The 'Dejerines': an historical review and homage to two pioneers in the field of neurology and their contribution to the understanding of spinal cord pathology

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Our purpose, in this number of Spinal Cord devoted to the French speaking Society of Paraplegia (AFIGAP), is to render homage to two very distinguished doctors, who by their work at the end of the XIXth and the beginning of our century contributed greatly to our knowledge of the nervous system and in particular the spinal cord (SC). This was at the time a field of considerable interest in France and abroad. Professor Jules Dejerine was from 1911 – 1917 the holder of the Chair for Nervous System Diseases created for Charcot. Dejerine and his American born wife, Augusta Klumpke, and had very limited means for investigation compared to actual technological advances. They relied mainly on their superb clinical observations and neuropathological examinations. Dejerine was also a pioneer in the growing field of neuroanatomy. In 1895 he published a treatise on the anatomy of the nervous system, which is still considered worlwide to be a masterpiece. Augusta Dejerine-Klumpke, the first woman Intern in Paris Hospitals, was not only a fine clinician, neuroanatomist and pathologist, but also contributed greatly to her husband's work. Amongst other things she is known for the 'Klumpke palsy'. She was also a pioneer in France, during the First World War and subsequent following years, in the treatment and rehabilitation (medical and vocational) of the large number of soldiers afflicted by wounds of the nervous system and especially of the SC. During the same period, many authors contributed to SC pathology, but only a few to the treatment and rehabilitation of these patients. This was brought to our attention, in the sixties, by Professor Pierre Houssa, pioneer in Belgium in the field of comprehensive care of those who have SC lesions. Augusta Dejerine-Klumpke also contributed to our present knowledge of heterotopic ossification following a SC injury, including its pathogenesis. Most of their clinical and pathological findings and discussions are recorded in Dejerine's famous monograph which was published in 1914: La séméiologie des Affections du Système Nerveux (The Semiology of the Diseases of the Nervous System).

Keywords: Dejerine; Dejerine-Klumpke; spinal cord; semiology; neuroanatomy; historical review

Introduction

The history of Medicine offers amongst other privileges, that of being able to appreciate and look into the life and the work of people who have made significant contributions concerning the Science and the Art of Medicine. The history of spinal cord pathology, traumatic and non traumatic, has been available at least in the French language since the time of Ollivier d'Anger's book 'Le traité de la moelle épinière et de ses maladies' published in 1827.¹ More recently, the reader can refer in the English language to the writings of Sir Ludwig Guttmann² and of Ohry and Ohry-Kossoy,³ who focussed perhaps more on traumatic spinal cord

conditions than on medical causes for such lesions. Joseph Jules Dejerine (1849-1917) (he was usually known as Jules), was a person who marked his epoch not only as neurologist, neuroanatomist and pathologist but also as a straightforward very hard working and loyal person. His reputation and that of his American born wife Augusta Dejerine-Klumpke (Figure 1) were acknowledged and respected not only in France but also wordwide. The quality of their achievements in all the domain of the rising speciality of neurology was recognised in several European countries, mainly in Germany and Great Britain, but also in North and South America where French culture was particularly appreciated. Towards the latter part of the XIXth century, thanks to Forel, His, Golgi, and Ramon y Cajal, the theory of the neurone as an

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Figure 1 Portrait of the Professor J Dejerine and his wife A Dejerine-Klumpke. *DFC Phototèque; Copyright AP-HP; Paris.*

independent entity was definitely proven. The turn of the XIXth and the beginning of the XXth century was considered as '. . . the time when 'dynamic' and 'topographic' viewpoints in neurology were in conflict, with the real issues becoming more and more clouded'.⁴ Dejerine was certainly one of the pioneers in making things clear.⁵⁻⁹ This period was marked by considerable progresses in medical sciences, especially within the fields of neuroanatomy, neuropathology, clinical investigations and neurophysiology, applied and experimental. In France, people such as Duchenne de Boulogne, Brown-Sequard, Claude Bernard, Vulpian, Charcot, Pierre Marie, and others, in their time, were certainly among the many outstanding figure-heads of the French School of Neurology.^{10,11} The Dejerines, have not only been unanimously placed at the same level but their pioneering work in the fields of correlative neuroanatomy has reinforced, even today, our clinical knowledge especially of the semiology of the nervous system.¹² The latter term has unfortunately nowadays become rather obsolete.

Jules Dejerine's achievements are not only known in the fields of neurology, but also in the new science of psychiatry developed by Charcot where 'he was a man who could listen to the suffered, understand and was patient.⁶ Nevertheless, according to his contemporaries, patience was not always his usual quality. His books, and publications, had an international renown in all of the fields of neuroanatomy⁵ and neuropathology. They were beautifully illustrated and the correlation between anatomy and pathology with an amazing precision, provided an explanation of the functional and organic courses of certain diseases of the central nervous system, such as aphasia, astereognosis, apraxia, secondary encephalic degeneration, as well as disturbances of the peripheral nervous system. Regarding the spinal cord, he contributed greatly to the study of: tabes dorsalis, Friedreich's disease, multiple sclerosis, Charcot's disease (amyo-

Table 1

Dejerine hand reflex or phenomenon: clonic contracontractions of the hand (wrist) on tapping the dorsum of the hand or the volar side of the forearm near the wrist; it occurs in normal persons but is exaggerated in pyramidal tract lesions.

Dejerine-Lichtheim sign: in subcortical aphasia.

Dejerine cortical sensory syndrome: impairment in the ability to make fine sensory distinctions.

Dejerine-Roussy syndrome: or thalamic apoplexy, sometimes referred to as thalamic hyperpathia.

Dejerine-Sottas syndrome: hypertrophic interstitial neuritis in childhood, which includes the consecutive sclerosis of the posterior columns.

Klumpke-Dejerine syndrome: lower plexus lesion including a Claude Bernard-Horner syndrome when the sympathetic ramo of T1 are involved.

trophic lateral sclerosis), combined and primitive sclerosis, transverse myelitis, poliomyelitis, haematomyelia (post-traumatic and non traumatic), syringomyelia, spinal cord ataxia, hereditary progressive hypertrophic interstitial neuritis in childhood, as well as other clinical and organic aspects of disorders of the spinal cord and its nerve roots (Table 1).¹² He also described in detail the clinical signs of intermittent claudication due to vascular pathology of the spinal cord,¹² and gave a precise description of the somatotopy and the connections of the ventral corticospinal tract as well as of the lateral and anterior spinothalamic tracts (the 'faisceau en croissant' de Dejerine: the 'crescent like' fasciculus) referred to as the 'anterolateral system', (ALS) (Figure 2).^{12,13} Through his anatomical and pathological studies of the upper cervical cord and medulla oblongata, he contributed to the knowledge of the somatotopy of the trigeminal cranial nerve, its descending tracts and nucleus, with its particular 'onion skin pattern' sensory topography. This is particularly interesting in the clinical features of a patient with ascending cervical syringomyelia or syringobulbia.^{12,14} With André Thomas (1867-1963), as one of his closest collaborators,⁹ he contributed among other studies to the anatomical study of the pyramidal tract, which was also one of Babinski's clinical interests.^{15,16} Together with André Thomas, he also published in 1902 a work of reference: 'Le Traité des Maladies de la Moelle Epinère' (A Treatise on Spinal Cord Diseases).¹⁷ Other books and articles were also published in the English language on the same subject, which were cited by these authors, but appear to have been forgotten nowadays.^{18,19} In the German language, it is also important to mention Kocher's works.²⁰ André Thomas was also well known, even as a young doctor at the time, by his studies on the autonomic nervous system, especially in relation to spinal cord lesions and

many of their neurovegetative aspects. This aspect was also of great interest for Dejerine.¹² Two publications on this subject later made André Thomas famous in this respect.^{21,22} He was also, like his teacher, a physiologist, pathologist, anatomist and experimentalist, as were many of these people at that time. Dejerine marked his influence and knowledge on his colleagues and his pupils especially those who were writing and submitting their medical theses.⁶ He also concentrated much of his energy, and he had much to spare, towards the study of muscular pathology and neuromuscular hereditary diseases. He was a great admirer of Sir Henry Head's work particularly in the field of peripheral nerve pathology.

It is impossible to go into more detail concerning Dejerine's considerable work in a presentation as short as this. The greatest homage to Dejerine is probably the very comprehensive book of E Gauckler, his pupil and collaborator, published in 1923: 'Le Professeur Dejerine' devoted to his professional and family life, as well as providing a complete list of his publications and honors. More recently, one can also refer to Boucher, in Lyon, who wrote an excellent article in 1977 also rendering homage to Dr Y Sorrel-Dejerine (1891-1986), Augusta Klumpke and Dejerine's daughter.⁷ As a very young doctor, she had organised with her mother, during the First World War, the treatment and rehabilitation of spinal cord injured patients and those with post-traumatic hemiplegia. We shall refer subsequently to this important step in. She was instrumental in the 1970-80s in having two books reprinted by Masson, the famous publishers in Paris. The first named 'L'Anatomie des Centres Nerveux, 1980 (The Anatomy of the Nervous System, 1980) was first published in 1895 by her father.⁵ This was a considerable achievement in human neuroanatomy. It was written with the very close collaboration of Dr A Dejerine-Klumpke. The second 'Séméiologie des Affections du Système Nerveux' (semiology of the diseases of the nervous system), which appeared when first published in 1900 as part of a medical treatise and then in 1914.¹² as a monograph, and it was republished in 1977. Dr Dejerine-Klumke was here again his collaborator and especially supervised the figures and charts (560!). After two biographical sketches, we would like to report shortly on some anatomical and semiological aspects of spinal cord injuries.

Jules Joseph Dejerine

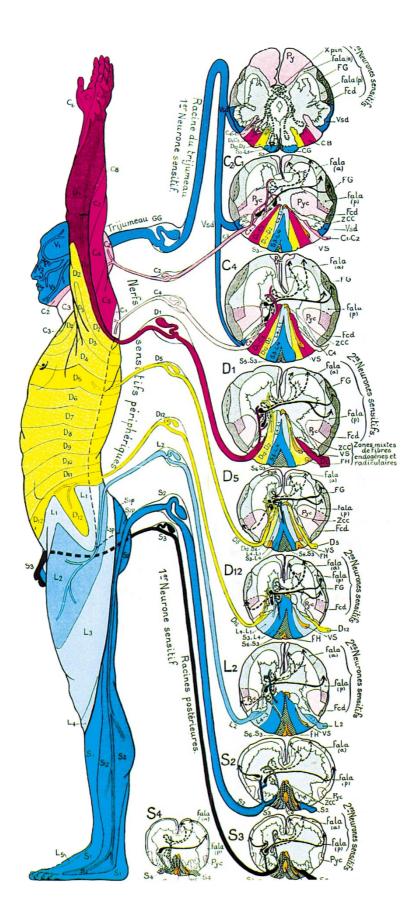
Jules Joseph Dejerine (de Gerine for some time by mistake)²³ was born on August the 3rd, 1849, at Plainpalais, in Geneva. His parents, from Savoy, in France, had modest means of existence but in spite of this, they encouraged their son to continue his studies.

In 1871, he decided to pursue medical studies in Paris. Amongst his many activities, he attended Brown Sequard's lectures and became a very close friend of him. Later, he became Vulpian's pupil, at the 'Hôpital

de la Pitié'. Vulpian was not only a great physician but also a renowned anatomist and physiologist.6,10,11,24 Dejerine had the greatest admiration for both of them. In 1879, he became Senior Registrar (Chef de clinique) in Hardy's Clinic at the 'Bicêtre' Hospital and as a Hospital Consultant (Médecin des Hôpitaux de Paris) in 1882. In 1886, after a memorable and 'heated' interview with Charcot, he was nominated Assistant Professor (Professeur Agrégé).^{6,11} In 1887, he became Chief Consultant at the Bicêtre Hospital and remained there for 8 years. During this time, Sir Charles Sherrington and many other international personalities came to visit him.⁶ Thereafter from 1895, he spent 23 years at the famous 'Salpêtrière' Hospital, at the Jacquard Pavillon. The Salpêtrière Hospital was then the Mecca of the French School of Neurology, Charcot being the head-figure.¹¹ In 1901, he became Professor of the History of Medicine and Surgery, in 1907, Professor in Internal Medicine. In 1911, he was appointed to the Chair in Nervous System Diseases in the Faculty of Medicine of Paris.²⁵ The post was previously occupied by Raymond who in 1894 succeeded the very famous and very charismatic neurologist, Charcot.^{6,11} In many respects these two personalities begged to differ, to say the least. Dejerine also became a member of the French Academy of Medicine. He did not travel much. spending his holidays, as an expert trout fisherman in the Aar river, near Bern, in Switzerland. He continued at the 'Salpêtrière' Hospital to receive visits from most of the famous neurologists from many different countries including Great Britain, Germany, United States, Western and Eastern Europe and especially Romania. He could read German and English easily in the text. This allowed him to maintain a continuous contact with other neurophysiologists and neurologists in Europe and in the American continent. He entered as an active member the Austrian (1904) and German (1907) Neurologists Associations.⁶ Amongst many national (he was Officer of the Legion of Honor) and international honors, in 1915 he received the Moxon Gold Medal, perhaps one of the greatest distinctions awarded by the Royal College of Physicians of London. In 1914 he was to give the Hughlings Jackson lecture at the Royal Society in London, but the outbreak of war interrupted the project. Dejerine died in 1917 having suffered from Bright's disease from nephritis which he had contracted several years previously.

Augusta Marie Klumpke

Augusta Marie Klumpke (1859–1927) was born in San Fransisco. The name Klumpke was of Danish origin.²³ When she was aged eleven years, she settled down with her mother, her four sisters and a brother near Lausanne in Switzerland. Her father remained in San Francisco. She was encouraged by her mother to pursue medical studies. The whole family (three of her sisters also became famous: an astronomer, a painter, and a musician), moved to Paris. She was the first woman to become *Interne des Hôpitaux* in France, this



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was after quite serious opposition from the phallo-centric medical community at this time.^{6,26,27} The difficulty was overcome by the Minister of Health and a group of influential medical colleagues, amongst whom were Vulpian and Charcot. She had in 1885, already published in relation to her medical memoir two interesting studies, one relating to radicular palsies of the lower brachial plexus and one on ocular disturbances (Claude Bernard-Horner syndrome). This is known as the Klumpke type of radicular palsy of the brachial plexus.^{28,29} After her thesis (1889)³⁰ based on polyneuritis, lead palsies (quite common at that time) and neuromuscular atrophies, she continued her studies on peripheral nerves. After her marriage with Jules Dejerine, in 1888, she performed a number of studies of secondary degenerations following lesions of the cerebral cortex, the course and connections of the Reil's ribbon (lemnisci), the red nucleus, the aberrant fibers of the pyramidal tract and the course of the cuneus fibers in the corpus callosum. This list is by no means exhaustive, as she also participated with her husband and several of his collaborators on many other studies concerning the clinical and neuropathological aspects of lesions of the nervous system. At the time, the degeneration method was in use by neuropathologists, leading to further knowledge on the somatotopy of different fasciculi in the central nervous system.^{15,31} She also described, different forms of so called 'pseudo-tabes dorsalis' ataxia with spinal cord localization, mainly of toxic origin.

Her collaboration with her husband was particularly outstanding in the realisation of the 'Anatomie des Centre Nerveux' (concerning mainly the cerebrum, the cerebellum and the medulla oblongata)⁵ and the 'Sémiologie des Affections du Système Nerveux.'¹¹ Dr Augusta Dejerine-Klumpke drew the sketches herself as well as many of the anatomical charts and diagrams published elsewhere.

As aforementioned she played a major role in the treatment and rehabilitation of patients with traumatic paraplegia and hemiplegia during the First World War, first at the 'Salpêtrière' Hospital (300 beds) and soon after the 'Invalides' military hospital in Paris under the leadership of Professor Jean Camus.³²⁻³⁴ In fact, she helped to codify the treatment of these patients and established not only a specialized medical but also a vocational rehabilitation programme in a property, near Fontainebleau, which was bequeathed to her artist sister by the French animal painter, Rosa Bonheur, and was supported financially by her family and American Friends.³³ She was in charge of the paraplegic department, the first in France's medical history. During and after the war period, she made several studies on traumatic lesions of peripheral nerves and their main trunks. In 1918, she published with Ceillier a remarkable study on the 'Paraosteoarthropathies of Paraplegic Patients by Spinal Cord Lesions'.^{35–37} This was translated into English and was published recently.^{38–40} Her pathogenic proposals are quite outstanding and most original as we can appreciate now in 1997. At the end of the war, these pioneers returned to their pre-war activities and therefore this pilot advancement ceased.³² It was taken up again after the Second World War following the example of Sir Ludwig Guttmann in Great Britain, of Munro, Bors, Abramason, Davis, Freeman, Talbot, Comarr, in the United States, and others in the Veteran's Administration Centers, as well as Jousse, in Canada. In France, the works of Maury, Benassy, Grossiord and Pelot have also to be mentioned. From 1914 to 1915, she was elected President of the 'Société de Neurologie'. One main feature in her professional life was being honored by receiving the 'Legion of

Figure 2 The first order neurone and the origin of the spinal sensory tracts (abridged). In violet colour: The nVth cranial nerve (trigeminal nerve), and the corresponding dermatomes V1, V2, V3. In pale pink: the upper cervical neurones (C2 and C4); the cutaneous radiacular zones of the neck (C3), of the skull (C2) and of the upper part of the shoulder (C4). In red: the lower cervical neurone T1 and the cutaneous radicular zones of the upper extremities (C5-T1), which receive their innervation from the plexus brachialis. *Remark:* nowadays, especially since Foerster,⁵⁵ there is a marked separation between C8 and T1. *In* yellow: the thoracic neurones T5 to T12 and the sensory radicular topography of the trunk (T2-T12). In pale blue: the lumbar neurone L2 and the sensory radicular topography of the thighs and the buttocks. *In dark blue:* the second sacral sensory neurone (S2) and the sensory radicular topography of L5-S3. *Remark:* modern authors^{50,51} agree, that L5 covers the lateral part of the leg and that S1 corresponds to the lateral part of the foot and heel and is represented on the foot sole, as in Figure 3; S2 lies in width, only a few centimeters of the posterior and lateral parts of the leg and thigh. This is due to the posterior overlap of the L2-L3 dermatomes at the thigh and L4-L5 at the leg. This is most obvious when there is an anaesthesia-analgesia of S2-S5 in complete lesions at S1. In black: the third sacral sensory neurone (S3) and the sensory radicular topography of S3. In orange (within the right funiculus posterior): the endogenous ascending radicular fibres. Pyc: lateral corticospinal-tract; Pyd: anterior corticospinal-tract; CG: Fasciculus Gracilis; CB: Fasciculus Cuneatus; vS: Schultze's bundle; zcc: cornu commisural zone; fH: Hoche's bundle; Co: Flechsig's ovale field; tGp: Philippe and Gombault's triangle at the sacral spinal cord level (S3-S6). The second order sensory neurones (a) the second order sensory neurones destinated to the thalamus: (grey oblique lines) and their anterior and posterior parts (fala a and fala p: tractus spinothalamicus anterior and lateralis); (b) the second sensory neurones destinated to the cerebellum: FcD, FG (for Gower's f): tractus spinocerebellaris posterior and anterior. The anterior and lateral spinal thalamic tracts ('Faisceau en croissant de Dejerine': the 'crescent like' funiculus) extend all along the spinal cord, lie at the periphery of the funiculus lateralis at the sacral and lumbar levels and, at the thoracic and cervical levels, are situation anteriorly and medially to the tractus spinocerebellaris posterior and anterior. In 'Sémiologie des affections du système nerveux. Copyright Masson publishers, Paris 1914

Honor', exceptional for a lady at that time, which was bestowed to her first in 1913 for her scientific work, and subsequently, in 1921, with the grade of Officer, for the work she had done to help numerous wounded soldiers, victims of brain and spinal cord injuries. The medal was given to her by her veteran paraplegic patients as an expression of their grattitude.

She was also given a token of Honor for having saved from drowning, in the Aar river, a young girl (and she also helped her husband out, who tried to rescue the girl) during a holiday near Bern, in Switzerland.⁶ She was an accomplished hostess, especially during Sunday afternoons and evenings, in their home in Paris where many friends and collaborators used to gather and discuss their projects and findings. Towards the end of the evening, the signal of departure was given by Professor Dejerine as he started to wind his clock.⁶ In 1920 the 'Dejerne Foundation' was established at the Paris Faculty of Medicine (laboratory, museum and library), consecrated to the furtherance of the neurological sciences.

Some aspects of the semiology of the spinal cord according to Dejerine

Motor function

At the time, there were many discussions concerning the somatotopy of the peripheral motoneurones. Dejerine defined the theory of the pluriradicular innervation of muscles and of the cells column disposition of the second motor neurones.^{41,42} The radicular innervation of muscles strikes us by its clarity and representation, in particular concerning the perineal muscles innervation (Figure 3).¹²

The reflexes

Reflexes of SC origin were all extensively studied in all of Dejerine's books and publications.¹² He, his wife and others, argued that the abdominal reflexes can be present where there are complete lesions of the spinal cord and they stated that they were not suprasegmental reflexes. This was also believed at the time to be the case for the Babinski reflex.^{12,16} It was also postulated that lesions of the SC with tendons areflexia and the absence of sensation and motor function below the lesion signified, that the lesion was anatomically complete. This was considered as 'Bastian's Law'.43 Dejerine refuted this theory just before and, at the beginning of the First World War, due to the tremendous influx of people with spinal cord lesions and his own animal experiments on the monkey.^{11,44,45} Later on, during and after the First World War, Bastian's theory followed a critical study by Mar-inesco⁴⁶ and by George Riddoch⁴⁷ and Guillain and Barré.^{48,49} One should not forget that, at that time, most of the patients with a spinal cord injury, except those with a very incomplete or a low lesion, did not survive. Most of them died very soon after injury in a

state of rapid progressive cachexia and toxaemia. These largely explain the disappearance of the tendon jerks or even of the cutaneous reflexes.

Figure 3 Radicular topography of the pelvis musculature in

the man, which does not correlate with the sensory radicular topography (see Figure 5). In 'Sémiologie des affections du système nerveux'¹². Copyright Masson publishers, Paris 1914

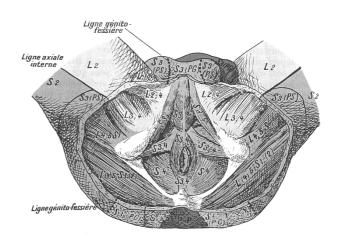
Sensory function

Dejerine's studies of mixed sensory and motor lesions, especially those concerning the different modalities of the Brown-Sequard syndrome, are impressive. He considered that this syndrome was very rarely complete.

Syringomyelia was extensively studied, both clinically and anatomically, in all of his books and articles on the subject.^{11,17} He insisted, as mentioned before, on the anatomical disposition of the spinal nucleus and its nerves within the trigeminal system. This is represented as the cutaneous 'onion skin pattern', which is clinically important for the diagnosis of ascending syringomyelia or syringobulbia.11,13,50 He also stressed the importance of examining those with high syringomyelia and syringobulbia the inner mucosae of the mouth as well as noting the paucity of the first clinical symptoms being limited to only two fingers. Incidentally, the term of syringomyelia, as such, was first described, at least in the French language, by Ollivier d'Angers¹ in 1827. This anomaly has been described in the XVIIIth century by other authors, mentioned in the book but the term 'syringomyelia' had not been coined as such.

The dermatomes, or the sensory radicular topography

The term 'dermatome' was not in use at the time, at least in France, during Dejerine's time but that of radicular sensory territory or topography, see Figures 2-5. The terminology such as 'skin fields' of afferent roots used by Sir Charles Sherrington,⁵¹ or radicular



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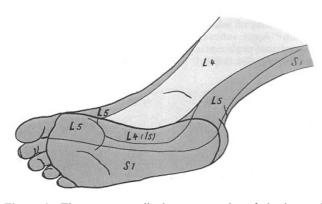


Figure 4 The sensory radicular topography of the internal malleolar area and the footsole. In 'Sémiologie des affections du système nerveux'. Copyright Masson publishers, Paris 1914

sensory distribution corresponded anatomically to the trajectory of sensory fibers, of the first or second order, within the spinal cord (Figure 2). Sherrington did use the term 'segmental', which had already been used in the German literature as segmental typus.¹⁷ The methods used to map these determinations included studies of the cutaneous manifestations of herpes zoster, 12,52 correlative clinical and pathological studies in humans,^{20,53,54} and in animals, using the method of the 'remaining sensibility'.⁵¹ Neurosurgery was not yet a speciality, but during the post-war period it brought many interesting data in the study and knowledge of dermatomes in man, especially by the work of Foerster,^{55,56} recently summarised by Bonica in his excellent synopsis on the anatomy applied to pain⁵⁰ and discussed from 1982 and still in 1997.^{57,58} One will notice that the S6 (sixth sacral root) was described by $\frac{59}{59}$ these authors, now referred to as the coccygeal nerve.⁵

The sclerotomes

The studies by J Dejerine and Augusta Dejerine-Klumpke concerning the sclerotomes in man are considered as a classic as well as the peripheral nerve supply of the skeleton.¹² These are still referred to in modern medical literature.⁵⁰ As is mentioned above, Dejerine insisted on the presence of any deep pressure sensation most distally, at the level of the foot and toes, as a sign of incompletness of the lesion, even in the absence of any form of superficial sensation below the lesion.

Conclusion

This historical review can only be considered as incomplete. We have been surprised by the number of studies concerning the spinal cord, its neuroanatomy and neurophysiology, published at the end of the XIX Century and during the first quarter of our Century. The work of the Dejerines must be considered as a

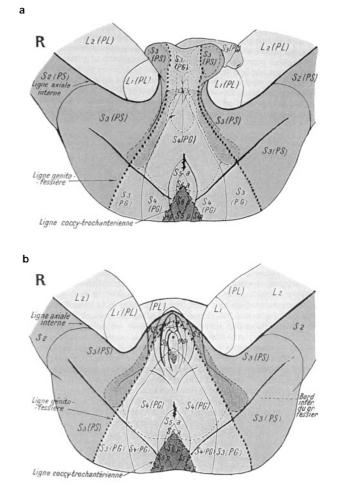


Figure 5 (a) The sensory radicular topography of the male perineum. (b) The sensory radicular topography of the female perineum. In *'Sémiologie des affections du système nerveux'*.¹² *Copyright Masson publishers, Paris 1914*

milestone in the actual comprehension of the physiopathological aspects of the spinal cord lesions (traumatic and non traumatic). Historical medical researches can be compromised by the difficulty of finding the relevant documents and our poor knowledge of the richness of clinical studies published during the above mentioned period. This review has proved to us, that many actual clinical medical studies should be revisited in the light of the works and writings of our predecessors. Our suggestion is that further historical studies on distinguished clinical and basic research neural scientists in other parts of the world should be published in the future, especially on the spinal cord.

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