

Obstetric brachial plexus palsy: reviewing the literature comparing the results of primary versus secondary surgery

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Abstract Obstetric brachial plexus injuries (OBPP) are a relatively common stretch injury of the brachial plexus that occurs during delivery. Roughly 30 % of patients will not recover completely and will need a surgical repair. Two main treatment strategies have been used: primary surgery, consisting in exploring and reconstructing the affected portions of the brachial plexus within the first few months of the patient's life, and secondary procedures that include tendon or muscle transfers, osteotomies, and other orthopedic techniques. Secondary procedures can be done as the only surgical treatment of OBPP or after primary surgery, in order to minimize any residual deficits. Two things are crucial to achieving a good outcome: (1) the appropriate selection of patients, to separate those who will spontaneously recover from those who will recover only partially or not at all; and (2) a good surgical technique. The objective of the present review is to assess the published literature concerning certain controversial issues in OBPP, especially in terms of the true current state of primary and secondary procedures, their results, and the respective roles each plays in modern-day treatment of this complex pathology. Considerable published evidence compiled over decades of surgical experience favors primary nerve surgery as the initial therapeutic step in patients who do not recover spontaneously, followed by secondary surgeries for further functional improvement. As described in this review, the results of such treatment can greatly ameliorate function in affected limbs. For best results, multi-disciplinary teams should treat these patients.

Keywords Obstetric brachial plexus palsy, Graft repair, Primary surgery, Secondary surgery, Nerve transfers

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Introduction

Obstetric brachial plexus injuries (OBPP) are a relatively common (2.9 per 1000 live births) [1] stretch injury of the brachial plexus that occurs during delivery. Even though a high percentage of these injuries achieve complete recovery spontaneously, this does not occur in roughly 30 % of patients [2], affecting some individuals for the rest of their lives, adversely compromising both movement and sensory function in the involved upper limb. Classically, two main treatment strategies have been used to treat these unfortunate newborns. The first and oldest approach has been to wait a number of years to see if and what degree of spontaneous recovery occurs, later performing different so-called secondary procedures that include tendon or muscle transfers, osteotomies, and other orthopedic techniques. The other newer strategy has been called 'primary surgery.' It consists of exploring and reconstructing the affected portions of the brachial plexus within the first few months of the patient's life, well before complete spontaneous recovery could ever happen. After primary surgery, secondary procedures can then be used to minimize any residual deficits.

In truth, primary surgery is not all that new; the first reports of this surgery being performed published in 1903 by Kennedy [2, 3]. However, using surgical microscopes and modern microsurgical techniques have rendered this approach to treatment much more mainstream. Alain Gilbert [4] was one of the first pioneers to report a series of

well-studied patients in whom he performed primary surgery with exceptionally good results, his patients recovering not only shoulder and elbow movements but also some hand function as well. Primary surgery generally consists of (1) sectioning the lesion—which generally is a neuroma-in-continuity—and replacing it with an autologous graft, (2) performing a nerve transfer, (3) doing a neurolysis (liberation of the nerve tissue from adhesions and fibrosis), or (4) all techniques or two of them combined (Fig. 1). Gilbert, among few others, worked hard to convince his initially reluctant colleagues of the advantages of this approach. Secondary surgeries could then theoretically be re-served only for those patients who fail to recover some movement after primary surgery, or for late referrals to whom primary surgery was not offered during the first few months of life. One of the obvious problems with deciding whether or not to perform primary surgery in newborns with OBPP is that if we section the lesion or perform reconstruction using grafts or nerve transfers, we lose the potential for spontaneous recovery, at least in the region of the plexus that we are sectioning. This makes two things crucial to achieving a good outcome:

- (1) the appropriate selection of patients, to separate those who will spontaneously recover from those who will recover only partially or not at all; and (2) a good surgical technique that ensures that the new nerve connections are performed properly to allow for the growing axons to successfully trans-verse the nerve union site.

The objective of the present review is to assess the published literature concerning certain controversial issues in OBPP, especially in terms of the true current state of primary and secondary procedures, their results, and the respective roles each play in modern-day treatment of this complex pathology.

Differences between adult and obstetric brachial plexus injuries

Traumatic brachial plexus injuries in adults are generally associated with high-energy accidents delivered over a few milliseconds, resulting in a higher proportion of root avulsions and severe stretch injuries, both pre- and post-ganglionic. Such injuries most commonly occur in the context of motor vehicle accidents. Their obstetrical counterpart has a completely different mechanism of injury: the stretching of roots and trunks involves a lesser degree of energy transfer over a much longer period of time (minutes or even hours; for as long as shoulder dystocia lasts during labor). Therefore, instead of the root avulsions and complete post-ganglionic sections of roots and trunks seen in adults, the typical lesion in infants is a neuroma-in-continuity, incomplete stretching that allows for some axon fibers to grow through and reach distal targets [5, 6] (Fig. 2). However, even though the aforementioned scenario is the one most frequently found, a mixed pattern of injury is not uncommon, as root avulsions or complete stretch lesions are seen in infants, and neuromas-in-continuity can also be observed in adults.

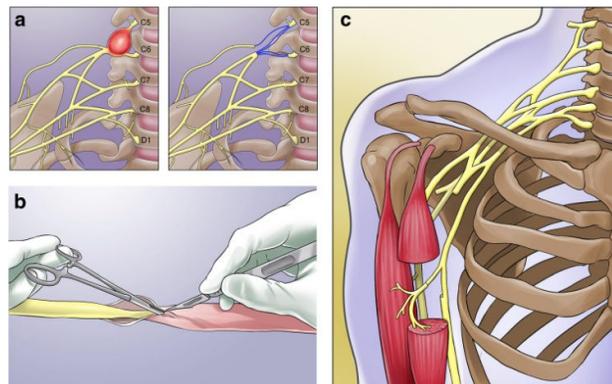


Fig. 1 a An in-continuity neuroma of the upper trunk of the brachial plexus is seen on the left. On the right, the neuroma was resected and cable grafts connect non-avulsed C5 and C6 grafts to the anterior and posterior divisions of the upper trunk and to the suprascapular nerve. b A neurolysis is a liberation of a nerve from external or internal compressive fibrotic tissue. c Example of nerve transfer, in this case a partial ulnar to biceps branch of the musculocutaneous (Oberlin's technique)



Fig. 2 The effects of trauma on the roots. Above: preganglionic rupture (avulsion), typical in high kinetic trauma that occurs in adults after a motorcycle accident. Center: postganglionic rupture. This root is available for grafting. Bottom: an incontinuity neuroma that allows the growing axons to pass in an alleatory way through it. This lesion is typical of obstetric brachial plexus palsies

With a neuroma-in-continuity, the growing pattern is not normal, with many axons innervating both agonist and antagonist muscles synchronically, or with the strength of innervation inadequate to invoke useful movement. The different pathophysiology observed in newborns is the main reason that the treatment of obstetric brachial plexus injuries must be understood in a completely different way than in adults. The presence of a neuroma-in-continuity guarantees two things:

(1) at least one root is not avulsed; otherwise, there would be no way to see a growing axon (i.e., with an upper trunk neuroma, at least C5, C6, or both roots must be in continuity); and (2) many patients recover spontaneously via the growth of severed axons through the neuroma-in-continuity. This is the reason why—whereas in adults the first objective of surgery is to connect proximal roots with distal targets that have no other way to connect to one other—in babies, the main goal of surgery is to resect the neuroma and orientate the growing axons toward a determined target, rather than allowing spontaneous regeneration to occur (Fig. 3).

If we do resect a neuroma and replace it with grafts, we have to ensure that the result will be better than what spontaneous recovery would provide. Here is where the biggest obstacle arises with primary surgery for obstetric brachial plexus injuries: identifying appropriate candidates for surgery and the correct time to perform the procedure, differentiating between those patients who will recover spontaneously to an adequate degree from those who will benefit from neuroma resection and reconstruction with grafts.

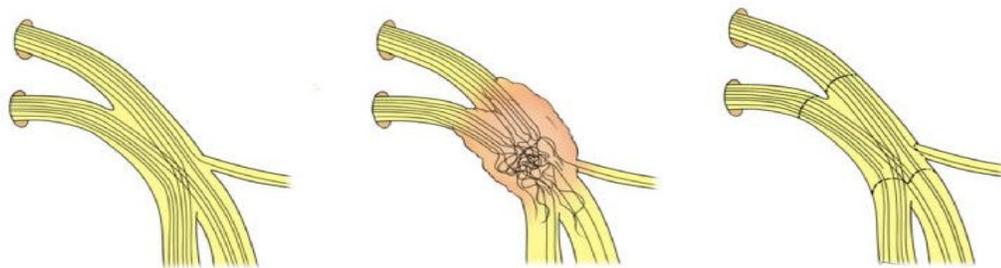


Fig. 3 The concept of replacing a neuroma with interposed sural nerve grafts. Left: normal upper trunk, with fascicles inside. Center: an incontinuity neuroma allows a disordered passing of the growing axons: few of them will reach the distal targets. Right: the neuroma was replaced with grafts, leaving a path to allow the growing axons to reach the distal targets in an organized manner

Physical examination: can OBPP be considered a single entity?

Newborns suffering traction injuries to their brachial plexus during delivery share a common mechanism of injury, but can exhibit a broad and highly varied constellation of symptoms and signs, many of which are far from being stable. In fact, dynamic changes are common from the time of delivery through the next few months of life. Classically, three different clinical syndromes have been described:

1. Upper-type obstetric brachial plexus palsy (aka C5-C6, upper trunk palsy, Duchenne-Erb syndrome) is characterized by lost or reduced shoulder abduction and elbow flexion, while hand function is preserved (Fig. 4a).
2. Complete obstetric brachial plexus palsy (C5-D1 palsy) paralyzes the shoulder, elbow, and hand. Sometimes, Bernard-Horner syndrome is associated, which some argue is an indirect sign that lower roots (C8-D1) have been avulsed, resulting in sympathetic plexus elongation. Some challenge this interpretation, however, having established that more widespread sympathetic innervation—including the C7 root—may exist in infants [7].
3. Lower-type obstetric brachial plexus palsy (aka C8-D1, lower trunk palsy, Dejerine-Klumpke syndrome) is rarely seen. Like complete palsy, it is also associated with Horner's syndrome, and characterized by complete hand and wrist palsy (Fig. 4b).

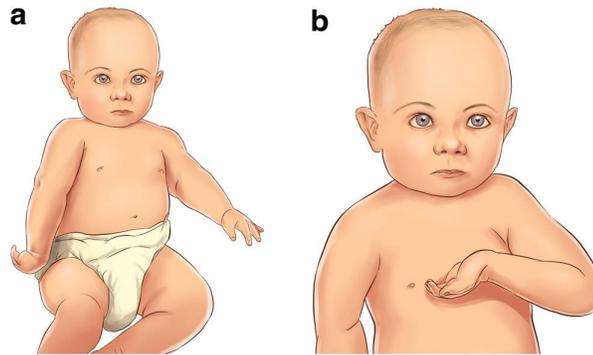


Fig. 4 a Duchenne-Erb (upper trunk) palsy. Shoulder abduction and elbow flexion are paralyzed. b Dejerine-Klumpke (lower trunk) palsy. The shoulder and elbow are preserved, but the hand is paralyzed

A range of mixed syndromes exist with variable components of the aforementioned three classic presentations. For example, the C5-C6-C7 syndrome is associated with shoulder abduction palsy, complete elbow palsy, and wrist extension palsy. Meanwhile, involvement of C5 through C8 adds some degree of impairment of hand function.

Given this constellation of presentations, we do not consider OBPP a single entity. Accordingly, treatment strategies differ considerably. For example, with complete brachial plexus palsy and an accompanying Horner's syndrome, early surgery is considered mandatory, usually when the patient is roughly 3 months old. On the other hand, with upper-type injuries lacking root avulsions, many surgeons prefer to wait until between the sixth and ninth month of life to attempt reconstruction.

The natural history of OBPP

To determine how useful primary surgery is, one must first identify the natural history of OBPP, keeping in mind that most affected infants will recover spontaneously. At present, the best review of this topic, for which 1020 papers were examined, was published by Pondaag et al. [2]. They established that the ideal study to determine the natural history of OBPP must be (1) prospective, (2) based on a closed

population and not on individual cases derived from different places toward a certain surgeon, (3) have a minimum follow-up of 3 years, and (4) use an appropriate scale to accurately assess outcomes. Incredibly, not even 1 of the 1020 articles they assessed fulfilled even 3 of these four criteria. Only papers satisfying one or two of the criteria were found, revealing how the actual natural history of OBPP remains unclear. Nevertheless, according to the authors, it became clear to them that at least 20 to 30 % of infants will have some permanent impairment related to their OBPP, prompting some kind of treatment for this subgroup of patients.

Comparing primary and secondary surgery for OBPP

Pondaag et al. [2] also systematically reviewed published series in an attempt to compare the evidence supporting primary versus secondary surgery, but were able to discern only nine papers that compared the two treatments, eight of them presenting evidence-based medicine (EBM) level 4 evidence and one level 5. Unfortunately, no pooling of data was possible because of insurmountable differences in patient selection, the indications for surgery, the scales used to measure neurological deficits, and other methodological issues.

The first reported study comparing conservative treatment versus primary surgery was mentioned before, and published by Gilbert and Tassin in 1984 [4, 8]. In this study, 44 non-operated upon children were compared against 38 who had undergone brachial plexus reconstruction. Unfortunately, though both patient groups came from the same city, they came from different institutions, which raise questions for issues like selection bias and differences in patient evaluations. Even though no statistical analysis was performed, it was clear that the final results of those patients who were operated upon were much better than among those who were not. Another crucial finding of this study that strongly influenced the management of OBPP for the next few decades was that biceps recovery before 2 months of age predicted spontaneous recovery, rendering surgery unnecessary. On the other hand, if biceps function had not returned by the time the infant reached 3 months old, then primary reconstruction should be performed. In 1988, Gilbert et al. [9] presented 178 cases with a minimal follow up of 2 years and stressed their initial findings, which were that 80 % of patients with a C5-C6 lesion achieved good to excellent results with surgery, versus 0 % of those treated conservatively, while 45 % of those with C5-C6-C7 injuries experienced similar results. Moreover, when primary reconstructive surgery and conservative treatment were compared with respect to the outcomes of later secondary procedures, the results of the former were better because these patients had more recovered muscles that could be transferred, and they lacked co-contractions between antagonist muscles. Thereafter, Haerle and Gilbert [10] published their results on 73 complete OBPP cases with a mean follow-up of 6.4 years. Although the results for elbow and shoulder function were not as good as with C5-C6 palsies (a total of 123 secondary procedures, mainly in the shoulders, were needed to improve function), the results for hand reinnervation were encouraging, probably because, with total palsies, the few available roots or extraplexual donors were directed to this target via the lower trunk. According to the authors, a useful hand was attained in 75 % of patients, though it took more than 3 years to arrive at such a favorable outcome. Interpreting their surgical results, these authors again argued for the usefulness of early exploration and repair of the obstetric plexus. The secondary procedures performed for the shoulders yielded good or average results in 77 %. Recovery of elbow flexion was very good, with 81 % of patients exhibiting good active flexion at 8 years of follow-up. The authors also observed an interesting phenomenon—their shoulder results tended to be less favorable in adolescents. They reasoned that these patients, their families, and their treatment teams paid less attention to rehabilitation than when they were younger children.

Comparing 22 patients who underwent primary reconstruction and 48 who did not, Boome and Kaye [11] arrived at similar conclusions as Gilbert: that nerve grafting yielded the best potential for recovery if biceps function was not recovered by 3 months. However, once again, no statistical analysis was performed.

A group led by Clarke published four more methodologically robust studies; the first examining the natural history of OBPP, the second, third, and fourth analyzing the effectiveness of pure neurolysis versus grafting, and the latter patients having a minimum follow-up of 4 years [12–15]. They used a newly-developed scale, the Active Movement Scale, as their primary outcome. The authors concluded that the results achieved via neurolysis were no better than the natural history of OBPP, while grafting results were better than what was observed in either other group.

Waters et al. [16] reached similar conclusions, except that they deemed grafting clearly better than conservative treatment plus secondary surgery only when patients had not re-covered biceps function by six (as opposed to three) months, and that shoulder external rotation results were generally poorer than for abduction, no matter what treatment approach was chosen.

Results published by Al-Qattan [14, 17], Xu et al. [18], Badr et al. [19], and Gu [18] were similar to those of the aforementioned studies, but selection bias or inconsistency in the scales used to measure outcomes makes it difficult to compare these series with the rest.

Cheng et al. [20] also concluded that accessory and phrenic nerve transfers yield excellent results, in terms of both shoulder and elbow recovery. However, it is crucial to state clearly that most of these investigators [21–23] considered use of the phrenic nerve for transfer contraindicated in children less than 3 years of age, because these patients might develop respiratory problems.

Investigators in Sweden [24] analyzed a closed population of 247 patients with prolonged follow-up (5 years) and concluded that shoulder results were much better with C5-C6 palsies when primary surgery was performed. Nevertheless, these authors precluded waiting more than 6 months for biceps recovery.

The role of primary surgery restoring function in the hand is more controversial than for the shoulder or elbow. In a retrospective study in which results were gleaned from 11 different surgeons across 9 institutions—a fact that enhances the results' generalizability but raises other issues like the consistency of evaluations and treatment—Kirjavainen et al. [25] evaluated 105 patients with hand palsies post reconstruction for a mean 13.4 years and concluded that their results support lower root reconstructive surgery, in that patients undergoing hand reconstruction with grafts had better outcomes than those who did not.

After analyzing the series described above that compared primary versus secondary treatment, we agree with Pondaag et al. [2] that, albeit the scientific evidence supporting reconstructive primary surgery for OBPP is far from ideal, the general consensus is that this approach is indicated in patients in whom the anticipated natural history is of no or inadequate spontaneous recovery, as in those with little to no return of biceps function by 3 months.

Given that the decision to perform primary surgery should be made well before complete spontaneous recovery has taken place, what is then crucial is to be able to predict which patients will not recover spontaneously. The next section examines this issue more closely.

Importance of optimizing the timing of surgery

No single algorithm is adequate to manage the extremely complex pathology that exists with brachial plexus injuries. As such, the decision regarding whether or not to proceed to surgery must be done on a case-by-case basis. The timing of intervention is overwhelmingly important in peripheral nerve surgery: if a procedure is performed too late, muscle atrophy appears so that, no matter how many axons reach the distal muscle target, functional (motor) recovery will be limited. As stated before, due to the unique etiology of OBPP, where a neuroma-in-continuity is frequently seen, the growing axons can bypass the lesion and reinnervate some target muscles, even though this growth is not straight. The problem is that this reinnervation is often either incomplete or directly deleterious, because agonistic and antagonistic muscles can be innervated via the same fascicles. This is a critical problem in OBPP patients.

As mentioned earlier, Gilbert and Tassin's classical rule—if the biceps does not spontaneously reinnervate by 3 months, surgery is indicated—has been challenged. For example, Michelow et al. [12] claimed that this

rule would lead to 12 % of patients undergoing surgery that was unnecessary. Therefore, a wider window of 3 to 9 months of post-natal follow-up has been adopted by many clinicians [26].

With C5-C6 injuries, conservative treatment is generally preferred. In this scenario, good quality myelograms, both with MRI and CT, may help to avoid unnecessary delays if a C5 or C6 avulsion is observed. If after this observational win-dow no spontaneous recovery is seen, there should be no delay proceeding to primary surgery.

On the other hand, if we have to deal with a complete palsy and associated Horner's syndrome, earlier surgery is generally preferred because no complete spontaneous recovery is expected.

Some evidence exists to suggest that OBPP patients might have a relatively longer window of opportunity for primary surgery, and that certain forms of late surgery could yield good results. Badr et al. [19] identified a subgroup of patients with good recovery of the elbow 9 months after birth, but poor shoulder function, who then benefited from primary surgery. El-Gammal et al. also analyzed 19 patients (14 with complete and 5 upper-type lesions) who they operated upon even later (mean age at surgery = 41 months, range 18 to 48) and follow-ed post-operatively for a mean of 2 years. The patients experienced marked recovery of elbow flexion (mean increase from 2.7° pre-operatively to 91.8° at last follow-up), elbow extension (from 10 to 56.7°), and hand function, especially related to sensation [27].

Comments on surgical strategies for primary surgery

As depicted in Fig. 1, three basic microsurgical techniques are employed in primary surgery: neuroma resection and reconstruction with interposed grafts, neurolysis, and nerve transfers. Results with the first technique have been reviewed previously in this paper, while consideration of the other two techniques follows.

Neurolysis

In a robustly designed and very influential paper already mentioned above, the group led by Clarke in Toronto [15] determined that neurolysis alone produces no beneficial effects in children with OBPP, whereas sectioning of the neuroma and reconstructing with grafts comprise the gold standard for obtaining good results. In other words, according to these authors, the natural history of an OBPP does not differ from the course of patients who undergo isolated brachial plexus neurolysis. As their primary outcome, these authors used the Active Movement Scale (AMS)—designed to evaluate 15 distinct movements of the upper limb—to retrospectively assess 108 patients with a mean follow-up of 4 years. Their conclusion is logical, because a stretch injury that causes a neuroma-in-continuity or a root avulsion should not benefit from neurolysis. Surgery was indicated if the baby had an AMS score of 3.5 or less, a Horner's sign, or a negative "cook-ie test" at 9 months (positive hand-to-mouth test). Mean age at surgery was 7.9 months. As just mentioned, the results were consistently better with reconstruction using grafts than neurolysis alone. Moreover, 56.3 % of the patients who underwent neurolysis alone ultimately required a secondary procedure, versus just 29.4 % of those who received graft reconstruction.

Nerve transfers

Pure nerve transfers in OBPP patients are uncommon, mainly because there is almost always at least one available root for reconstruction with grafts. Nevertheless, some interesting series in which isolated nerve transfers were assessed should be mentioned.

Ladak et al. [28] reviewed ten patients with upper-type lesions in whom three nerve transfers were performed (spinal accessory to suprascapular nerve, ulnar nerve fascicle to bi-ceps branch, and triceps branch to axillary nerve), and observed statistically significant improvements versus baseline for both elbow flexion and shoulder abduction. That a plexus approach was not adopted, so that the likely neuroma-in-continuity

was not resected, may have favorably biased re-sults, however, since some spontaneous recovery could also have occurred.

El-Gammal et al. [29] reviewed 46 babies (mean age at surgery 14 months) with intercostal nerve transfers to 12 different targets, albeit mainly to the musculocutaneous nerve (n = 31) and observed an overall rate of 'success' of 76 % (93 % for elbow flexion).

Following publication of the original report by Oberlin et al.—wherein using ulnar nerve fascicles for the biceps and brachialis branches of the musculocutaneous nerve [30] was suggested to reconstruct elbow flexion in partial palsies in adults—the treatment protocol for partial brachial plexus injuries in adults changed completely over the course of the following two decades. The greater than 90 % success rate of this novel approach modified the earlier tendency to perform reconstruction using grafts gleaned from nerve roots [31].

As might be expected, OBPP also were influenced by this new treatment tendency. Noaman et al. [32] retrospectively evaluated their results in seven children, in whom M3 level strength (a 'good result') was achieved in five.

In a more recent series involving 18 patients [33], good to excellent results for biceps contraction were obtained in 14 (77.8 %). The pre-operative Al-Qattan scale score and an absence of osseous alteration in X-rays of the hand were maintained at final follow-up. Consequently, it was concluded that no permanent damage to the developing hand resulted from extracting ulnar nerve fascicles for the biceps.

In a multicenter study of 31 patients undergoing nerve transfers to reanimate elbow flexion, Little et al. found that 87 % of their patients obtained functional elbow flexion, and 77 % complete return of function [34]. The authors included five patients in whom they performed double transfers to re-innervate the biceps and brachioradialis [26, 35]. Recovery of supination recovery was better in patients with an avulsion than in those without [34].

Even given the initially documented success of Oberlin's technique in newborns, it is important to note that classical reconstruction with grafts from non-avulsed roots also generates excellent results in this patient population. Therefore, if no further evidence appears to suggest its superiority, it is generally recommended that Oberlin's ulnar nerve transfer for upper-type obstetric brachial plexus palsy be reserved for those patients with (1) avulsion of the C5 and C6 nerve roots, (2) a late presentation with good recovery of shoulder function, (3) an upper trunk neuroma-in-continuity with good intra-operative nerve conduction to the shoulder muscles, consistent with good shoulder function pre-operatively but no biceps movement (dissociated recovery), and (4) failure of graft reconstruction for elbow flexion.

Triceps branch nerve transfers to the axillary nerve are another proven technique in adults. Zuckerman et al. assessed seven OBPP patients with upper palsies in whom they performed this procedure in seven patients, and noted good results for both shoulder abduction and deltoid muscle strength in all of them.

Terzis et al. [36] obtained better results using the suprascapular versus the axillary nerve as a target for shoulder reanimation during primary surgery. However, the authors did not compare the two approaches directly. Moreover, the patient sample was heterogeneous and many patients were admitted for secondary procedures later. This being said, both of these nerves are generally adequate targets when the objective is to enhance shoulder function.

Another popular nerve transfer for elbow flexion in children is the pectoral to biceps branch technique. In 2003, Blauw et al. published their results with pectoral to the musculocutaneous nerve transfers in 25 children, and noted excellent results in 17 and fair results in 5, while 2 patients, who subsequently underwent a Steindler flexorplasty, were deemed complete failures. Leiden et al. [37] also consider this technique to be trustworthy, in terms of generating favorable ultimate outcomes.

Finally, contralateral C7 transfers are a controversial technique, especially because it seems that, at least in adults, patients must move their healthy limb to move their re-innervated limb, due to limitations in brain plasticity in adults. In 15 patients with upper-type palsies, Lin et al. [38] noted marked dependence between the 2 limbs, though in some patients this did not appear to result in any significant impairment.

Reconstruction with grafts plus nerve transfers

The overwhelming majority of primary reconstructions fall within the category of grafts plus nerve transfers. Even after an appropriate neuroma resection and reconstruction with grafts, the option of enhancing results using, for instance, a spinal accessory to suprascapular nerve transfer is frequently adopted.

Chuang et al. [39] retrospectively analyzed 78 OBPP patients divided into 2 groups: those with more severe lesions, associated with complete palsies, with more than 80 % having avulsion of two roots (44 infants), and those with less severe injuries, consisting of upper-type palsies with no root avulsions (n = 34). For the former, the surgeons generally utilized spinal accessory to suprascapular nerve transfers for the shoulder, intercostal to musculocutaneous nerve transfers to restore elbow flexion, and other available roots for the hand. They achieved abduction greater than 90° and external rotation greater than 60° in 86 % of patients; elbow flexion (positive hand-to-mouth test) was adequately restored in 59 %; and good results were achieved in the hand in 63 %. As anticipated, outcomes in the non-avulsed group were much better than in those with avulsions, averaging 132° (80–180°) of abduction, 67 % of external rotation (range 50–90°), and at least M3 elbow flexion strength in 100 % of cases. O'Brien et al. [40], in 52 patients, obtained only slightly worse results with similar surgical techniques in somewhat older patients (mean age at surgery 4.9 months in Chuang's series versus 9.8 months in O'Brien's).

Tse et al. [41] compared their results obtained for shoulder external rotation, but not shoulder abduction, after reconstruction of the suprascapular nerve using grafts from C5 or the spinal accessory nerve as the source for nerve transfer. This study

failed to demonstrate any statistically significant differences between the two groups of patients, though the nerve transfer group had more complete palsies and avulsions at baseline, and tended to undergo surgery at a younger age.

Secondary procedures: potentially improving early results

Secondary procedures are those surgeries involving tendon transfers, arthrodesis, or osteotomies. They can be used in two different scenarios: (1) in patients who, after primary surgery, have reached a certain level of recovery, but still have some deficits that are considered treatable with a secondary procedure; and (2) in patients who did not undergo primary surgery, have some level of spontaneous recovery, but also still have deficits. As stated earlier in this paper, primary surgery not only optimizes the likelihood of recovering lost upper limb function but also innervates new muscles that can then be used for secondary procedures that might enhance function further. Therefore, the emergence of primary surgery does not indicate the end of secondary surgeries, but rather the birth of a number of additional surgical options.

Most frequently, even after highly successful primary surgery for shoulder and elbow restoration, there is persistent weakness with some movements, thereby limiting the patient's use of the upper limb. Shoulder external rotation is one example. Patients with this deficit are unable or limited in their ability to perform self-care activities like grooming, feeding, and washing themselves. Selecting the right surgical procedure could greatly increase external rotation. For didactic reasons, we will divide the secondary procedures into three categories, based upon the anatomic targets: (1) shoulder, (2) elbow, and (3) hand.

Secondary procedures for shoulder restoration

Strecker et al. [42], in an older series, use the classical Sever-L'Episcopo procedure—transferring the latissimus dorsi and teres major tendons to the external aspect of the humerus in an attempt to enhance external rotation—and achieved a mean improvement of 80° in 16 patients. Three transient and one permanent axillary palsy were observed by stretching the nerve at the quadrangular space. This surgery has been largely abandoned due to the resultant loss of internal rotation and abduction.

Another possible approach to improving external rotation of the shoulder is the Hoffer procedure, a variation of the Sever-L'Episcopo procedure with better results both function-ally and cosmetically (Fig. 5). Phipps and Hoffer [43] used this procedure on 56 patients with Erb's palsy and managed to follow 35 of them for more than 2 years. Post-operative active external rotation averaged 31° with only two recurrent internal rotation contractures. Post-operative strength in external rota-tion increased in 29 of the 35 patients, with abduction strength increasing one or more grades in 13 of 35. Murabit et al. [44] studied 18 patients who underwent this procedure and im-proved significantly with no increased deficits: younger age and better shoulder movement were positive predictors in the studied population, something that Gilbert et al. [45] also noted. Edwards et al. reviewed ten patients using the same technique and also obtained excellent results: active shoulder abduction improved from a mean 72° pre-operatively to 136° post-operatively, while post-operative active shoulder external rotation averaged 64° [46]. Ozben et al. [47] also obtained good results transferring the latissimus dorsi and teres major in the absence of any subscapularis tendon release for pallia-tive shoulder motion, and their improvements were statistical-ly significant for both abduction and external rotation.

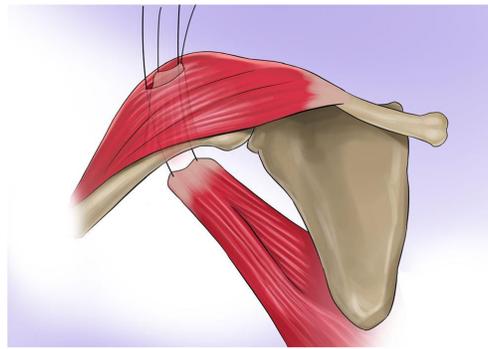


Fig. 5 This diagram shows the Hoffer procedure, a transfer of the latissimus dorsi and teres major muscles to the rotator cuff posteriorly through a transaxillary approach. This procedure is designed primarily to improve external rotation of the shoulder

In older children, a rotational osteotomy of the proximal humerus is a valid procedure to improve external rotation. Kirkos et al. [48] reviewed 22 children, averaging 10 years old, in whom they achieved a mean of 27° of active external rotation. According to these authors, to undergo this proce-dure, patients should be older than 2 years, and should have a functional elbow and hand, no dysplastic glenoid, acceptable passive shoulder range of motion, and obviously a functional latissimus dorsi and teres major muscle. Obesity may be a relative contraindication to this procedure.

Gilbert's group [49] evaluated their results after transfer-ring the latissimus dorsi muscle to improve abduction and external rotation as a secondary procedure in 203 children, and observed that patients with C5-C6 palsies experienced the best results for both abduction and external rotation, while C5-C7 and complete palsy patients only experienced im-provements in external rotation.

Terzis et al. [50] analyzed 67 patients (68 limbs) who underwent a total of 197 secondary procedures for shoulder reconstruction, half of these patients having undergone previ-ous primary surgery. Mean follow-up was 7.5 years. Using Mallet's scale—a measurement instrument designed specifi-cally to evaluate the shoulder in patients with OBPP—a score of 18 or more was considered a good result. Numerous differ-ent procedures were employed, including muscle or tendon transfers, tendon releases, arthroplasties, and osteotomies. The mean Mallet's score increased from 13.4 pre-operatively to 20.7 post-operatively, and abduction improved by a mean of 69°. Having an upper-type palsy and undergoing surgery prior to 4 years of age were two predictors of a favorable outcome. However, no statistically significant differences were observed in the outcomes of patients who had had pri-mary surgery versus those who had not.

Secondary procedures for elbow recovery

Steindler flexorplasty is a procedure that is very commonly used to reconstruct elbow flexion (Fig. 6a, b). Again, it was the group of Gilbert et al. [51] who studied 26 cases—all of them having previously undergone primary surgery and plexus reconstruction. Elbow flexion in these patients improved from a mean 14° pre-operatively to 97° post-operatively. One strong predictor of a good response was strong wrist extension pre-operatively.

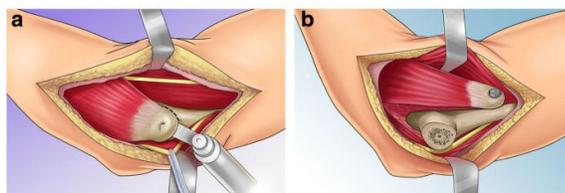


Fig. 6 Steindler procedure. a Flexor-pronator muscles arising from medial epicondyle are detached together with a bone cuff. b They are transposed to a more proximal point on the humerus and fixed with a screw, so that their moment for elbow flexion is increased enough to permit active elbow flexion

Al Quattan [52] reported on his experience with Steindler's flexorplasty in nine OBPP patients; and, after a mean follow-up of 5 years, eight patients had a good outcome with mean active elbow flexion against resistance of 110°. The outcome in the remaining patient was poor. The authors maintained that the following selection criteria—good (M4) grip strength with the fingers, good wrist and elbow extension, and at least M2 strength on elbow flexion—predict a good outcome. Similarly, Tonino et al. [53] obtained good functional results with the Steindler procedure in 23 of 26 patients (88 %).

Another possibility for elbow flexion restoration (in patients who present late for surgery) is to transfer a free functioning gracilis muscle from the thigh. This surgery is more complex, incorporating both a nerve and a vascular anastomosis, and its rate of success is lower than with primary surgery. Nonetheless, it is one further option for late referral cases, patients who have failed other procedures, and patients for whom the Steindler procedure is contraindicated. El-Gammal [54] presented 18 cases in which there were significant improvements in both elbow flexion range and strength. Three of the 18 (17 %) also experienced some partial improvement in hand function.

Secondary procedures for hand and wrist restoration

Surgically restoring hand function in OBPP patients is a challenge, both for primary and secondary procedures, with results only palliative in most cases. In one series of 56 patients in which tendon transfers were performed to restore absent wrist extension, Duclos et al. achieved a positive result in 75 %, with 42.5 % experiencing active and 32.5 % static wrist extension [55]. Patient age did not appear to influence their results.

Ruchelsman et al. [56] studied 21 patients, at an average age of 5.5 years, on whom they operated to obtain active wrist extension, and achieved active extension in 66 %, even better than the results noted by Duclos et al.

In patients with a complete brachial plexus palsy, after primary reconstruction of the lower trunk for hand function, only 26 further procedures were deemed possible to restore further hand function in Haerle's series [10]. These included transfers to restore finger extension in 13 patients and to restore intrinsic function in 3, as well as 5 joint releases and 4 tenodeses. As mentioned previously, at 8 years of follow-up, 76 % of the patients with complete brachial plexus palsies had a useful hand via a combination of primary and secondary procedures.

Bertelli et al. [57] transferred a functioning brachialis muscle to forearm extensors, resulting in good recovery of wrist extension and pronation. They transferred this muscle to the pronator teres, the extensor carpi radialis brevis, or the extensor carpi radialis longus, obtaining an average of 20° active motion recovery in wrist extension and 14° in pronation.

Another possibility for hand function reconstruction is a free functional muscle transfer using the gracilis muscle. Duclos et al. [58] performed this operation on four children with complete hand palsy who had undergone primary surgery with no reinnervation of the hand, and achieved good finger flexion in three of them. Meanwhile, Zuker and Manktelow [59] reported on their experiences with 30 forearm reconstructions using the same technique, and more than half of their patients obtained good wrist flexion.

To improve supination, Amrani et al. [60] transferred the pronator teres in 14 patients with either a C5-C6 or C5-C6-C7 palsy, at a mean age at time of surgery of 7.6 years. Active supination was improved consistently, increasing from a mean 3.6° pre-operatively to 77° post-operatively.

Conclusions

The natural history of OBPP, and the optimum choice and timing of treatment still has to be empirically verified. Nevertheless, considerable published evidence compiled over decades of surgical experience favors primary nerve surgery as the initial therapeutic step in patients who do not recover spontaneously, followed by secondary surgeries for further functional improvement. As described in this review, the results of such treatment can greatly ameliorate function in affected limbs. For best results, multi-disciplinary teams composed of clinicians, surgeons, physiotherapists, and occupational therapists should treat these patients. At many centers, the same surgeons perform primary and secondary procedures; meanwhile, at others, the procedures are performed by different surgeons on the same team. In both instances, the whole team must be well versed in all the procedures, and thoroughly discuss each therapeutic step when the time is appropriate. The permanent presence of a rehabilitation team is mandatory throughout the entire process. Given that the treatment of OBPP palsies can take two decades or even more, this complementary work is the best chance for recovery for those patients who suffer from these complex and disabling injuries.

Compliance with ethical standards

Ethics No ethical conflict and no financial support were present or received upon writing, editing, or sending the present work.

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

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