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REVIEW ARTICLE

Surgical treatment of birth-related brachial plexus injuries: a historical review

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Abstract
This historical review presents the relevant data about the evolution of the surgical treatment of neonatal brachial plexus palsy. Starting with the first clinical description by Smellie in 1754, we will present the initial enthusiasm for the surgery followed by a lack of interest that lasted many years, the resurgence of interest in operative management in the 1970s, and the consolidation in the 1980s of surgery as the standard indication in cases of neonatal brachial plexus palsy without a functional spontaneous recovery.

Keywords Neonatal brachial plexus palsy · Surgical treatment · History

Introduction

Neonatal brachial plexus birth palsy consists of flaccid paralysis of an upper limb secondary to injury to one or several brachial plexus elements occurring during delivery. Although known since antiquity, neonatal brachial plexus palsy was mixed with other congenital anomalies for centuries. It began to generate medical interest only toward the end of the nineteenth century.

Early clinical descriptions

William Smellie (1697–1763) (Fig. 1a), a Scottish-born physician and instructor in obstetrics who practiced and taught in London, is commonly credited with the earliest known English language description of brachial plexus injury at birth, in 1754. Smellie published a three-volume collection on obstetrics. The first volume, "A Treatise on the Theory and Practice of Midwifery," was published in 1752 [41]. In 1754, the second volume, "A Collection of Cases and Observations in Midwifery," was published [38] (Fig. 1b). The third and final volume, "Collection of Preternatural Cases and Observations in Midwifery," was published in 1764 [39], 1 year after Smellie's death. Though none of these three volumes contain images, Smellie published a set of 37 images of the human pelvis, the abdominal region and fetuses, and forceps in 1754, which were masterfully illustrated (Fig. 1c), mainly by Jan van Rymsdyck. The title of this volume was "A Set of Anatomical Tables with Explanations and an Abridgment of the Practice of Midwifery" [40]. William Smellie became a prominent figure within midwifery in Britain and throughout Europe, and the 1370 pages of his books were translated into French, German, and Japanese.

In the chapter "Of laborious cases from the presentation of the forehead or face, in which the women were delivered by the forceps" of the second volume of his treatise (case III, pp. 506–509) [38], Smellie mentions a case of bilateral upper limb paralysis following a difficult delivery with face presentation that resolved in a few days and discussed the relation of brachial plexus injury with difficult obstetric manipulation:

"In the year 1746, about nine o'clock in the morning, I was called by a gentleman who had formerly attended my lectures, to a woman in labour, and found the child's face presenting… Both blades (forceps) being thus introduced in the same direction, and the handles locked together, I pulled gently, moving the head from ear to ear, until it was brought lower down into the pelvis, then with the assistance of two fingers pressed above it, I turned the chin,

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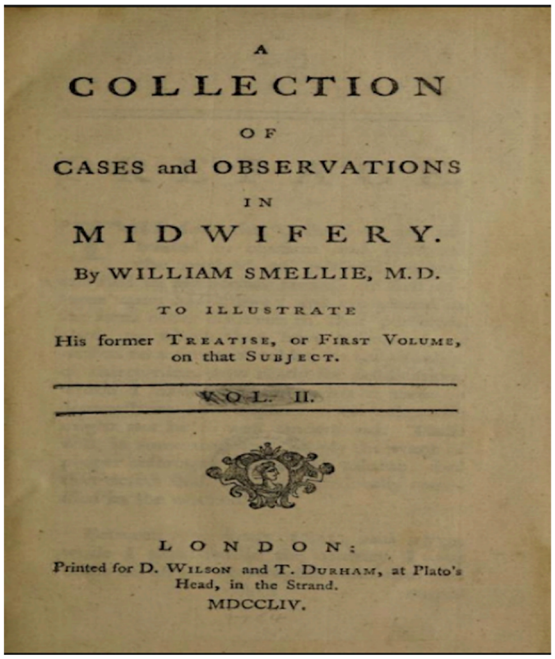

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Fig. 1 William Smellie and his treatise on midwifery. **a** Portrait of William Smellie (1697–1763). **b** Second volume of the treatise, in which he described a case of birth-related brachial plexus palsy for the first time. **c** Table 12, illustrating the complimentary volume "A Set of Anatomical Tables, with Explanations, and an Abridgement of the Practice of Midwifery," and showing a gravid uterus

and anterior part of the neck forwards, from the lower part of the ischium, to the space below the pubes, so that the forehead was at the same time turned from the left ischium to the lower part of the sacrum and coccyx. Lastly, I moved the handles towards the pubes, and delivered the woman of a child, whose face was swelled, and whose head was compressed ...the long compression had rendered the arms paralytic for several days, though this misfortune was soon remedied by friction and embrocations."

A few other observations of spontaneously resolving upper plexus paralysis appeared in the literature in the following years [5, 20], but it was only in 1872, 118 years after Smellie's second book was published, that a French physician, Guillaume Duchenne (Fig. 2a), first brought the condition to

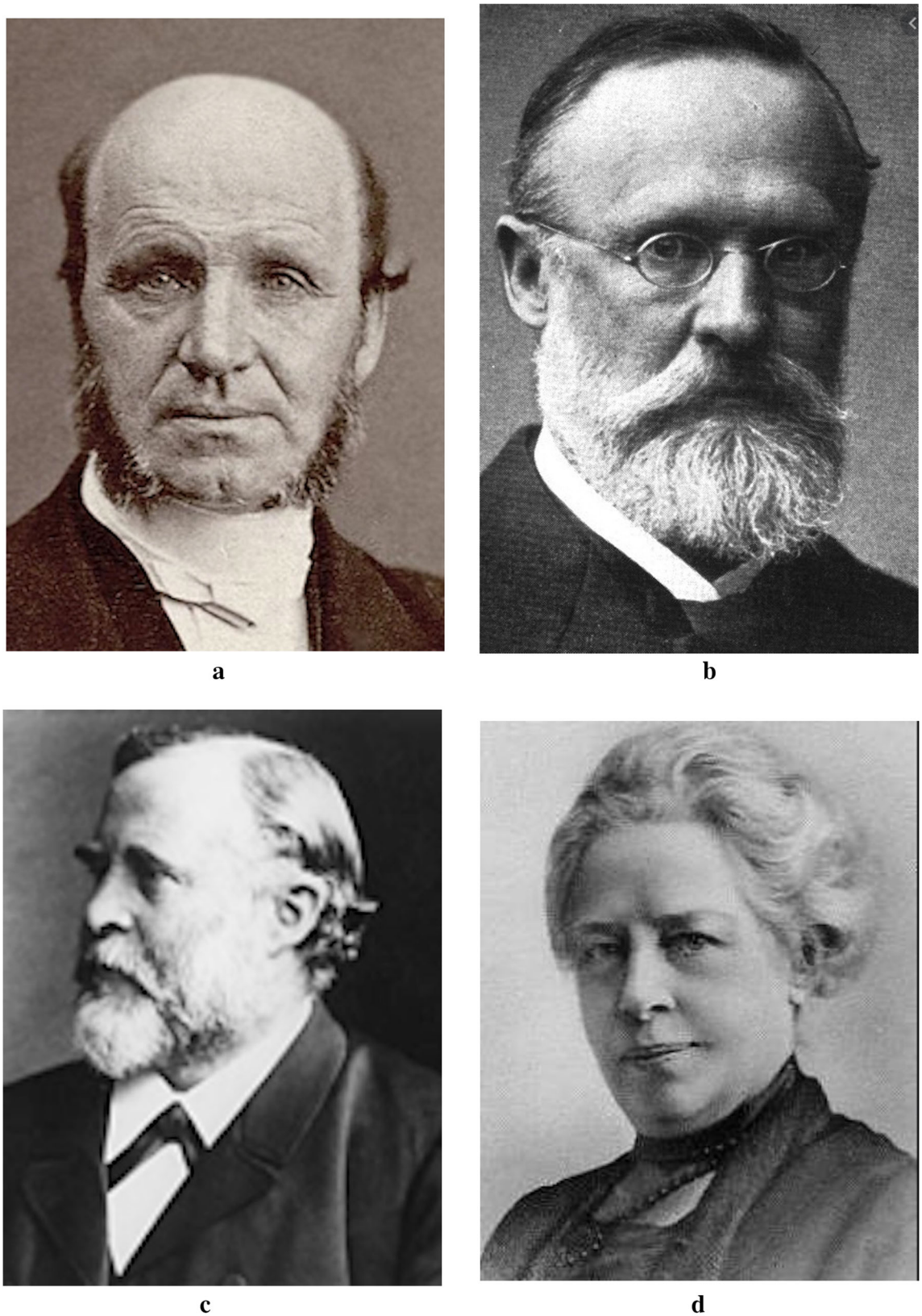

Fig. 2 Authors who described the different types of brachial plexus palsy. **a** Guillaume Benjamin Amand Duchenne described the upper brachial plexus palsy in children. **b** Wilhelm Heinrich Erb described the upper brachial plexus palsy in adults. **c** Otto Ludwig Adolf Seeligmüller described the total plexus paralysis in children. **d** Augusta Marie Déjerine-Klumpke reported the first description of an isolated involvement of the distal roots of the brachial plexus

the notice of the medical profession. He attributed the clinical picture of the neonatal brachial plexus palsy to an injury brought about by force applied during the birth of the child in an effort to deliver the shoulder. In the third edition of Duchenne's book "De l'Electrisation Localisée et de son Application à la Pathologie et la Thérapeutique" [6], he described four children with upper brachial plexus lesions and introduced the term "obstetric paralysis," thus confirming that the lesion of the brachial plexus was caused by traction during birth and that it was not a congenital lesion. Duchenne also described the typical position of the upper limb of these newborns, in which the arm is kept extended at the side, medially rotated at the shoulder joint, and the wrist is volar flexed, while the fingers are extended (Fig. 3). This typical limb posture was given the name "policeman's tip" [35]. However, possibly to avoid any doubt about the incorruptibility of the police, the phenomenon was subsequently designated "waiter's tip position."

Shortly afterwards, in 1874, the German neurologist Wilhelm Erb (Fig. 2b) described a form of paralysis of the brachial plexus in four adults with the same characteristics as those described previously by Duchenne in children [9].

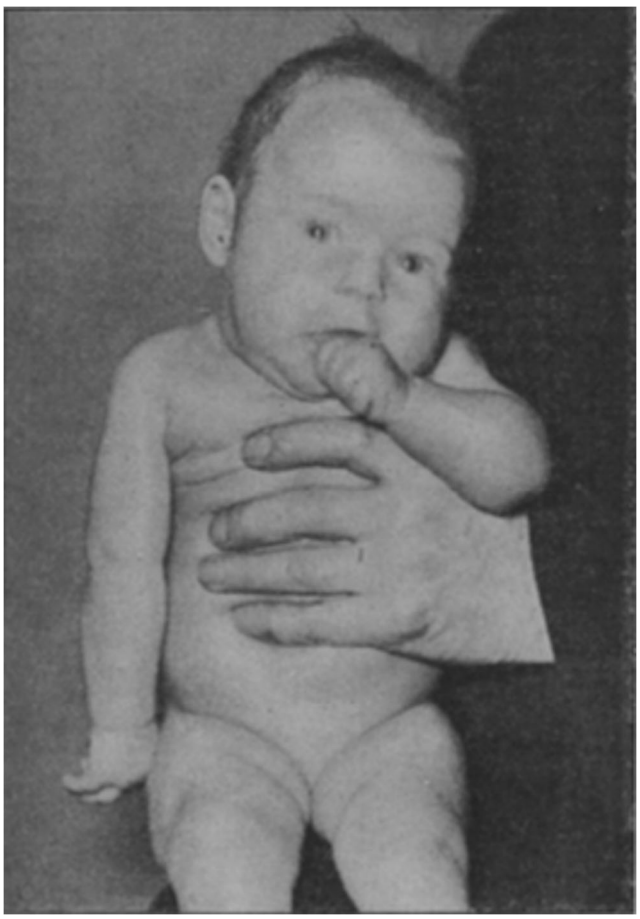

Fig. 3 The typical limb posture of an upper brachial plexus lesion on the right side as described by Duchenne [23]

Fig. 4 **a** William Thorburn, who was the first to suggest, in 1903, that excessive traction during delivery was the cause of neonatal brachial plexus palsy. **b** Engelhard agreed with Thorburn's theory and **c** presented this photograph demonstrating the result of excessive stretching during the delivery

He studied several anatomical preparations and concluded that the paralysis in his patients was caused by damage of the fifth and sixth cervical nerve roots. Later on, Erb found in healthy individuals that electrical stimulation of a distinct point on the skin, 2 to 3 cm above the clavicle, along the posterior border of the sternocleidomastoid, and at the level of the carotid tubercle of the sixth cervical vertebra, provoked contraction of the same muscles as those affected in his patients, i.e., the deltoid, supraspinatus, biceps brachii, brachialis, and brachioradialis. This point corresponds to the position where the anterior divisions of the fifth and sixth cervical nerves unite to form the upper trunk. It came to be known as "Erb's point" [10] and is the usual location of injuries in the most common form of neonatal brachial plexus injury. Duchenne and Erb have become linked through the hyphenated eponym of Erb–Duchenne brachial plexus palsy.

In 1827, Flaubert described complete brachial plexus paralysis in an adult after a forcible reduction procedure for an old shoulder dislocation [14]. However, it was only 50 years later that Adolf Seeligmüller (Fig. 2c) described three cases of total plexus paralysis associated with Horner's syndrome in infants [34].

In 1885, the neurologist and neuroanatomist Augusta Déjerine-Klumpke (Fig. 2d) provided the first description of paralysis of the lower roots of the brachial plexus, which compromised function and sensation in the forearm, wrist, hand, and fingers [24]. She also noted that involvement of the sympathetic fibers (Horner's syndrome) in this type of paralysis was relatively frequent. This form of neonatal brachial plexus injury has become known as Klumpke's palsy and is much less common than Erb's palsy.

Etiology and pathogenesis

Toward the end of the nineteenth century, interest in the etiology and pathogenesis of neonatal brachial plexus injury developed. Biomechanical and anatomical experimental work was done in an attempt to understand the physiopathological mechanisms of this condition.

In 1897, Fieux made a theoretical study of the mechanical factors involved in brachial plexus injury and confirmed his theories using rabbits [13]. In 1898, Duval and Guillain [7] studied the anatomy and relationships of the intraspinal segment of the plexus, calculated the angles of the roots with the spinal cord, and then induced plexus lesions with traction of

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the upper limb. They confirmed the work of Fieux and stated that the different lesions observed in traumatic brachial plexus can be explained by the anatomical disposition of the roots. In 1903, Thorburn was the first to suggest that the lesion in neonatal brachial plexus palsy is usually the result of excessive traction or rupture of the brachial plexus during delivery [49] (Fig. 4a). To test Thorburn's assumption, Engelhard [8] (Fig. 4b) investigated the influence of different positions during delivery and assisted deliveries on a dead fetus, in which the brachial plexus was dissected (Fig. 4c). In his doctoral thesis presented in 1906, he wrote that the pressure theory was highly improbable and that the possible etiology was excessive stretching of the plexus during the delivery. He mentioned that care should be taken to avoid inferior traction of the head of the fetus in cephalic presentations with shoulder dystocia and to avoid excessive lateral movements of the fetus body in breech presentations.

Early surgical interventions

Motivated mainly by dissatisfaction with the results from clinical treatment, the beginning of the twentieth century saw the appearance of direct surgical treatment of these lesions and 1903 was a golden year for this surgical endeavor [2].

Three years after the first detailed repair of a stretch injury to the brachial plexus of an adult had been reported in Manchester [48], Robert Kennedy, an assistant surgeon at the Western Infirmary in Glasgow (Fig. 5), published the first description of surgical treatment for an obstetrical brachial plexus lesion. He reported three cases in which a neuroma of the upper trunk was excised and the three distal divisions (suprascapular nerve and anterior division and posterior division of the upper trunk) were sutured to the fifth and sixth nerves, using "fine chromatized catgut" [22]. The resection of the lesion resulted in a gap, and in order to suture the stumps directly, the infants were immobilized with the shoulder pushed upward and the head tilted to the side, such that the operation and suturing were performed after the shoulder and head had been brought closer together. At the time when the paper was published, only one of the patients had had sufficient time for recovery. This male infant was operated in 1902 at the age of 2 months, and the pictures of the patient that were taken at the age of 9 months [22] and 2 years [23] after the surgery showed a result that would gratify most present-day surgeons (Fig. 6a–c). One year later, Kennedy had extended his surgical series to five cases of neonatal brachial plexus palsy [23]. Furthermore, also in 1903, Vincent Warren Low (Fig. 7) of St Mary's Hospital in London, jointly with the neurologist Wilfred Harris, presented the first attempted intraplexal neurotization for treatment of a neonatal brachial plexus lesion at a meeting of the British Medical Association, with subsequent publication [19]. They

Fig. 5 Robert Kennedy, a surgeon at the Western Infirmary in Glasgow, who published the first description of surgical treatment for neonatal brachial plexus palsy in 1903

implanted the distal stump of a ruptured C5 spinal nerve into a healthy C6 spinal nerve in a child with Erb's palsy.

Two years later (1905), Alfred Taylor (Fig. 8), a professor of surgery at Cornell University Medical College in New York, together with a neurologist (LP Clark) and a pathologist (TP Prout), described seven surgical cases using Kennedy's technique. They recommended early surgeries: at the age of 3 months in cases of partial paralysis and 1 month in cases of complete paralysis. They also investigated the effect of stretching the brachial plexus in stillborn children and demonstrated the site and manner of rupture [4]. In 1907, Taylor published a further nine cases in a nicely illustrated paper [46], and in 1920, he described his experience with 200 cases of obstetrical paralysis, 70 of which were treated operatively [45]. The results were not precisely described, but the mortality rate was low (4%).

Other cases were published during this early period by Lange [27], Fairbank [11], and Spitzy and Lange [43], but it was not until 1917 that surgical intervention became a generally accepted solution. In that year, Wyeth and Sharpe [50] published 81 cases of obstetrical paralysis that were treated surgically. The results were not

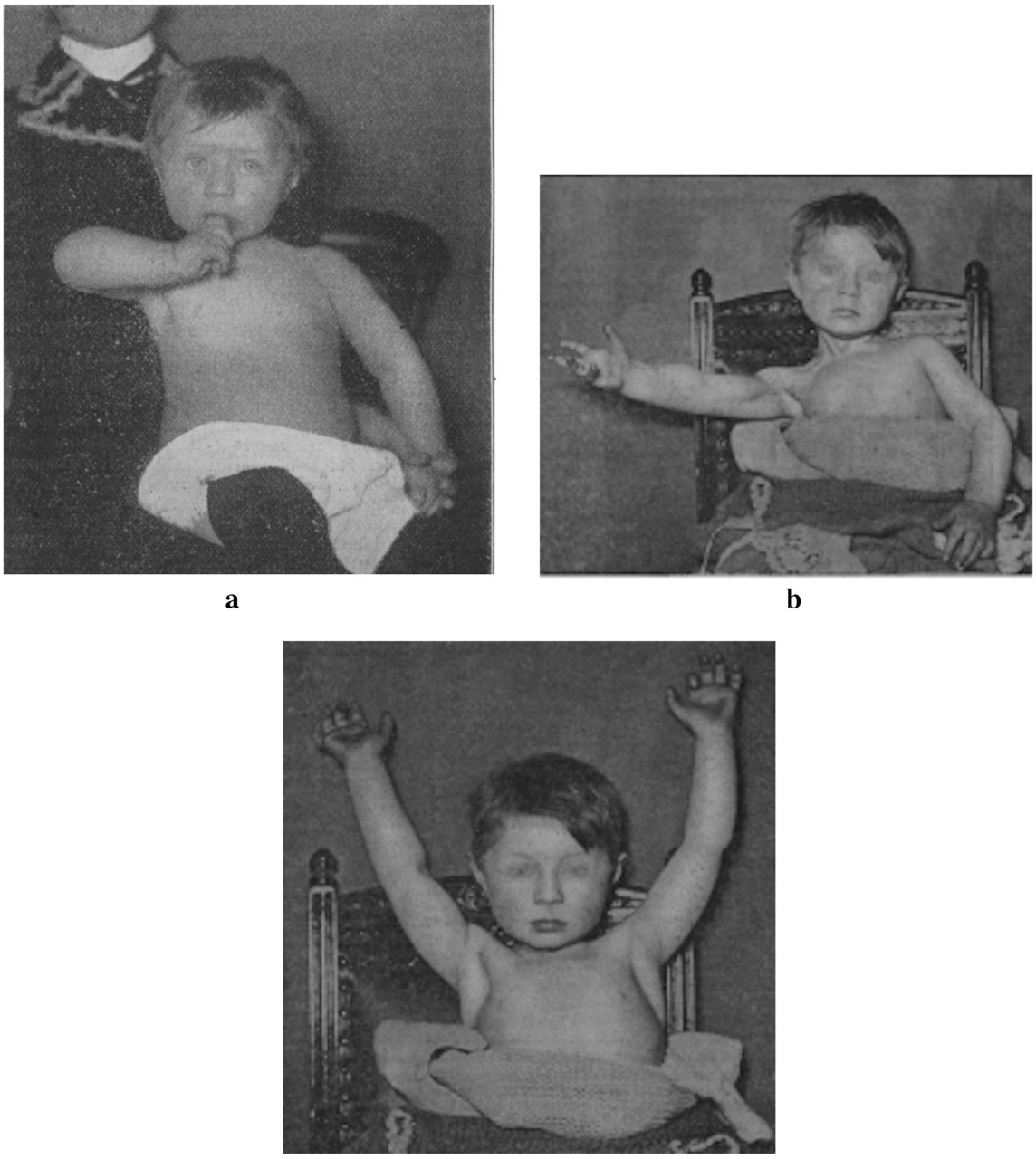

Fig. 6 Surgical results from a patient with right-side Erb–Duchenne paralysis operated by Robert Kennedy in 1902. **a** Nine months after the operation: restoration of the power to abduct the arm and flex it at the elbow joint. **b** Two years after the operation: recovery of supination. **c** Two years after the operation: recovery of deltoid function [22, 23]

presented, but the authors recommended that surgery should be performed at 1 month of age if the paralysis was complete and at 3 months if it was incomplete.

The last surgical report of this early period of surgical treatment was published by Lauwers in 1930 [28], which was the first report in the French literature. This author described nine surgical cases that he had treated personally, mostly by means of neurolysis.

The emergence of secondary surgery

After 1930, the early interest in surgical treatment for neonatal brachial plexus palsy progressively decreased and this approach fell out of favor. The main reasons for this were the difficulties of surgery, the risks of anesthesia in babies, the high mortality rates during or just after operation, and the usually poor results. During

Fig. 7 Vincent Warren Low, of St Mary's Hospital in London, who published the first attempt to perform intraplexal neurotization, as the treatment for Erb–Duchenne paralysis, in 1903

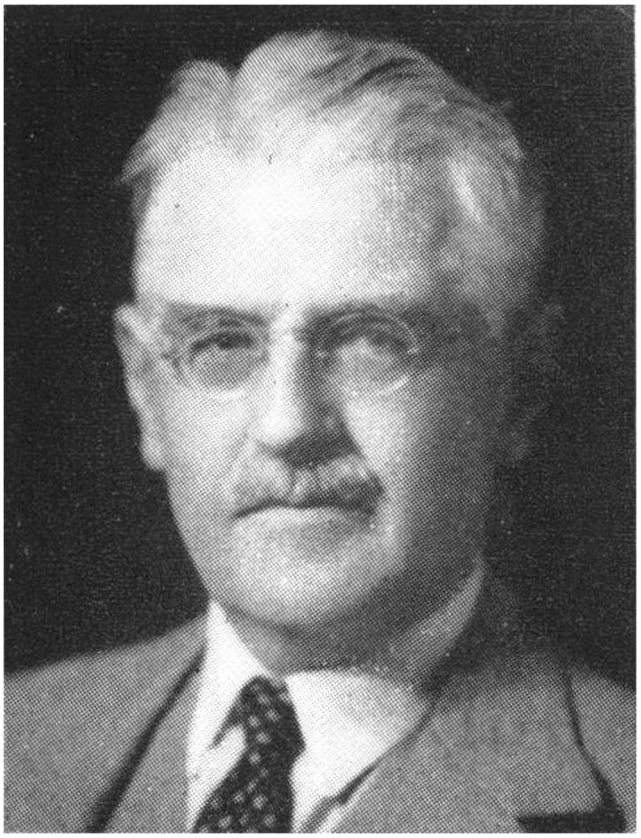

Fig. 8 Alfred Taylor, professor of surgery at Cornell University in New York, who published surgical cases of neonatal brachial plexus palsy using Kennedy's technique, for the first time in the USA, in 1905. In 1920, he described his experience with 200 cases of obstetrical paralysis, among which 70 were treated operatively

this period of lack of interest in direct plexus repair, the secondary operations were extensively practiced and developed. The reports of James Sever (Fig. 9), an orthopedic surgeon at Boston Children's Hospital, certainly contributed to the decrease in interest in surgical treatment. In 1916, he reported on 470 infants [36] with neonatal brachial plexus palsy, and in 1925, he published a follow-up report on 1100 infants with brachial plexopathy [37]. He recommended against operating on the brachial plexus itself because good outcomes were uncommon and a number of cases were worsened by surgery. Although he considered that most cases were nonoperative, he developed a surgical procedure that released the pectoralis major and subscapularis muscles, while leaving the nerves themselves undisturbed, to treat children who only had limited range of motion as the result of the nerve damage to the brachial plexus. Joseph Battiato L'Episcopo, an orthopedic surgeon in New York, published two critical articles on tendon transplantation and restoration of muscle balance in the shoulder for brachial plexus patients [25, 26]. He adapted Sever's procedure and, through a combination of muscle releases and transfers, achieved increased external rotation and abduction. This technique became known as the Sever–L'Episcopo procedure and it is still used today, with some modifications.

For approximately 50 years, surgical treatment of neonatal brachial plexus palsy entered into a more conservative period with the adoption of a "wait-and-see" attitude. As a result, publications about primary surgical repair of neonatal brachial plexus palsy became scarce.

Resurgence of primary surgery

The revival of efforts toward surgical repair of the brachial plexus in children is often ascribed to the advent of microsurgery in the late 1960s. This is not completely correct, because besides the improvement in instrumentation and lighting, there were also increased understanding of peripheral nerve anatomy, physiology, and pathophysiology; improvements in making the diagnosis and implementing child anesthesia; and development of brachial plexus surgery in adults. All of these

Fig. 9 James Warren Sever, an orthopedic surgeon at Boston Children Hospital, who published a follow-up report on 1100 cases of neonatal brachial plexus palsy in 1925 and recommended against operating on the brachial plexus itself

Fig. 10 Alain Gilbert, a French hand surgeon, who in 1977 operated his first case using microsurgical technique and revived interest in reconstructive brachial plexus surgery for neonatal brachial plexus palsy

contributed to the renewed interest in operative management in the 1970s.

World-renowned surgeons contributed to the revolutionary change in brachial plexus surgery. Herbert Seddon in the UK, Hanno Millesi in Austria, and Algimantas Narakas in Switzerland should be commended for their work in adults, and Alain Gilbert (Fig. 10) and his associates in France deserve credit for reviving interest in reconstructive surgery for neonatal brachial plexus palsy. Gilbert operated his first case of this condition in October 1977. The patient was a 4-year-old girl with very severe sequelae of complete paralysis. The brachial plexus was repaired by means of grafts, but even though several muscles of the upper extremity became reinnervated, the patient was unable to use them [16]. Subsequently, and up to 2018 (when he stopped counting), Gilbert performed 1823 brachial plexus surgical repair in neonatal brachial plexus palsy cases. Currently, he operates about 40 cases per year (Alain Gilbert, 2019—personal communication) and probably has the world's largest operative experience of this condition.

In the 1980s, thanks to initiatives from surgeons in many different countries, there was a revival of interest in surgical repair that consolidated this type of treatment as the standard indication in cases of neonatal brachial plexus palsy without spontaneous recovery [1, 3, 12, 15, 17, 18, 21, 29–33, 42, 44, 47].

The increasing understanding of peripheral nerve injuries in cases of neonatal brachial plexus palsy has led to great advances in the magnitude of functional recovery after surgical treatment. Currently, management in these cases focuses on early diagnosis and on surgical intervention when indicated.

Thanks to the pioneering spirit of the physicians listed in this article and the continuous improvements in anesthetic and operative techniques, surgical treatment today can offer the hope of effective functional recovery from this challenging injury.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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