Eponyms for Hirschsprung's disease

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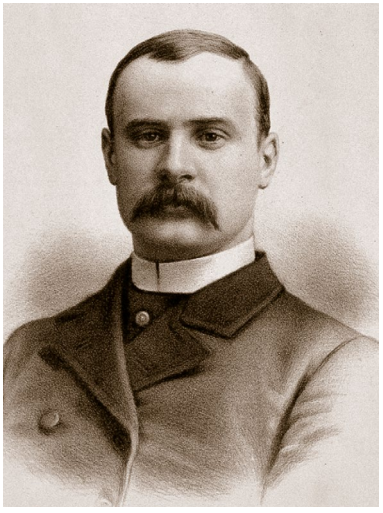
"Even though the etiology and genesis of these ulcers remain inevitably controversial, it seems highly plausible to me that ulcerations occur secondary to the primary disorder, and that these injuries are associated with the aforementioned dilatation and hypertrophy of the intestinal wall. Based on history, and as long as there are no new observations available, it will be difficult to issue a well-founded opinion regarding the development of the aforementioned conditions, which means I will refrain from conducting any theoretical deduction. However, considering that defecation impairment occurs from the very first stage of life, it seems undeniable that the underlying condition, which has been brought from the maternal womb, should be attributed either to an abnormality occurred during embryonic development or to a fetal pathological process."

Communication by **Harald Hirschsprung**
at the German Society of Pediatrics' conference
held in Berlin in 1886[1]



Frederick Treves
(1853-1923).

INTRODUCTION

Hirschsprung's disease (HD) has been subject to many –more than some people could think of– clinical descriptions over time. But it was a Danish pediatrician named Harald Hirschsprung who first presented it formally as a distinct pathological entity in 1886, after he reported the autopsy of two infants with congenital megacolon. Since then, his name has remained intimately related to this condition, adding to the extensive list of eponyms present in medical nomenclature. The use of eponyms not only reflects the history of scientific breakthroughs, but also provides a human and cultural dimension to medical knowledge. This paper aims to analyze the various eponyms associated in one way or another with HD by reviewing their origin and their relevance from a historical, clinical, and humanist standpoint.

John Raffensperger, the American pediatric surgeon who most fondly wrote on the history of pediatric surgery, said that "*if this condition were to be named in a more precise and fair manner, then three key figures should probably be honored. Constipation associated with aganglionic distal colon should be known as Hirschsprung-Treves-Swenson disease*"[2]. Hirschsprung and Swenson will be discussed in detail later, but Treves, who ironically lacks an HD eponym, is also to be mentioned.

Frederick Treves (1853-1923) was an English surgeon who became famous after he conducted an appendectomy to King Edward VII. In 1898, a few years after Harald Hirschsprung had reported his two cases, he not only accurately described the macroscopic pathology of the condition, but also guessed there was a congenital defect in the

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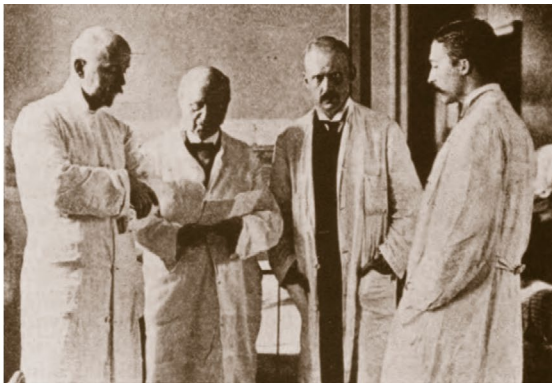


Harald Hirschprung (1830-1916). To the right, next to his colleagues *(second on the left)* at Copenhagen's Queen Louise Hospital.

rectum. He wrote: "*I dare think there is solid evidence supporting the idea that all 'idiopathic colon dilatation' cases in young children stem from congenital defects in the terminal portion of the bowel, that there is actual mechanical obstruction, and that dilatation is not idiopathic*"[2,3].

Treves demonstrated his theory in a 5-year-old girl with a hugely distended sigmoid narrowing towards a rectum the size of an index finger. In January 1897, Treves conducted a sigmoid colostomy, and a few months later, resected the colon and the rectum while exerting traction on the splenic flexure towards the anus, where it was sutured. The girl survived, with only a minor infection between the new anus and the vagina. She was seen again in 1957, 62 years after surgery, with an adhesion-caused intestinal obstruction[2].

Georg Meissner (1829-1905) and Leopold Auerbach (1828-1897).

HARALD HIRSCHSPRUNG (1830-1916)

Harald Hirschsprung was born in Copenhagen (Denmark), but his family –he was the oldest of six siblings– was Jewish-German. His father was in the tobacco business, but Harald decided to study Medicine and completed his degree in 1855. In 1862, he got married to Mariane Hertz, with whom he had three daughters[4,5].

Until 1864, he worked in the general medicine field, and in 1870, he was appointed clinical chief of the only pediatric hospital in Copenhagen back then –the Rigensgade Hospital–, a small 20-bed institution operating as a maternity hospital. Subsequently, he worked at the Queen Louise Hospital until he retired in 1904 at the age of 73 as a result of "cerebral sclerosis".

Hirschsprung is regarded by various authors as the true pioneer of pediatrics in Denmark. He was also described as a shy, reserved person, but with a strong determination. As an anecdote, he had a dispute with Queen Louise, who had ordered that biblical texts were placed at the head of every hospital bed, but Professor Hirschsprung insisted animal pictures were chosen instead.

Even though he was not the first to observe this clinical entity, his comprehensive clinical and anatomical description decisively contributed to it being defined as a distinct condition. Although this eponym has been widely accepted since the late 1800s, other authors such as Ruysch, Mya, or Galant also made an early contribution to the understanding of this disease. In Spain, the first references date back from the early 1900s, with L. Urrutia's (1914) and Enrique Súñer's (1935) work standing out[4].

GEORG MEISSNER (1829-1905) AND LEOPOLD AUERBACH (1828-1897)

In the mid-1800s, within an intense period of anatomical and physiological exploration, various key scientific breakthroughs were carried out by two European scientists –Georg Meissner and Leopold Auerbach. These advances represented a significant milestone in the understanding of the digestive tract's nervous system. Their names have remained associated with the submucosal and myenteric plexuses, respectively, which are two key structures in the autonomous regulation of intestinal function. Their observations were not mere anatomical findings, but opened the door to the study of the enteric nervous system as a

functionally independent and sophisticated entity whose disorders represent the basis of conditions such as congenital megacolon or HD.

Georg Meissner was a German physiologist born in Hannover. He was a professor at Basel, Fribourg, and Göttingen universities, and described a network of submucosal nerves in the digestive system in charge of regulating gastrointestinal blood flow, which was named after him[6].

Leopold Auerbach was a Polish anatomist and neuropathologist. Born in Wroclaw, he studied in Wroclaw, Berlin, and Leipzig. In 1862, Auerbach first described a nerve plexus between the circular-internal and longitudinal-external intestinal muscle layers, responsible for the control of gastrointestinal motility. Named after him, this plexus is crucial for the peristaltic propulsion of bowel content[7,8].

Orvar Swenson (1909-2012).

ORVAR SWENSON (1909-2012)

Orvar Swenson was born in Helsingborg (Sweden) in 1909. Son of Mormon missionaries, he emigrated with his family to Missouri (USA) in 1917. After the early death of his parents, and together with his brother Alvin, he opened a match manufacturing business for the Boy Scouts and a company named "Woodcraft." He subsequently graduated at Harvard University Medical School in Boston. He completed a one-year internship in HD research, during which he developed an intestinal resection procedure that became a cure for the disease[9].

Swenson got married to Elizabeth Criley and had three daughters. In 1945, he was appointed professor of Pediatric Surgery at Harvard. In Boston, he worked with Dr. Robert Gross and later at Boston Floating Hospital, before he moved to Chicago in 1960 as the chief of surgery of Chicago's Children's Memorial Hospital.

ORVAR SWENSON (1909-2012)

Swenson studied the bowel of HD children by using balloons both proximal and distal to the colostomy. To his surprise, there was vigorous peristalsis in the dilated bowel proximal to the colostomy, whereas no peristaltic activity was observed in the distal rectum. X-ray studies by Edward Neuhauser also demonstrated that the rectal segment was abnormally narrow in these patients[2].

These findings persuaded Swenson that the resection of the narrow distal segment, followed by anastomosis of the bowel proximal to the anus, would be a healing solution.

The procedure was first carried out in dogs to show that the distal rectum could be resected, and that by preserving the sphincter mechanism, the dogs maintained continence. Subsequently, Swenson applied this technique in one boy, resecting the narrow and aperistaltic segment distal to the colostomy using the "pull-through" technique –as he called it. This patient, named Joseph Murphy, was alive and had a normal intestinal function in December 1986, according to Raffensperger[2].

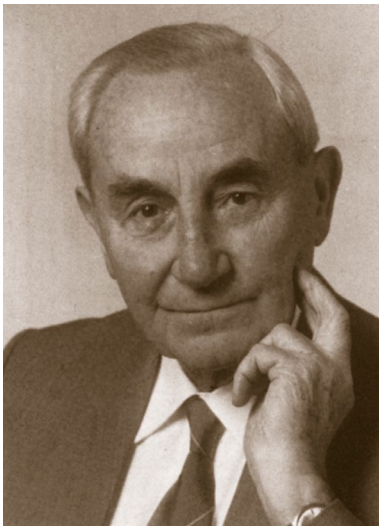
In 1948, together with Alexander Bill, he published the surgical technique bearing his name. This technique revolutionized treatment and became the gold-standard method for decades. According to John Raffensperger, Swenson "provided the scientific curiosity and genius required to correlate HD physiology and pathology and to design a healing surgery".

Histology remains the gold-standard diagnosis of HD. It was also Swenson's group who, in 1955, proposed a full-thickness rectal biopsy to achieve diagnosis. The submucosal suction biopsy technique was first published by Dobbins and Bill in 1965, and later refined by Noblett in 1969[10].

Throughout his career, Swenson received numerous international awards and distinctions, and he was president of the American Pediatric Surgical Association from 1964 to 1965. He wrote up to 100 papers published in many medical journals. His *Pediatric Surgery* book was a reference read by pediatric surgery residents worldwide. He also enjoyed sailing and woodwork. Swenson passed away in South Carolina at the age of 103.

FRITZ REHBEIN (1911-1991)

In the history of surgery, surgeons have frequently changed or tried to improve the techniques of others. Raffensperger wrote that "this may be due to ego, to a lack of understanding of the original method, or to a true desire to refine the results." Whatever the reason, the objections to the deep pelvic dissection required by Swenson's pull-through surgery, which sometimes caused damage to sphincter innervation, led to the development of other procedures, such as those by Rehbein, Duhamel, or Soave.



Fritz Rehbein (1911-1991).

Bernard Duhamel (1917-1996).

Rehbein was born in Westuffeln (Germany). He studied Medicine in Munchen, Bonn, Hamburg, and Heidelberg, and started his postgraduate studies at the University of Göttingen in 1936. His education was interrupted by WWII, during which he served as a surgeon in a field hospital. On October 17, 1951, he successfully completed the first repair of an esophageal atresia in Germany, under ether-drop anesthesia. Additionally, in 1964, he founded the *Zeitschrift für Kinderchirurgie* journal, a precursor of the current *European Journal of Pediatric Surgery*, which he was editor of until 1983[11].

In 1953, he developed the procedure bearing his name for HD. Contrary to Swenson, his method avoided deep pelvic dissection, with lesser risk of incontinence and nerve damage. Since an aganglionic rectal stump was left in place, some patients developed persistent constipation, enterocolitis, or incontinence, but Rehbein successfully applied it in nearly 400 patients[2].

BERNARD DUHAMEL (1917-1996) AND THE RETRORECTAL TECHNIQUE

Bernard Duhamel was born in Paris (France) in 1917. His father was an award-winning author, member of the French Academy, and his mother was an artist. After finishing his studies in Medicine, he started his surgical education in Paris in 1939. Subsequently, he completed an internship in Pediatric Surgery at Paris' Hôpital des Enfants Malades[10].

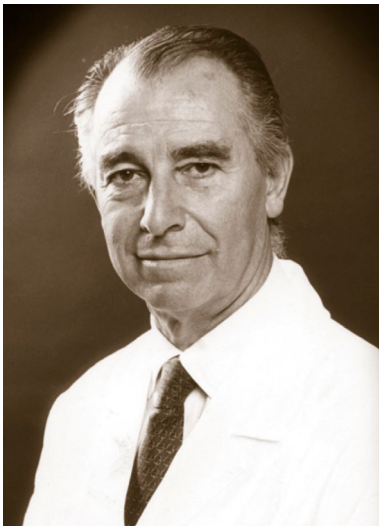
In 1954, he was appointed chief of Pediatric Surgery at *Saint-Denis Hospital,* and in 1955, he was promoted to surgery professor. Duhamel and Prochiallté conducted the first successful esophageal atresia surgery in France, and founded the first French neonatal surgery unit in 1947. In 1953, he published his book *Chirurgie du Nouveau-Né et du Nourrisson*, which confirmed him as an international figure[10,12]. He was elected president of the French Pediatric Surgery Society in 1967, and was a prominent member of the French Academy of Surgery. He was the first editor of the *Annales de Chirurgie Infantile* journal, which merged with *Zeitschrift für Kinderchirurgie* in 1991 to become the *European Journal of Pediatric Surgery*, as previously stated. Duhamel not only was a surgeon and a researcher –he was also a musician who directed an important amateur orchestra for 25 years[12].

In 1956, he proposed an alternative to Swenson's technique –a lateral anastomosis between the normoganglionic colon and the aganglionic rectum, while preserving the latter to maintain continence. This technique was widely adopted in Europe and has been subject to changes over time to improve results.

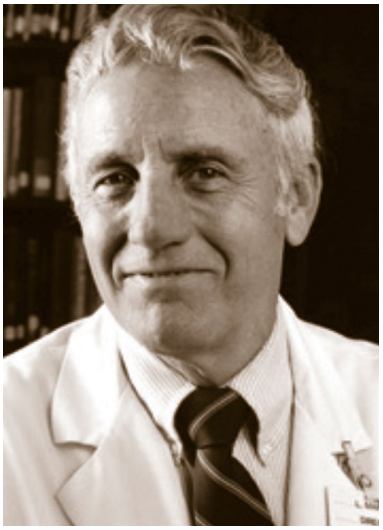
FRANCO SOAVE (1917-1984) AND THE DEVELOPMENT OF THE ENDORECTAL TECHNIQUE

Born in Naples (Italy) in 1917, Franco Soave graduated in Medicine at the University of Genova in 1943. He completed his residency in surgery at the Surgical Clinic of Turin University in 1950. He returned to Genova as an assistant professor from 1951 to 1954, and was also part of the medical staff. He was then appointed chief of surgery at Instituto Gaslini's Hospital, and also senior professor and chief of the Pediatric Surgery department at the University of Genova, where he worked until his last days[13].

In 1964, he developed the technique bearing his name, which involves the de-mucosalization of the aganglionic rectum and the introduction of the normoganglionic colon through the preserved muscle layer[14]. This technique aimed to reduce complete-rectal-resection-associated



Franco Soave (1917-1984).



Lester Warren Martin (1923-2020).

complications and to preserve sphincter function. It was recently published that this technique had already been carried out by Afro-American surgeon Asa G. Yancey[15] 12 years before it was reported by Soave, but in an adult patient.

Soave published more than 160 papers and was part of various editorial committees, including that of the *Journal of Pediatric Surgery*. He was awarded honorary memberships in several international pediatric surgery societies and was president of the Italian Pediatrics Society from 1970 to 1972. In 1984, he published his most significant work presenting an alternative surgery for HD in the American Pediatric Surgical Association. Regrettably, he passed away that very same year.

In 1998, Mexican surgeon Luis de la Torre Mondragón published a modification of Soave's technique, initially named "transanal endorectal descent," but later known as "de la Torre" technique globally. Developed at Mexico's National Institute of Pediatrics, this technique involves conducting the whole surgery –including mucosectomy, resection of the aganglionic colon, and anastomosis– transanally, thus removing the need for laparotomy or laparoscopy[16].

LESTER W. MARTIN (1923-2020): A TECHNIQUE FOR THE LONG SEGMENT

Lester Warren Martin, an American Missouri-born surgeon, started his medical education at Missouri University before he moved to Harvard to complete his studies. In this period, he served in the army during WWII[17]. He was trained in Pediatric Surgery in Boston under the guidance of Robert Gross, and in 1957, he moved to Cincinnati to become the first full-time pediatric surgeon in town. He led the Pediatric Surgery department for 21 years, and his contributions to our specialty are numerous[18]. He conducted the first liver and kidney transplantations in Cincinnati, and developed a new approach for long segment HD known as Lester Martin's technique –a side-to-side healthy ileum anastomosis with a segment of the aganglionic colon. He also designed an ileoanal reservoir procedure for ulcerous colitis in pediatrics[19].

In the USA, he is known for his contribution to the Pediatric Surgery education program, thanks to which he was awarded the Daniel Drake medal from the University of Cincinnati in 1990. He also received the Ladd medal from the American Academy of Pediatrics' Section on Surgery.

The history of HD, which is reflected in the eponyms associated with clinical descriptions and surgical techniques, illustrates the profound bond between the progress of medical knowledge and the names accompanying it. Since Harald Hirschsprung –and even if there are previous descriptions of HD–, every first and last name represent not only a technical contribution, but also a distinct stage in the understanding and treatment of this complex pathology.

Acknowledging these eponyms is not a merely historical review, but an exercise to assess the progressive development of medicine. It also helps pay tribute to those who transformed the lives of thousands of patients thanks to their innovation and devotion.

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