeep, *Bird's Nest View from a Dermatologist's Eye*, “International Journal of Trichology” 2016, vol. 8, n. 1, pp. 1–4.

⁴ E. Sakalauskaitė-Juodeikienė, D. Jatuzis, S. Kaubrys, *Plica polonica: From national plague to death of the disease in the nineteenth-century*, “Indian Journal of Dermatology” 2018, vol. 84, pp. 1–5.

⁵ A. Ohry, N. Ohana, op.cit.

⁶ M. Marczevska, op.cit.

were assigned to it. Currently, p. neuropathica is a rare disease, and isolated cases are observed among the population of India, especially in social groups with low material status. Although plica has been detected for centuries, it still is an unfathomable disease, difficult to identify and explain.⁷

History of *Plica neuropathica*

The disease has been already described thoroughly by often forgotten in medical textbooks Joseph Romain Louis Kerckhoffs (1789–1867).⁸ However, it was named ‘p. neuropathica’ only in 1884 by a physician from Manchester J.F. Le Page.⁹ He described the condition as a neuropathic disease in which scalp, as well as hair, are ‘structurally healthy’ and plication is caused by ‘structural changes produced by force within’. As a result, the hair arrangement is not messy but symmetrical. Two years before the publication, Le Page examined a 17-year-old patient. She presented with headache, general numbness and sensation of pins and needles over her scalp. To relief the pain she had washed and combed her hair but after that, she ‘felt a tearing sensation on the right side of her head’, and her hair was being drawn up, as a result, making a hard lump. Physical examination revealed that hair on the left side of her scalp was not drawn up or tangled. As patient also manifested symptoms of hysteria, Le Page claimed that ‘high nervous tension found vent in the hair itself’ causing plication.

Throughout the years, new cases have been described. In 1969, the first case of p. neuropathica in dark-skinned female was reported.¹⁰ A 46-year-old woman with another sudden onset of matted hair after washing them was presented. The previous episode resulted in cutting her hair short. The scalp itself appeared healthy, but the hair was divided into sections and tangled. The patient was under the care of psychiatrist due to her depression and anxiety. Blood test results were within the norm; there was no evidence of alopecia or dermatophytosis. Further analysis showed that her condition might have

⁷ E. Elghblawi, *Case Report: Acute Hair Matting in a Libyan Girl from the Outskirts*, “World Family Medicine Journal/Middle East Journal of Family Medicine” 2016, vol. 14, n. 9, pp. 21–23.

⁸ A. Ohry, N Ohana, op.cit.

⁹ J.F. La Page, *On Neuropathic Plica*, “British Medical Journal” 1884 (Jan 26), vol. 1, n. 1204, p. 160.

¹⁰ M.H. Simpson, J.F. Mullins, *Plica neuropathica*, “Archives of Dermatology” 1969, vol. 100, n. 4, pp. 457–458.

been caused by the unconscious habit of ‘vigorously rubbing together strands of kinky hair’ while sleeping.

In the case of a 60-year-old woman with moderate hypertension reported in 1992,¹¹ tangling of hair has progressively developed due to massages with a herbal mixture with mustard oil. There was no history of use of any bleach, detergent shampoo, dyes etc.; also, no abnormalities in her blood chemistry, detrimental for dermatophytosis and without any signs of infection on the scalp were detected.

In 2004, a case of a 14-year-old Sikh boy with multiple entangled, twisted hair-weaves after shampooing, was described.¹² Medical history revealed only traction alopecia over the frontal scalp. However, in his case, the psychological evaluation did not indicate any behavioral disturbances.

Although in many cases entanglement is associated with shampooing or poor hair hygiene,¹³ in 2006, a 14-year-old girl developed the disease in the absence of shampooing. This case, as opposed to many regular cases of *p. neuropathica*, shows that matting of scalp hair in this condition can be linked to underlying illnesses as this patient suffered from chronic illness and acute infection.

The first case of the disease among infants was reported in 2014. It was an 11-month-old Hindu boy with multiple, solid, irregular masses of matted hair.¹⁴ Aside from that, there were no other changes on the scalp. Because of religious beliefs his hair was only washed with water without soap, never cut or combed, and regularly oiled.

New cases of *p. neuropathica* continue to occur to this very day. In 2017, in Italy,¹⁵ a case of an autistic patient with matting hair was communicated. In turn, in 2019 in Portugal,¹⁶ a case of a 55-year-old Caucasian woman with diffuse scalp dermatosis was reported. There was much negligence in hair-care because of a belief that it would have caused pain. The patient scalp had many alterations. Finally, the woman was diagnosed with *p. neuropathica* with alopecia.

¹¹ N.K. Bansal, C.M. Kuldeep, *Plica neuropathica*, “Indian Journal of Dermatology, Venereology and Leprology” 1992, vol. 58, pp. 131–132.

¹² S. Dogra, A.J. Kanwar, *Plica Neuropathica: A Mystery*, “Pediatric Dermatology” 2004, vol. 4, pp. 477–478.

¹³ J. Kwinter, M. Weinstein, *Plica neuropathica: novel presentation of a rare disease*, “Clinical and Experimental Dermatology” 2006, vol. 6, pp. 790–792.

¹⁴ S. Verma, B.K. Thakur, *Plica “Neuropathica” in an 11-Month-Old Boy*, “Pediatric Dermatology” 2014, vol. 6, pp. 116–117.

¹⁵ M. Siragusa, S. Giusto, R. Ferri, A. Centofanti, C. Schepis, *Plica neuropathica (matting hair) in an Autistic Patient*, “The Journal of Dermatology” 2017, vol. 9, pp. 212–213.

¹⁶ A.F. Monteiro, A. Lencastre, *Whole-Scalp Cobblestone Plica Neuropathica with Alopecia*, “Skin Appendage Disorders” 2019, vol. 5, pp. 390–392.

Confusion with *Plica neuropathica*

Although numerous medical cases mention *p. neuropathica*, the disease is still considered to be vague. There are no particular clinical and dermoscopic features that could help doctors to identify it. There are also no medical recommendations and golden standards that provide diagnostic solutions. What is more, pathophysiology of this condition has been still unraveled. Still some medical cases pay attention to common factors that might contribute to the process of matting of the hair.¹⁷

Gupta and Kumar described cases of two women, whose hair started to tangle after they have had chemotherapy.¹⁸ Also, Joshi, in his article, presented patient treated with azathioprine, and found out that after treatment, her hair got matted and formed a stiff mass.¹⁹ Suresh, in 2001 and 2012 pointed out the disorder happened to woman, who had been suffering from anxiety.²⁰ Elghblawi noticed the disease in a 16-years old girl from war-torn Libya.²¹ Moreover, there are pieces of evidence that *p. neuropathica* is caused by using ionic or herbal shampoos and conditioners.²²

All these cases are connected since the disease is more prevalent among women. In the past, it was also common that women suffered from *p. neuropathica* more often than men, probably because of hair's length that predisposes them to tangle and matt.²³ Sethi, Kaur, Gupta and Puri in their article published in 2016 tried to explain how risk factors affect the origin of *p. neuropathica*. They mentioned that chemotherapy and immunosuppressive drugs might change the life cycle of hair, causing either no hair-producing or that hair shaft is narrow and defective, what is connected with the process of matting.

¹⁷ S. Anisha, K. Sukhjot, G.K. Sunil, P. Sandeep, op.cit.

¹⁸ S. Gupta, R. Kumar, A. Vijay, S.K. Jain, op.cit.; S. Kumar, B.K. Brar, P. Kapoor, *Drug-associated Plica Polonica: An unusual presentation*, "International Journal of Trichology" 2019, vol. 11, n. 2, pp. 80–81.

¹⁹ R. Joshi, S. Singh, *Plica Neuropathica (Plica polonica) Following Azathioprine-induced Pancytopenia*, "International Journal of Trichology" 2010, vol. 2, n. 2, pp. 110–112.

²⁰ P.N. Kumar, B. Antony, A. Chakravarthy, A.M. Koyamu, *Plica neuropathica (polonica) in schizophrenia – a case report and review of literature*, "Indian Journal of Psychiatry" 2001, vol. 43, n. 3, pp. 281–283; P.N. Kumar, V. Rajmohan, *Plica neuropathica (polonica) in schizophrenia*, "Indian Journal of Psychiatry" 2012, vol. 54, n. 3, pp. 288–289.

²¹ E. Elghblawi, op.cit.

²² S. Anisha, K. Sukhjot, G.K. Sunil, P. Sandeep, op.cit.; J. Kwinter, M. Weinstein, op.cit.

²³ S. Gupta, R. Kumar, A. Vijay, S.K. Jain, op.cit.

Interestingly, most of the described cases originate from India.²⁴ Precisely, plica is common among Hindu ascetics (Sadhus), where proper hair hygiene is lacking. Also, the disease occurs more often within populations living in poor sanitary conditions, which cannot afford adequate scalp hygiene.²⁵

There also begs the question, what causes disease among psychiatric patients. Although Sethi mentioned anxiety as a risk factor, yet he did not explain it. The most probable answer is emerging from Kumar's paper published in 2019. The author stated that maybe it is because patients perform maniac manipulations with their hair.²⁶

The greatest unknown is the past of p. neuropathica; the first documented case came from 1884 when J.F. Le Page described the disease in a 17-year-old, mentally-ill woman. There is no earlier publishing mentioning the condition directly, but in more recent articles, some authors compare it to dreadlocks or more often to p. polonica. They either use terms p. polonica and p. neuropathica as synonyms or to point out some differences, finding out they were two separate diseases. Even Le Page noticed that p. polonica and p. neuropathica are different disorders. But due to some obvious similarities between them, it should be properly analyzed with all new medical cases and decide whether they are different or the same disease.²⁷

What about *Plica polonica*?

Plica polonica, often referred to as 'kołtun', was a rare condition associated with irreversible twisting, entanglement and matting of the hair. It was characterized by a hard impermeable mass of keratin and felting of the outer layer of hair. This medical condition was considered to be characteristic for Poland, Lithuania, and neighbouring countries.²⁸

²⁴ S. Anisha, K. Sukhjot, G.K. Sunil, P. Sandeep, op.cit.

²⁵ S. Gupta, R. Kumar, A. Vijay, S.K. Jain, op.cit.; E. Elghblawi, op.cit.; A.F. Monteiro, A. Lencastre, op.cit.; S. Kumar, B.K. Brar, P. Kapoor, op.cit.; R. Joshi, S. Singh, op.cit.; P.N. Kumar, B. Antony, A. Chakravarthy, A.M. Koyamu, op.cit.; P.N. Kumar, V. Rajmohan, op.cit.; L. Gupta, M. Balai, A.K. Khar, A. Mittal, *Plica neuropathica*, "Indian Dermatology Online J." 2015, vol. 6, n. 4, pp. 310-311.

²⁶ S. Anisha, K. Sukhjot, G.K. Sunil, P. Sandeep, op.cit.; S. Kumar, B.K. Brar, P. Kapoor, op.cit.

²⁷ S. Gupta, R. Kumar, A. Vijay, S.K. Jain, op.cit.; S. Anisha, K. Sukhjot, G.K. Sunil, P. Sandeep, op.cit.; E. Elghblawi, op.cit.; S. Kumar, B.K. Brar, P. Kapoor, op.cit.; R. Joshi, S. Singh, op.cit.; P.N. Kumar, B. Antony, A. Chakravarthy, A.M. Koyamu, op.cit.; P.N. Kumar, V. Rajmohan, op.cit.

²⁸ A. Ohry, N. Ohana, op.cit.

The origin of the disease is associated with the Mongolian invasion of Poland in 1287. They were supposed to bring the pile to the country, where it has remained for many years.²⁹

In Europe, there were many historical beliefs about the cause and symptoms of the disease. Usually, it was associated with the dirt, lack of hygiene and the negligence of the hair.³⁰ It was also believed that the cause of it might be related to some supernatural powers, which punish people for their misdeeds.³¹ Mental illness, anxiety or the usage of herbal shampoo would also predispose a patient to the development of plica.³²



Fig. 1 The longest preserved p. polonica. The Faculty of Medicine Museum of the Jagiellonian University. Source: public domain.

Due to the symptoms, p. polonica is not the only term, which defined the afore-mentioned phenomenon. Because of the severe and sharp headaches, from which the patients were suffering, people called it also ‘bolączka’ or ‘gwoździec’ (the pain was supposed to be as severe as when a spine or nail is perforating the skull). The occurrence of the illness was supposed to be connected with the evil spirits, from where originated the term ‘wieszczyce’.³³ Plica was also commonly referred to as ‘gościec’, which meant rheumatism. Polish plait was supposed to be one of the symptoms of rheumatic disease, which

²⁹ Ibidem; J. Chevallier, *Histoire d'une maladie qui n'a jamais existé: la plique polonaise!*, “Histoire des sciences médicales“ 2018, An. 2, pp. 233–242.

³⁰ M. Marczevska, op.cit.

³¹ E. Sakalauskaitė-Juodeikienė, D. Jatuzis, S. Kaubrys, op.cit.

³² S. Gupta, R. Kumar, A. Vijay, S.K. Jain, op.cit.

³³ J.J. Pruszyński, J. Putz, D. Cianciara, op.cit.

could manifest itself by causing pain in muscles and bones. Previously mentioned ‘gość’ [Eng. guest] was considered to be the enemy of the human, which steals into his body and in the end reveals its existence by causing a plait.³⁴

Although literature mentions that the 13th century was the period when the Polish plait first appeared, it was not until the 16th and 17th centuries when it began to be described regularly by physicians. In 1584, Johannes Schenck von Grafenberg referred to that ‘phenomenon’, and a few years later, in 1599, Laurentius Staringelius described the condition in detail. In the 17th century, in numerous European cities like Paris, Basel, Venice, Hamburg, Leipzig, etc., new dissertations relating to p. polonica were frequently published.³⁵

The first clinical description of the disease appeared in the 18th century; the author of the publication was a lecturer of anatomy at the Vilnius University Stephanus Bisius. He stated that the Polish plait was not an illness, but a ‘human mistake’ resulting from negligence and numerous superstitions. At the beginning of the 19th century, Joseph Frank, a German physician in Vilnius, also presented his explanation. He described the Polish plait as a ‘national plague’, an infectious disease transmitted from generation to generation, whereas Ludvicus Knothe in his inaugural dissertation on p. polonica (1830) approached the subject differently, as he considered many factors influencing the deformation. For that reason, he proposed a specific treatment for the disease. Namely, he ordered to look for the cause of the condition, and only then to take care of the plait. The fully grown plait could be cut down without harming health.³⁶ In the 19th century, there also appeared numerous explanations of p. polonica provided by French and Polish scholars.³⁷

P. polonica as the name of the condition suggests that it was endemic among the Polish population. Yet, historical sources recall cases of the plait in neighbouring countries, especially in Lithuania.³⁸ It cannot be ignored, that plica was also reported by Jean-Louis Alibert (1768–1837) who described a 38-year-old French woman from Lille.³⁹

³⁴ Ibidem; M. Marczevska, op.cit.; H. Widacka, *Plica polonica czyli kołtun polski*, Silva Rerum, https://www.wilanow-palac.pl/plica_polonica_czyli_koltun_polski.html (7.04.2008).

³⁵ E. Sakalauskaitė-Juodeikienė, D. Jatuzis, S. Kaubrys, op.cit.

³⁶ Ibidem.

³⁷ J. Chevallier, op.cit.

³⁸ S. Gross, *Elements of pathological anatomy*, Philadelphia 1857, p. 335; E. Sakalauskaitė-Juodeikienė, D. Jatuzis, S. Kaubrys, op.cit.; H. Widacka, op.cit.

³⁹ J. Chevallier, op.cit.

Like *p. neuropathica*, also *p. polonica* has not been fully explained yet. Numerous historical sources provide readers with many different causes, symptoms, and explanations of the afore-mentioned condition. However, none of them gives an accurate description of the disease. Thus, in the mid-19th century scholars started to question if the condition exists. In 1862, a special commission in Cracow composed of 23 physicians under the leadership of Prof. Jozef Dietl suggested that *p. polonica* ‘does not exist’.⁴⁰ A similar opinion was declared by Henryk Dobrzycki, a Polish graduate of the Surgeon Medical Academy in Warsaw. In 1876, in his dissertation ‘On plait, commonly called *plica polonica*’ he argued that *p. polonica* is not a disease but an effect of ‘obscurity, prejudice and lack of hygiene’.⁴¹ Despite that, in the 21st century, there were communicated cases which were similar to affliction mentioned above. In 2000, the German magazine ‘Der Hautarzt’ came out with an article about a young man who had so-called dreadlocks – bunches of felted, matted hair. The clinical picture of this patient has resembled the *p. polonica*.⁴² In 2008, one could also read about a German 62-year-old male patient with mental illness, whose hair was very matted.⁴³

Comparison of *p. polonica* and *p. neuropathica*

As already mentioned, the term *p. neuropathica* was coined in the 1880s.⁴⁴ Not long before, in 1876, Dobrzycki, had researched on plait and concluded that *p. polonica* is in fact not a disease, but a result of specific living conditions.⁴⁵ This marks the time when the phenomenon of *p. polonica* has begun to die off. However, the amount of documented cases of *p. neuropathica* began to increase from then on. The occurrence of mental illnesses closely associated with tangled hair was still of major interest to many physicians. A closer look at their studies leads us to the question: what exactly differed those two diseases?

⁴⁰ Ibidem; H. Widacka, op.cit.

⁴¹ E. Sakalauskaitė-Juodeikienė, D. Jatuzis, S. Kaubrys, op.cit.

⁴² A. Friedli, J. Pierriard-Wolfensberger, M. Harms, *Die Plica Polonica im 21. Jahrhundert*, „Der Hautarzt“ 2000, Bd. 51, Nr. 3, pp. 201–202.

⁴³ F. Wolf, M. Scherr, D. Scherthöffer, J. Bäuml, H. Förstl, *Trichoma (Plica polonica) – a contemporary case with a historical disease*, „Neuropsychiatrie: Klinik, Diagnostik, Therapie und Rehabilitation: Organ der Gesellschaft Österreichischer Nervenärzte und Psychiater“ 2008, Bd. 22, Nr. 2, pp. 124–126.

⁴⁴ R. Joshi, S. Singh, op.cit.

⁴⁵ E. Sakalauskaitė-Juodeikienė, D. Jatuzis, S. Kaubrys, op.cit.

P. polonica hails from the countries in Central and Eastern Europe, where it was associated with some kind of supernatural influence. It was strongly believed that it could not have been treated by simply cutting off one's hair and that the results of doing so could have been lethal.⁴⁶ The characteristic symptoms included: inflammation of the scalp, which is also excessively tender and the hair-follicles secrete in abundance a reddish viscous fluid, 'which agglutinates the hair and by desiccation unites them into a solid mass'. *P. polonica* was also believed to break the patient by causing ulceration, seizures, headaches, blindness and even insomnia. It was the effect of the disease circulating in the patient's blood. The status of the disease has evolved from a national plague (it was believed to be both contagious and congenital) into an endemic nervous-rheumatic syndrome and ended as an archaic phenomenon. People from lower social classes were more prone to getting *p. polonica*, but generally, it might have affected anyone, regardless of his status, age and gender.⁴⁷

P. neuropathica was first reported in the 19th-century England, but it was also common among the members of the Sadhus tribe in India.⁴⁸ When it comes to the treatment methods cutting off plica was advised.⁴⁹ The disease manifested itself as hysteria. Other symptoms include irregular menstruation, frontal headache, prickling of the scalp and other neurotic symptoms. Contrary to *p. polonica*, in this case, the group of potential patients has been narrowed down to mostly women, who were more prone to developing symptoms of the mental illness. However, scientific research, providing the reasoning behind the cause of this phenomenon, have not been conducted.

Concerning the similarities in aetiology of both conditions, one may observe these are diseases that occur among mentally unstable people. The lack of hygiene causes acute matting and tangling of hair; it also results from the usage of herbal soaps and shampoos as well as the friction produced while rubbing the hair.⁵⁰ However, the literature still does not demystify the exact reason for developing the symptoms of mental illness by the patients suffering from plica.

⁴⁶ *Ibidem*.

⁴⁷ *Ibidem*.

⁴⁸ S. Kumar, B.K. Brar, P. Kapoor, *op.cit*.

⁴⁹ E. Elghblawi, *op.cit*.

⁵⁰ S. Anisha, K. Sukhjot, G.K. Sunil, P. Sandeep, *op.cit*.

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