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**Perspective Study** 

# The History of Restless Legs Syndrome: Some Early Clinical Observations

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The term Restless legs syndrome [RLS] was introduced by my father Karl-Axel Ekbom [1]. For several years he had been greatly interested in diseases combined with paresthesia and pain, particularly in the extremities. In 1943 he encountered a peculiar and tormenting type of paresthesia deep in the legs which he had never heard of before. The sensory disturbances proved to be the main symptom of a mainly disregarded, easily recognizable, independent disease of practical importance. The sensations appeared when the limbs were at rest, particularly in the evening and at night, and they were typically relieved by movement. Patients stated that they were experiencing an almost irresistible urge to move the legs and they had to get up and to walk around in order to get relief. Objective signs were lacking.

In 1944 he reported on 8 patients [2] and in 1945 he published his doctoral thesis, as a monograph [3], that was entitled "Restless legs": 'A clinical study of a hitherto overlooked disease in the legs characterized by peculiar paresthesia ["Anxietas tibiarum"], pain and weakness and occurring in two main forms, asthenia crurum paraesthetica and asthenia crurum dolorosa'. In his first series of patients he was able to collect 34 severe and 120 mild cases of the paresthetic form of the disease, and 15 cases of the painful form. The clinical picture was described in great detail. "Restless legs" proved to be easily recognizable and was found to be very common, about 5 % in a "normal" population. Many sufferers had one or more close relatives with creeping sensations in their legs that in these cases suggested hereditary factors as an etiological background. He also described restless legs in pregnancy and later as a symptom of iron deficiency anaemia, in carcinoma [4], and in blood donors [5]. The pathogenesis of restless legs remained obscure but it seemed reasonable to assume a disturbance at some site in the limbs. Vasodilating agents were thus tried and caused improvement in 23 out of 29 cases.

In the early literature he found only few brief descriptions of similar symptoms. In a monograph from 1861 Wittmaack referred to a condition that was called "Anxietas tibiarum" [6], being thought of as a manifestation of hysteria. Similar complaints were briefly commented upon by Beard in his book on neurasthenia [7] and by Oppenheim in his standard work [8]. The complaints seemed even to have been regarded as "a common minor ailment" only [9].

In 1955, however, Macdonald Critchley [10] noted that the

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famous English physician and anatomist Sir Thomas Willis already in the  $17^{\rm th}$  century described symptoms similar to RLS [11]. Willis emphasized motor symptoms in arms and legs that occurred when the sufferers were going to sleep: ["Leeping and Contractions of the Tendons"]. And "so great Restlessness of the Members" ensued that the diseased were "no more able to sleep". The sufferers could be most severely afflicted: as being "in a Place of the greatest Torture". Symptoms were thought to be originated in the spinal cord.

Another two most interesting descriptions of probable RLS have been found in old French literature by Konofal et al [12]: thus, Boissier de Sauvages wrote in 1763 on Anxietas tibiarum, and the description was claimed to meet the present RLS criteria. However, it was also said that the symptom of restlessness ceased as soon the patients went to bed. Konofal and colleagues also cited from a book of Gilles de la Tourette, published in 1898, a description of symptoms suggesting RLS and possibly – for the first time - so called periodic limb movements at sleep onset. The sensory and motor aspects of RLS were considered to be well described. Gilles de la Tourette included the description of RLS in a general report on what he named "neurasthenia".

In 1974 I reported together with my father KA Ekbom [13] on an early, previously overlooked, admirable description of symptoms being similar to restless legs, namely in Magnus Huss' extensive work of 1849 on chronic alcoholism [14]. Huss mentions, among others, three chronic alcoholics who in supine position had creeping sensations in both legs, between the knees and the ankles, in combination with inability to keep the legs still. The symptoms were most troublesome at nighttime, being so severe that they prevented sleep and forced the sufferer to get up.

To sum up: After the first original descriptions by KA Ekbom a vast number of papers on RLS have been published. Early comprehensive ones are for instance the thesis of Jolivet [15] and that of Coccagna et al. [16]. The disorder has been known by a number of synonyms: anxietas tibiarum, Ekbom syndrome or disease, Wittmaack-Ekbom disease, leg jitters, fidgety legs, paresthésies agitantes nocturnes des membres inférieurs, impatiences, unruhige Beine, la sindrome delle gambe senza riposo. However, it is now officially named Willis-Ekbom disease [WED].

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