

A Propos de deux cas de spondylose rhizome'lique. A history of Bechterew disease

Ohry A*.

Reuth Medical Center, Tel Aviv & Sackler Faculty of Medicine, Tel Aviv University, Israel

ABSTRACT

My grandfather, gynecologist Doctor Henryk Hersz-Lejba Lewin from Radom, Poland, introduced his MD thesis to the Faculty of Medicine, Geneva, Switzerland, in 1918, on: "A Propos de deux cas de

spondylose rhizome'lique". After briefly describing his life, a discussion on the history of Bechterew disease will be given.

Key words: Gynecologist, Lewin, history, Bechterew disease

***Corresponding author:**

Dr Avi Ohry
Reuth Medical Center, Tel Aviv & Sackler Faculty of Medicine
Tel Aviv University, Israel
e-mail: aohry@post.tau.ac.il

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"Medicine is learned by the bedside and not in the classroom. Let not your conception of disease come from words heard in the lecture room or read from the book. See, and then reason and compare and control. But see first." (Sir William Osler)

INTRODUCTION

Jan Bohdan Glinski was wrong [1]: on page 197, in his third volume "Słownik Biograficzny Lekarzy i Farmaceutów Ofiar Drugiej Wojny Światowej", he wrote that my grandfather, gynecologist Dr Henryk Hersz Lejba Lewin, died in ghetto of Czestochowa. In fact, upon the German invasion to Poland, he left Radom with his close family members, crossed the Russian border, survived the Soviet Gulags, and arrived in Palestine in 1943 with General Władysław Anders' army he died in Israel in 1961. Recently, I found his MD thesis introduced to the Faculty of Medicine of Geneva University in 1918.

Dr Hersz Lejba Henryk Lewin

He was born in Puławy in 1892. He and his brother Moszko (Mozes) went to Geneva to study medicine.



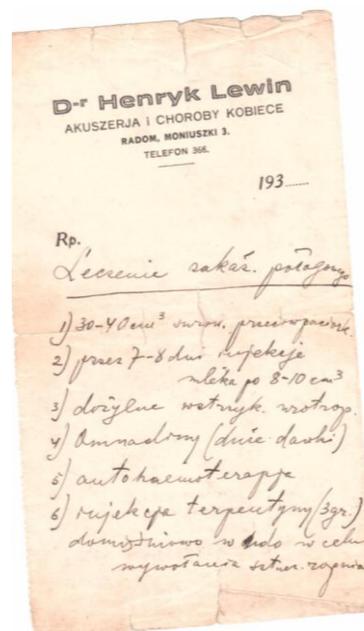
(The medical student, Henrik Lewin on the left, Geneva 1910, author's private collection)

Upon finishing his studies in 1914 at Geneva University, he returned to Poland and was recruited to the Russian Tsarist Army, so he could present his thesis only after being discharged from the Army in 1918. He decided to move to Radom, where he opened his private practice and also worked as a gynecologist to the Jewish Hospital. The building still exists but it is not a hospital anymore.



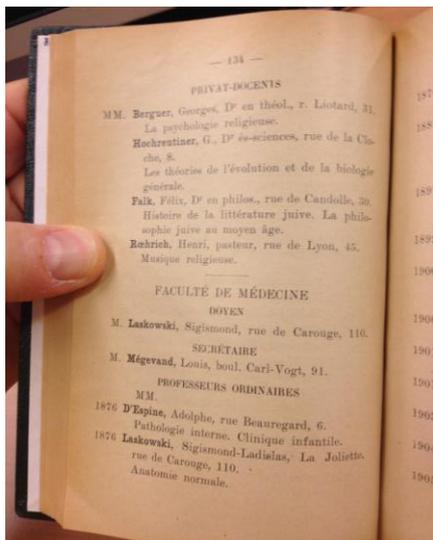
Gynecologist to the Jewish Hospital, Source: [2]

Just a note: my father-in-law, Dr (of law) Edward Kossoy [3], had graduated the high school in Radom, named after Prof. Tytus Chałubiński (Radom, 1820–1889, Zakopane) who was a Polish physician–writer, co-founder of the Polish Tatra Society, and a professor at the Medical-Surgical Academy in Warsaw. With E. Kossoy, we published a book in 1990 on the history of the Feldshers.



A private prescription of Dr Lewin in Radom. Author's private collection

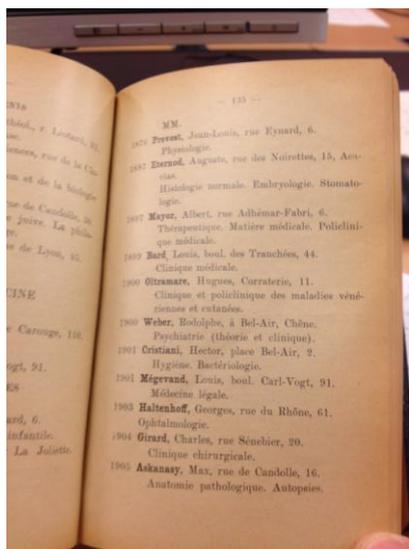
The title of this article was taken from my grandfather's thesis submitted in 1918 to the *Faculte'de Me'decine, Universite de Gene've* [4]. The thesis begins with a description and definition of the syndrome, clinical course, treatment and prognosis. The typical vertebral and skeletal pathology is described. The etiology was still unknown: Infectious? Toxic? Nutritional? How do these possible elements affect bones, ligaments and joints? Sometimes, nerves or spinal nerve roots are affected, leading to joint contractures. Usually, states the author, the clinical course is painless,



Bard had described the *Bard's symptom*-a clinical sign that “makes it possible to distinguish between acquired and congenital nystagmus” [13] and Bard's syndrome, an eponym which describes pulmonary metastases from cancer of the stomach [15]. No references are given in Levine’s thesis; in 1911 Batten’s article, there are only two names which are mentioned: Piere Marie (1853-1940), and Andre’ Léri [16].



Piere Marie (1853-1940) [15]



Author's private picture

Among them, appears Professor Louis Bard, the chief of the “Clinique Me’dicale” who was Dr. Levine’s instructor [12,13].



Louis Bard, source: [14]



André Léri (1875 - 1930) [15]

Marie provided the first description of acromegaly, muscular atrophy type of Charcot-Marie (1886), pulmonary hypertrophic osteoarthropathy (1890); cerebellar heredotaxia (1893); cleidocranial dysssostosis (1897); and finally -rhizomelic spondylosis (1898) [17]. Léri is associated with Melorheostosis, “candle bone disease“, a rare disorder of bone (Léri-Joanny syndrome), Léri- J-E Weill syndrome (a skeletal dysplasia combining dorsal subluxation of the distal

end of the ulna (Madelung deformity) with mesomelic short stature), and Léri sign (a pyramidal sign, or joint reflex seen in spastic hemiplegia) and Léri's disease-paroxysmal pain precipitated by exposure to cold and high humidity.

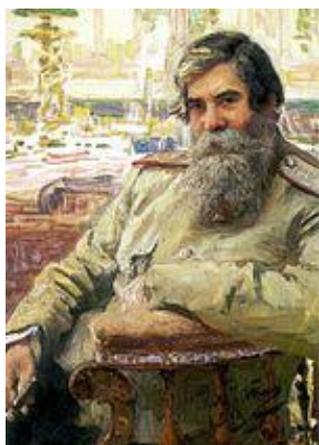
Ankylosing spondylitis

The Italian anatomist - surgeon Realdo Colombo, successor of Andreas Vesalius in Padua, published in 1559, a pathological-anatomical description of skeletal changes which fit to ankylosing spondylitis (AS) [18,19]. Most probably, the Irish doctor, Bernard Connor (1666–1698) gave the earliest account of ankylosing spondylitis. He served as a physician to Jan Sobieski (1629–1696) King of Poland in 1694, and also in the court of King Louis XIV (1638-1715). He published a book calling into question the religious basis of miraculous cures, before he succumbed probably due to malaria in London at the age of 32 years.

In 1818, Benjamin Brodie became the first to fully describe a patient who most probably had active ankylosing spondylitis who also had accompanying iritis [20]. In 1824, Benjamin Travers (1783–1858), a British surgeon, published in *The Lancet*, an account of a "curious case of ankylosis of great part of vertebral column".

In 1858, David Tucker published a report which clearly described a "famous" A.S. patient. Davies-Colley (1885) Wilks, and Charles Hilton Fagge (1838-83) had described in 1877 their patients at Guy's Hospital [21].

The French term of *spondylose rhizome'lique*, (or *arthrose de la colonne vertébrale*), Spondylarthrite ankylosante, pelvispondylite rhuma-tismale, spondylose rhizomélique, maladie de Bechterew [22], is better known in today's literature as ankylosing spondylitis or Vladimir Bechterew, Pierre Marie and Ernst Adolf Gustav Gottfried von Strumpell disease [23].



Władimir Michajłowicz Biechtieriew, source: [24]

This chronic and progressive disease is characterized by arthritis, inflammation, spinal immobility, sacro-ilitis, and cardio-pulmonary and ophthalmic complications. Batten used the French term for the disease. He also mentioned that when his patient (the patient's picture appears in the article), developed eye symptoms, he consulted with Professor Paton, an ophthalmic surgeon at St. Mary's Hospital and at the National Hospital for the Paralyzed and the Epileptic [25]. Professor Leslie Paton (1872-1943) is associated with *Paton's line* in Papilloedema and *Gowers's syndrome*. Surprisingly, the previous 1904 Gerspachen's article on tuberculose spondyloarthritis, gives a full detailed description of the various etiologies for spondylitis, and among the cited 48 references, we find the names of Léri, Marie, Babinski, Forestier, Brissaud, Strumpell, von Bechterew, and Grancher [26,27].

The Italian Giovanni Mingazzini's 1905 article [28], also gives (in German) full clinical descriptions of these syndromes, with updated references, which are absent in Levine's and Batten's papers. Mingazzini described encephalitis lethargica in 1921.

In 1929 article we can find this statement: "Thirty-five years ago von- Bechterew wrote the first of a series of papers which described a syndrome consisting of stiff spine, nerve root pains, nerve root degeneration, alterations of sensation and muscle atrophies of a radicular distribution, which he ascribed to a pachymeningitis and compression of the nerve roots. Following the descriptions of Strümpell and Marie, much discussion ensued on the classification of the various types of spondylitis, and with the exception of the work done in France and the reports by Camus, Leri, Sicard and Barré, the neurologic side of the syndrome has received little attention. In the English and American literature, references to the radicular syndrome have been few indeed, and under this title reference had not been made to spinal osteoarthritis as the causative agent, until the appearance of Rosenheck's contribution in 1924, on radicular pain in spondylitis deformans" [21]. It is clear that in our modern eyes, there are some confusion in the clinical descriptions of these (close) syndromes" [29].

It is feasible, however, that the flow of information was slow, and the availability of new coming journals was lacking. Researches were most probably, not aware of new publications and novel research. Especially from North America: The American orthopedic surgeon, Professor Abel Mix Phelps (1851-1902), was associated with "his" method to treat spondylitis, the Phelps brace, Phelps gracilis test, Phelps operation – surgery for talipes, Phelps orthosis and the Phelps splint.

Phelps graduated from Michigan University (1873), and practiced orthopedic surgery

in Germany, under Schede, Esmarch, Volkman, Billroth and Thiersch [30]. He became a prominent American orthopedic surgeon and an esteemed academic teacher. Another American pioneer in orthopedic surgery, Lewis Albert Sayre (1820–1900), performed the first operation to relieve hip-joint ankylosis and introduced the method of suspending and plaster-casting scoliotic patients. He and Phelps wrote an account of *“The history of the treatment of spondylitis and scoliosis by partial suspension and retention by means of plaster-of-Paris bandages”* [31].



Lewis Sayre, source: [32]

The American surgeon Fred Houdlett Albee (1876-1945) further developed spinal operation in Pott's disease, spinal fusion, and more [32].

"Kümmell's disease" or delayed post-traumatic osteonecrosis of a vertebral body, was coined by the German surgeon, Hermann Kümmell (1852- 1937).



Surgeon, Hermann Kümmell (1852-1937)
source: [33]

Paul Schober (1865-1943), is associated with a clinical test of the flexibility of the back, mainly used in patients with ankylosing spondylitis. The patient is *“standing, the spine is marked in two places, 10 cm above and 5 cm below the fifth lumbar vertebra. The patient is then asked to bend forward as deeply as possible. If the distance between the two points is now <20 cm, there is decreased movement in the spine”* [34]. Forestier's disease, (Forestier-Ott syndrome, Forestier and Rotés-Querol syndrome) *“is a condition fairly common in older men, marked by widespread ossification in spinal ligaments and tendons. Changes in spine simulate ankylosing spondylitis. Sacro-iliac joints spared. ESR normal”*. Jacques Forestier (1890-1978) [35]. Jean-François Calot (1861-1944) from the Hôpital Rotschild in Berck-sur-Mer, and the Hôpital Cazin-Perrochaud, developed a special 1896 method to treat the deformed tuberculous and spondylitic spine [36,37].



Paul Schober (1865-1943), source: [38]

The Literary aspect

As a child, the famous Czech author, Karel Čapek, suffered from scarlet fever, and later developed Bechterew disease: he often complained of head and spinal pain, and suffered from typical spinal deformity.

“While reading his books, I find it worth remembering that Karel Čapek was often in immense pain—he suffered from severe inflammation of the spine from the time he was a child. In the last story of Tales from Two Pockets, Čapek’s brilliant collection of mystery a narrator who suffered in this way writes: “I’ve had such respect, such a reverence in me; everything seems more important to me now...each little thing and each human being, do you understand? Everything has enormous value. Whenever I see a sunset, I tell myself it was worth that incredible pain. And people, their work, their ordinary lives...all of it has value because of that pain. And I know it’s a

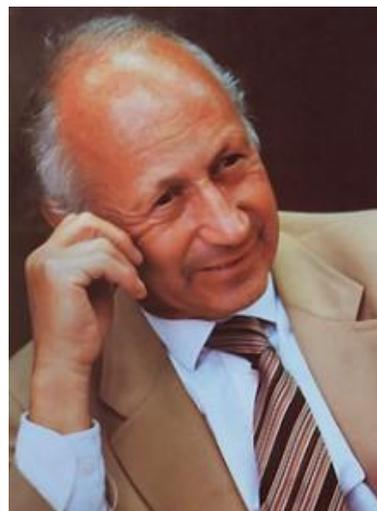
terrible and unspeakable price to pay—but I truly believe that it isn't some evil or punishment; it's only pain, and it serves to...to give life this enormous worth—“ Mr Skrivanek stopped, not knowing how to go on [39].



Karel Čapek, Source: [40]

Čapek's photograph show a serious face "marked by big eyes, a full mouth and a high forehead. Capek's smooth skin and slicked-down hair with an unruly tuft sticking up in back gave him something of an adolescent air. His wife thought his face resembled that of a beautiful African child. He would generally sport a cigarette-holder between his fingers, for he was a steady smoker, breaking each cigarette in half before placing it in the cherry wood tip. The palm of the other hand would be resting on his ever-present cane. This sometimes gave him a deceptively jaunty, Chaplinesque look; actually, the cane was a symbol of the cross that Capek had to bear most of his life, the painful spinal disease that made walking difficult, prevented him from turning his head and had a crippling effect on his personal life" [40,41].

Norman Cousins (1912-90), "was an American political journalist, author, professor, and world peace advocate" [42], wrote the: "Anatomy of an Illness" (As Perceived by the Patient) Bantam, 1981, about his Bechterew's disease. "He was told that he had little chance of surviving, Cousins developed a recovery program incorporating mega-doses of Vitamin C, along with a positive attitude, love, faith, hope, and laughter induced by Marx Brothers films. I made the joyous discovery that ten minutes of genuine belly laughter had an anesthetic effect and would give me at least two hours of pain-free sleep he reported. When the pain-killing effect of the laughter wore off, we would switch on the motion picture projector again and not infrequently, it would lead to another pain-free interval" [42].



Norman Cousins, Source: [43]

Conflicts of Interest

None

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