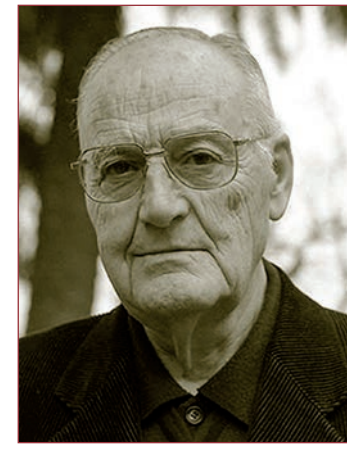
JAUME ROTÉS I QUEROL FORESTIER-ROTÉS QUEROL DISEASE

The eponyms

Forestier-Rotés Querol disease. Ankylosing vertebral hyperostosis, diffuse idiopathic skeletal hyperostosis[1-3], or senile ankylosing hyperostosis of the spine[4]. A disease characterised radiologically by the presence of ossification of the anterolateral parts of at least four contiguous vertebral bodies. It is also known as Forestier-Rotés disease[1], Forestier disease[5], ankylosing hyperostosis of Forestier and Rotés-Querol[3], and Forestier-Rotés-Querol syndrome[6]. Other names this disease has received include deforming spondylosis, spondylitis ossificans ligamentosa, and hyperostotic spondylosis[3].



Jaume Rotés i Querol (1921-2008)

Forestier-Jacqueline-Rotés Querol sacroiliac point. In the palpation of the sacroiliac joint, a point situated immediately under the posteroinferior iliac spine, where the base of the articulation is revealed and the finger can detect the presence of pain, evidence of synovitis[7].

Jaume Rotés i Querol (1921-2008)

Jaume Rotés i Querol was born on 15 January 1921 in Balaguer, where his father had a shop. He lived there until the end of the Spanish Civil War. He then began to study medicine at the *Universitat de Barcelona*, where from his fourth year on, he did an internship in medical pathology with Agustí Pedro i Pons[9]: "I discovered medicine when I met Professor Pedro i Pons in my fourth year and began to study with him. [...] I became a doctor during those years, doing patient rounds with him, which had a profound effect on my profession"[10]. He joined *Clínica Mèdica A,* at *Hospital Clínic de Barcelona* with Nicolàs Magriñà Ferrer[11]. From starting work at Pedro i Pons' clinic until 1947, he alternated working on the ward with working in the Rheumatology Dispensary, above all studying the patients with rheumatic disease.

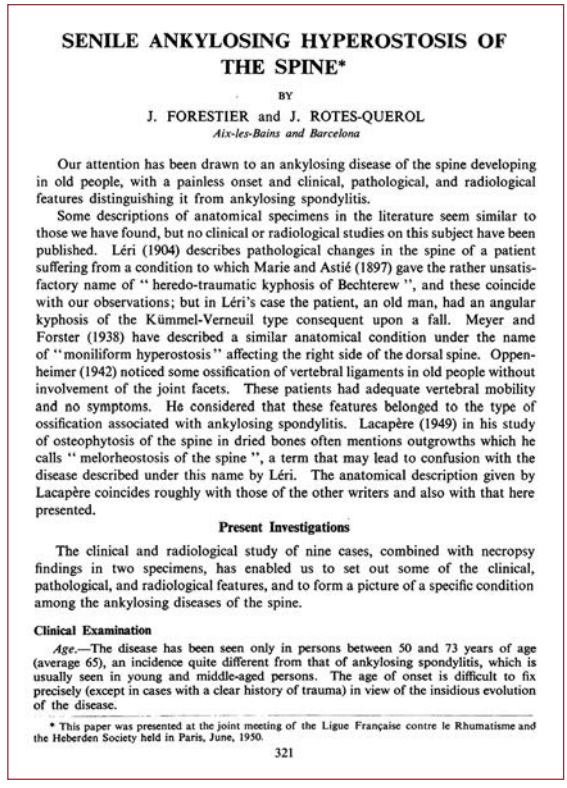
On completing his university studies, he decided he wanted to devote himself to rheumatology, so, encouraged by Pedro i Pons, who had studied both French and German medicine, he travelled to complete his training in Paris. There he worked with Stanislas de Sèze at *Hôpital Lariboisière*, remaining there from October 1947 to July 1948[11]. During those months, he became de Sèze's assistant, undertaking fascinating studies with him: "We published the first study on pain-avoiding postures in patients with herniated discs, which was a complete novelty"[10]. Two of the papers they wrote were *La hernie retromarginale anterieure* (The anterior retromarginal hernia) and *Les actitudes antalgiques par hernie discale* (Antialgic postures due to herniated discs). A year later, however, he spent the summer in Aix-les-Bains, where he worked with Jacques Forestier, who was already a renowned rheumatologist, helping to compile clinical histories. Rotés proposed to Forestier that they analyse the patients' dossiers. From that study arose a first work on peripheral arthritis in spondyloarthritis, which he followed up with publication of a book on spondyloarthritis ("writing

that book, which was quite successful, was when I made the first discovery", Rotés said[10]).

Forestier-Rotés Querol disease

To write this book, Rotés and another of Forestier's assistants, François Jacqueline, analysed 200 cases of ankylosing spondyloarthritis. Rotés became aware of something he found strange: "In some extremely rare cases, spondyloarthritis sets in when the patients are very young, but when these patients are old, they have never had pain from spondyloarthritis, have no sacroiliitis, and have some highly particular formations"[10]. He mentioned this to Forestier, who immediately became interested in the subject and gave them two new cases (autopsies). Rotés studied them and reached the conclusion that they were not dealing with spondyloarthritis or spondyloarthrosis. It was then that they described ankylosing vertebral hyperostosis. They presented this study at the joint meeting of the *Ligue Française contre le Rhumatisme and the Heberden Society* held in Paris in June 1950. It was published in *Annals of the Rheumatic Diseases* that same year[12]. The summary of this study[12], written by the authors themselves reads:

> "1) An ankylosing disease of the spine in old people is described, which may be distinguished from ankylosing spondylitis.
> 2) The pathological and radiological features have been studied in nine patients and two necropsy specimens.
> 3) X-ray films reveal the presence of bony outgrowths or hyperostoses, mainly in the dorsal region, but sometimes extending from the upper part of the sacrum to the axis. They arise from the antero-lateral aspect of the vertebral bodies and grow upwards in a 'candle-flame' formation over the lumbar disk spaces; at the level of the dorsal disks they are often thickened. They have a bony structure with a dense cortex similar to that of the head of the femur, the



SENILE ANKYLOSING HYPEROSTOSIS OF THE SPINE*

BY

J. FORESTIER and J. ROTES-QUEROL

Aix-les-Bains and Barcelona

Our attention has been drawn to an ankylosing disease of the spine developing in old people, with a painless onset and clinical, pathological, and radiological features distinguishing it from ankylosing spondylitis.

Some descriptions of anatomical specimens in the literature seem similar to those we have found, but no clinical or radiological studies on this subject have been published. Léri (1904) describes pathological changes in the spine of a patient suffering from a condition to which Marie and Astié (1897) gave the rather unsatisfactory name of " heredo-traumatic kyphosis of Bechterew ", and these coincide with our observations; but in Léri's case the patient, an old man, had an angular kyphosis of the Kümmel-Verneuil type consequent upon a fall. Meyer and Forster (1938) have described a similar anatomical condition under the name of "moniliform hyperostosis" affecting the right side of the dorsal spine. Oppenheimer (1942) noticed some ossification of vertebral ligaments in old people without involvement of the joint facets. These patients had adequate vertebral mobility and no symptoms. He considered that these features belonged to the type of ossification associated with ankylosing spondylitis. Lacapère (1949) in his study of osteophytosis of the spine in dried bones often mentions outgrowths which he calls " melorheostosis of the spine ", a term that may lead to confusion with the disease described under this name by Léri. The anatomical description given by Lacapère coincides roughly with those of the other writers and also with that here presented.

Present Investigations

The clinical and radiological study of nine cases, combined with necropsy findings in two specimens, has enabled us to set out some of the clinical, pathological, and radiological features, and to form a picture of a specific condition among the ankylosing diseases of the spine.

Clinical Examination

Age.—The disease has been seen only in persons between 50 and 73 years of age (average 65), an incidence quite different from that of ankylosing spondylitis, which is usually seen in young and middle-aged persons. The age of onset is difficult to fix precisely (except in cases with a clear history of trauma) in view of the insidious evolution of the disease.

* This paper was presented at the joint meeting of the Ligue Française contre le Rhumatisme and the Heberden Society held in Paris, June, 1950.

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First page of the article *Senile ankylosing hyperostosis of the spine* (1950), where J. Forestier and J. Rotés i Querol first described the disease bearing their name[12]

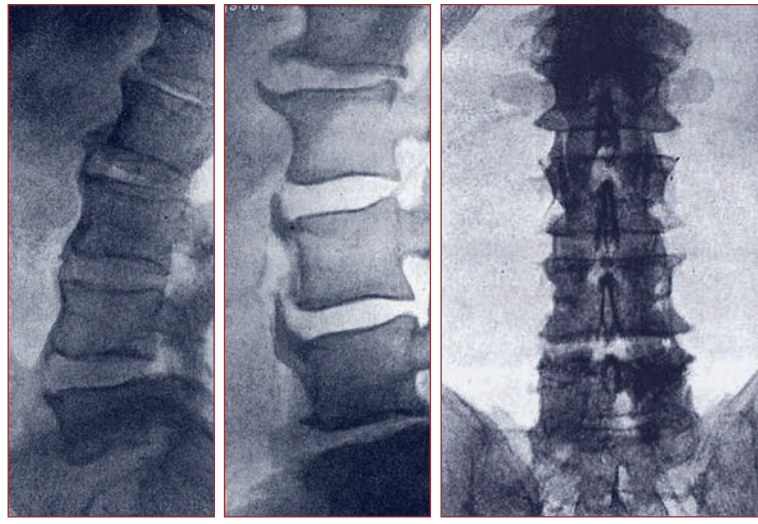
cancellous bone being in continuity with that of the vertebral bodies. They may co-exist with osteophytes, but have entirely different anatomical features.

4) Points of difference between this disease, ankylosing spondylitis, and spinal osteo-arthritis are tabulated.

5) The aetiology and pathogenesis of the condition are discussed.

6) The condition is defined as 'senile ankylosing hyperostosis of the spine'.

7) The mild nature of the symptoms gives little indication for active treatment".



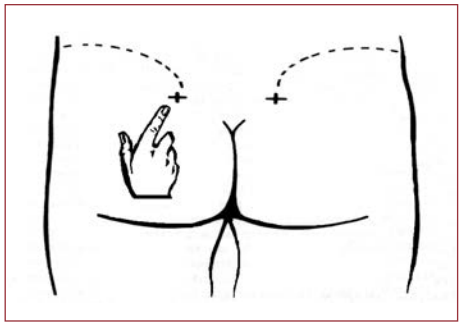
Three of the figures illustrating lumbar involvement in Forestier-Rotés Querol disease[12]

Rotés gave an interesting and detailed review of this disease in one of the chapters of his book *Reumatología clínica* (Clinical rheumatology)[1], starting with Lobstein's introduction of the term hyperostosis to describe increased bone mass in 1833. Even though this disease was not described until 1950, it is very common among vertebrates (it is also found in cats, horses, dogs, whales, and dolphins, and evidence has also been found in fossilised animals: crocodiles, dinosaurs, etc.). In humans, it is found radiologically in 6% of people over 40 years of age and in 12% of those over 70. Rotés also explained that even though in their first description they labelled it "senile", because the patients in the nine cases they studied were between 50 and 73 years old, with a mean age of 65, they dropped this adjective after they discovered numerous cases in 40 to 50 year-olds. However, the disease's link to the aging process in the broader sense was evident.

So, in summary, Forestier-Rotés disease[1], ankylosing vertebral hyperostosis or diffuse idiopathic skeletal hyperostosis[2], as described by

E. Lience, is a diffuse disease of the locomotor apparatus characterised by the calcification and ossification of the common anterior vertebral ligament, which attaches to the body of the vertebra. This condition bears no relation with vertebral osteoarthritis and its aetiology is unknown. It is more frequent in diabetics (with manifest or hidden diabetes). The generally painless process should be suspected when examination reveals rigidity in the lumbar or dorsal region —especially in patients over 50 years of age. The lateral X-ray shows an ossified band that descends attached to the anterior aspect of the vertebral body (above all between T4 and T12); the anteroposterior X-ray shows the band as a bridging shape similar to syndesmophytes on the right-hand side of the thoracic spine. It tends to be absent on the left side, apparently due to the aortic pulse. In the lumbar region, the band is discontinuous and ascending osseous proliferations appear in a candle-flame shape. In the cervical region, the lateral X-ray shows bony formations, sometimes huge, attached to the anterior face of the vertebral bodies, which do not generally form a continuous band. Extra-spinal locations have been reported, including ossifications in insertion sites of tendons and ligaments in the shoulders, knees, hips, feet, ankles, and hands[2,3].

The criteria for diagnosis are threefold: calcification or ossification of the common anterior vertebral ligament on the anterolateral aspect of at least four contiguous vertebral segments; relative sparing of the intersomatic spaces in the affected vertebral segments; and normal radiographic findings for the sacroiliac segments. The prognosis is benign, since it causes little disability. In general, it requires no treatment, apart from analgesics[2]. Although it is often an X-ray finding without repercussions, it can sometimes cause ankylosis and vertebral destabilisation, spinal-cord or visceral compressions, and disorders in the peripheral joints due to para- and juxta-articular ossifications[1]. Occasionally, extrinsic compression of the oesophagus due to exuberant ossifications in the cervical spine can cause dysphagia[3].



Forestier-Jacqueline-Rotés Querol sacroiliac point[7]

Another eponym is derived from Jaume Rotés: "Forestier-Jacqueline-Rotés Querol sacroiliac point"[7]. In the palpation of the sacroiliac joint, this point is situated immediately under the posteroinferior iliac spine, where the base of the articulation is revealed and the finger can detect the presence of pain, evidence of synovitis. One must search carefully and compare it with the symmetric side: it is painful, occurring relatively frequently in any type of sacroiliitis and is, in some cases, the only positive sign. It is recommended to seek it while the patient lies prone across the examination table, so that the base of the joint is maximally revealed and is easier to palpate. It is also the point of choice for the puncture of possible abscesses in infectious sacroiliitis, as well as the incision site for a biopsy.

Return to Barcelona

Rotés worked for three years with Forestier and then returned to Barcelona. He had also published other works: *Études statistiques sur les simptômes de début de la spondyloarthrite ankylosante* (Statistical studies of initial symptoms of ankylosing spondyloarthritis) and *La spondyloarthrite ankylosante* (Ankylosing spondyloarthritis) with Forestier and Jacqueline.

In 1956, he defended his doctoral thesis at the *Universitat de Barcelona*; entitled *Contribución al estudio de las manifestaciones osteoarticulares de la brucelosis* (Contribution to the study of osteoarticular manifestations of brucellosis). His thesis was supervised by Agustí Pedro i Pons and was published as a book in 1959.

He continued working at *Clínica Mèdica A*, in *Hospital Clínic de Barcelona*, until 1957, when he was appointed head of the Rheumatology Dispensary of the General Pathology Clinic. In 1968, Alfons Balcells i Gorina took over this post, and shortly afterwards the Rheumatology School of the Faculty of Medicine was created. The director was Jaume Rotés[11]. He retired from hospital activity at 67, becoming an honorary consultant of the *Hospital Clínic de Barcelona*.

In his extensive professional life, he was head of the Central Rheumatology Department at *Hospital Clínic de Barcelona* as well as the founder and director of the Professional Rheumatology School at the School of Medicine of *Universitat de Barcelona*, where he was also assistant professor of pathology and professor in charge of the rheumatology courses.

Among other posts, he was president of the *Sociedad Española de Reumatología* of which he was later honorary president. Furthermore, he was a founder and editor-in-chief of *Revista Española de Reumatología* (Spanish Journal of Rheumatology). He was also an honorary member of the American Rheumatism Association (1980).

He published many articles and books. The latter include *Tratamiento actual de los reumatismos, para el médico práctico* (Current treatment of rheumatisms, for the practical doctor), a book that has gone through several editions; *Estudios sobre el síndrome psicógeno del aparato locomotor* (Studies on the psychogenic syndrome of the locomotor apparatus); *La gota* (Gout), with J. Muñoz Gómez; *Semiología de los*

reumatismos (Semiology of rheumatisms), with E. Lience and D. Roig i Escofet; *Tratamiento de la artritis reumatoidea: saberes y práctica* (Treatment of rheumatoid arthritis: Knowledge and practice), with R. Sanmartí i Sala; and *Reumatología clínica* (Clinical rheumatology).

Apart from the 1950 article discussed above, he published numerous articles in prestigious national and international journals (*Annals of the Rheumatic Diseases, Arthritis and Rheumatism, British Journal of Rheumatology, Clinical and Experimental Rheumatology, Journal of Rheumatology, The Lancet, Medicina Clínica, Minerva Medica, Revista Clínica Española, Revista Española de Reumatología, Revue du Rhumatisme et des Maladies Ostéoarticulaires, Rhumatologie*, etc.). Two of these articles are especially noteworthy. In 1957, he published with A. Argany in *Revista Española de Reumatología* the article *La laxitud articular como factor de alteraciones del aparato locomotor* (Joint laxity as a factor in abnormalities of the locomotor apparatus), which was the first discussion of this topic to be published. In 1996, he published an editorial in the *British Journal of Rheumatology*[13] where he reviewed, after

On 3 December 2002, the *Generalitat de Catalunya* (Government of Catalonia) awarded Jaume Rotés i Querol the *Creu de Sant Jordi* (Cross of Saint George)

almost half a century, the disease that bears his name under the title *Clinical manifestations of diffuse idiopathic skeletal hyperostosis (DISH)*.

Rotés' work has been widely recognised, as is evident, for example, in the *Liga Reumatológica Española's* prize bearing his name: the Rotés Querol Prize on Quality of Life in Patients with Ankylosing Spondylitis. In 2000, a virtual library was created, specialising in rheumatology. Unique in its category, it bears the name *Biblioteca virtual Jaime Rotés Querol*[14], with the support of the *Sociedad Española de Reumatología*.

On 3 December 2002, the *Generalitat de Catalunya* (Government of Catalonia) awarded him the *Creu de Sant Jordi* (Cross of Saint George), "in recognition of his prestige as a rheumatology specialist, a significant symbol of which is the virtual library in this discipline bearing his name. Among his scientific contributions, his description of a new disease excels: ankylosing vertebral hyperostosis, also known as Forestier-Rotés disease"[15]. A few days later, the *Societat Catalana de Reumatologia*, on the occasion of awarding this prize, paid him homage[16]. He died in Barcelona on 29 January 2008.

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