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Original article

Treatment of dystonia in extensor hallucis longus and digitorum muscles with neurotomy of the branches of the deep fibular nerve: Preliminary results

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ABSTRACT

Introduction: Dystonia in extensor hallucis and/or digitorum muscles can be observed in pyramidal and extrapyramidal lesions and results in pain in these toes, spontaneous or when walking, problems and discomfort when putting on shoes and socks, and cutaneous lesions on the toes. The objective of this study was to assess the efficacy and safety of deep fibular nerve neurotomy for the extensor hallucis longus (EHL) and/or the extensor digitorum longus (EDL) branches in the treatment of extension dystonia of the hallux and/or other toes.
Patients and methods: A deep fibular nerve neurotomy was performed in 20 patients ($n = 19$ for the EHL, $n = 6$ for the EDL). We retrospectively analyzed the treatment's efficacy and safety and assessed the patients' self-reported improvement and overall treatment satisfaction.
Results: Dystonia totally disappeared in 15 cases (75%); it persisted at a minimal level in the other patients. The patients reported a decrease in pain ($P < 0.01$) and fewer difficulties putting on shoes and socks ($P < 0.001$) and had a high median level of satisfaction (8.5/10). Adverse effects were rare and transient. The identification of the nerve branches was sometimes difficult.
Discussion: Deep fibular nerve neurotomy for the EHL and/or EDL branches seems to be an effective treatment for extension dystonia of the hallux and/or other toes and its consequences for the adult neurological patient. However, these encouraging preliminary results should be confirmed by prospective, longer-term studies.
Level of evidence: IV.

1. Introduction

Extension dystonia of the hallux and/or toes is a classical deformity [1], encountered in two distinct contexts: extrapyramidal deformity [2] and pyramidal lesions secondary to neonatal or acquired brain injury [1,3]. It is related to hypertonia of the extensor hallucis longus (EHL) and/or the extensor digitorum longus (EDL) muscles. These extension spasms can lead to troublesome symptoms such as spontaneous or induced pain (either on the dorsal side of the toe(s) or of their tendon, which, when under tension, can become painful), problems putting on shoes and socks, discomfort when wearing shoes, cutaneous lesions on the dorsal side of the hallux [3–7], as well as problems when walking related to the pain induced and to the instability that can occur at the end of the stance phase [3,5,6].

The hyperextension mechanism always involves the EHL and/or the EDL, which can be hypertonic, retracted, or both. In its isolated form, this deformity involves hyperextension of the metatarsophalangeal (MTP) and the interphalangeals (IPs). If it is associated with tension on the flexor system, it often presents as hyperextension of the MTP and flexion of the IPs. Hyperactivation of the EHL and/or the EDL first of all can be caused by true dystonia of these muscles. When it occurs in the swing phase, it can also be related to a tenodesis effect on the EHL and/or the EDL when the foot drops because of a deficit in the levator muscles. Finally, it can correspond to an attempt to compensate for weakness in the anterior

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tibialis anterior muscle by the extensor digitorum muscles, which are accessory dorsal flexors. In these three cases, the severity of the hyperextension can be aggravated by retraction of the muscles involved.

Over the past few years, botulinum toxin treatment (BT) has proved to be effective in treating dystonia of the EHL in extrapyramidal lesions such as after an acquired brain injury [3–5]. However, this treatment often requires high doses of BT [5], limiting its use in other hypertonic muscle groups, and necessitates repeated injections. Neuro-orthopaedic treatment aims to prevent these two pitfalls. Lengthening the EHL tendon at the muscle–tendon junction is a therapeutic possibility [8,9], all the more indicated in isolated hyperactivity of the extensor muscles when muscle retraction is present, even though to our knowledge the literature reports no specific data on neurological hyperextension of the toes concerning this technique, nor any description of EDL lengthening procedures. In our experience, toe tendon lengthening procedures tend to give inconsistent and transient results, and the degree of lengthening is often difficult to predict (and is therefore a source of under- or over-correction). As for other locations, neurotomies have been shown to be effective in treating muscular hypertonia associated with pyramidal syndrome [10]. We have developed a selective neurotomy technique of the motor branches of the deep fibular nerve (formerly referred to as the anterior tibial nerve) leading to the EHL and the EDL muscles.

In the present study, we analyzed the efficacy and safety of deep fibular nerve neurotomy in adult patients suffering from disabling extension dystonia of the hallus and/or the other toes.

2. Patients and methods

2.1. Patients

In this preliminary open multicenter study, we included patients presenting with dystonia in of the hallux and/or other toes, causing problems (pain, cutaneous lesions on the toes, problems putting on shoes and socks, discomfort when walking), for which surgical treatment had been recommended during a multidisciplinary consultation. This consultation, which grouped physicians in physical and rehabilitation medicine, orthopaedic surgeons, and a neurosurgeon, allowed us to analyze the impact of dystonia of the toes, to select the targets that should profit from neurotomy (EHL and/or the EDL), and obtain the patient's informed consent. We included patients whatever the source of the dystonia, excluding patients suffering from severe language or psychiatric disorders because they would have been unable to take part in a subsequent telephone interview. The study was conducted in compliance with the principles of the Declaration of Helsinki and the patients were requested to provide oral consent to participate in the study.

Deep fibular nerve neurotomy was performed in 20 patients between July 2011 and January 2013 (Table 1). Twelve (60%) were female, and the median age was 52 years (range, 27–64 years). They suffered from stroke (14 patients, 70%), traumatic brain injury (two patients, 10%), cerebral palsy (one patient), craniostenosis (one patient), primary generalized dystonia (one patient), and cerebrotendinous xanthomatosis (one patient). For acquired lesions, the median duration was 9.5 years (range, 2.2–23.1 years). Six patients (30%) had been treated with BT for dystonia of the EHL, with at least partial efficacy for five of them.

2.2. Treatment

Neurotomy for fibular nerve branches that innervate the EHL and/or the EDL was performed under general anesthesia, with the patient in the supine position. A vertical cutaneous incision on the anterolateral side of the leg provided access to the space separating the tibialis anterior medially and the EHL and EDL laterally, making it possible to first approach the deep fibular nerve trunk and to repair the motor branches for both muscles. There was always a common branch for the EHL and EDL, and a varying number of additional branches for each of the muscles (from one to three) [11]. Since the course of the nerve varies between patients, selective fascicular electrical neurostimulation was used to identify motor branches and avoid sensory branches. Once they had been identified, the muscular branches were resected from three-fifths to four-fifths over a distance of 1 cm [12], such that the muscle response to upstream electrical stimulation was clearly reduced.

The neurotomy targeted the EHL branches for 14 patients (70%), the EDL alone for one patient (5%), and both for five patients (25%) (Table 1). It was bilateral in two patients (for the EHL in one case and the EDL in the other). The median follow-up period was 9.5 months (range, 3–21 months). In most cases, the patients underwent other therapeutic procedures in the same surgical time: one tibial nerve neurotomy for the gastrocnemius, the soleus, and tibialis posterior was done in three patients (15%), tendon lengthening in 16 patients (80%), which in 11 (55%) involved the Achilles tendon, 11 the flexor hallucis longus, and 12 the flexor digitorum longus (tenotomy). Two patients (10%) underwent split tibialis anterior transfer, and three (15%) a subtalar arthrodesis.

2.3. Evaluations

A retrospective study was first conducted by comparing the pre- and postoperative consultation data (carried out between 3 and 6 months after the procedure). The location of the dystonia was recorded (hallux, other toes, or both), its severity (0 = absent, 1 = minimal, 2 = moderate, 3 = severe, based on its amplitude and the frequency with which it occurred), its triggering features (permanent or initiated when standing or walking), the problem(s) occurring in both passive functions (spontaneous pain in the dorsal side of the toes or the extensor tendons, problems and discomfort when putting on shoes and socks, toe calluses and ulcers) and when walking (pain). In addition, intra- and postoperative adverse events were systematically recorded.

In the second part of the study, all patients were contacted by telephone in May 2013 by two investigators who were not members of the surgical team (CR and EA) to assess the self-reported changes induced by the neurotomy. The patients were first requested to evaluate their main dystonia-induced complaint before surgery (among those listed above) and to score the overall impairment related to the dystonia when standing and walking before and after the procedure on a numeric scale (from 0 = no impairment to 10 = major impairment). In addition, they were asked to assess the changes over time in dystonia-induced impairment on the Global Assessment Scale (GAS) [13], which uses a Likert scale from −4 (major deterioration) through 0 (no change) to 4 (major improvement). Finally, the patients were requested to estimate their overall satisfaction with the treatment of the EHL and/or the EDL dystonia using a numeric scale (from 0 = not satisfied at all to 10 = very satisfied).

2.4. Statistical analysis

Continuous variables were expressed as median and range, the ordinal variables as median and interquartile interval. The preoperative and postoperative comparisons were made using a Chi^2 test for the qualitative variables and a Wilcoxon test for the quantitative variables. The significance threshold was set at 0.05 (bilateral significance). The statistical analyses were performed using SPSS software (version 20, SPSS Inc., Chicago, IL, USA).