Surgical treatment for the thumb-in-palm deformity in patients with cerebral palsy (Review)

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This is a reprint of a Cochrane review, prepared and maintained by The Cochrane Collaboration and published in The Cochrane Library 2006, Issue 1

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This record should be cited as:

This version first published online: 19 October 2005 in Issue 4, 2005.
Date of most recent substantive amendment: 27 June 2005

ABSTRACT

Background
Thumb-in-palm deformity in patients with spastic cerebral palsy is a deformity that impairs the ability to use the thumb and thus severely limits hand function. From the variety of operative procedures that have been described, it may be clear that there is no consensus on the surgical treatment of thumb-in-palm deformity.

Objectives
To review the efficacy of surgical interventions for the thumb-in-palm deformity in patients with spastic cerebral palsy; to review the selection criteria to surgically treat thumb-in-palm deformity in these patients; and to review the outcome assessment used in these studies.

Search strategy
We identified studies for inclusion from searches of several electronic databases: the Cochrane Central Register of Controlled Trials (Issue 4, 2003), MEDLINE (1966 to December 2004), EMBASE (1980 to December 2004) and CINAHL (1982 to December 2004). We also cross-checked the reference lists of these studies to identify additional studies.

Selection criteria
We considered a trial eligible for inclusion when it met the following criteria. 1) It was described as a randomized clinical trial, clinical controlled trial or prospective study that compared pre-operative- with post-operative outcome assessment. 2) It concerned patients with thumb-in-palm deformity affected by spastic cerebral palsy. There was no restriction in age. 3) It compared or described any surgical intervention to the thumb. 4) It followed subjects for at least six months. 5) Outcomes described included one or more of the following items: rate of success; functional improvement; active and passive range of motion of the thumb; grasp and release; pinch grip; complications and side effects; and quality of life.

Data collection and analysis
Two authors assessed each study using a scoring system. Meta-analysis was not possible because the selected studies were poorly designed, and the results were presented in an incompatible form. Therefore, we compiled a descriptive summary of the results of the individual studies. We did not attempt to acquire the raw data for re-analysis.

Main results
We identified 14 prospective studies that compared preoperative and postoperative outcomes as eligible for inclusion in this review. We found no randomized clinical trials or controlled clinical trials. The studies with the best available evidence were prospective studies that compared pre- with post-operative assessment. After assessment, we ultimately included nine studies.

Participants
In all the included studies the participants were more or less homogeneous regarding the most important prognostic indications. The nine included studies treated 234 patients. Age at operation ranged from 4-48 years (Median approximately 11 years).

Interventions
Twenty-four different specific interventions were performed, or combined, aiming to 1) stabilize the first metacarpophalangeal joint, 2) weaken the spastic thumb adductors, and 3) augment thumb abduction and extension.

Outcome measures
All of the included studies assessed whether the thumb had stayed out of the palm at follow-up. Additional outcome measures varied among studies.

Selection criteria
There was no consensus on the selection criteria for eligibility for surgical treatment. There was also considerable variety in the use of methods of assessment among the studies. There is no standardized method to evaluate the pre- and post-operative data, and most of the assessment methods were not validated. It was impossible to compare the outcomes among studies. Judgement about the effectiveness of one particular surgical intervention was not possible, because different surgical interventions and co-interventions were used within most studies.

Nonetheless, generally, the outcome of surgical treatment of thumb-in-palm deformity was considered satisfactory to both patients and to surgeons in all studies.

Authors' conclusions
Because the methodological quality of the studies is poor, it is impossible to provide a reliable judgement of the role of surgery for thumb-in-palm deformity. This review has demonstrated the need for randomized clinical trials or controlled clinical trials on the surgical treatment of thumb-in-palm deformity. Surgical reconstruction appears to improve hand function, to facilitate hygiene, and to improve the appearance and quality of life. For patient selection, a validated classification system should be developed to determine the type and extent of the cerebral palsied hand. The influence of age, intelligence, and voluntary muscle control still needs to be investigated. Investigators should focus on one particular surgical intervention or a specific group of interventions to find out which procedures produce the best functional improvement.

Plain Language Summary
Thumb-in-palm surgery appears to lead to modest improvement in hand function and quality of life in people with cerebral palsy. Cerebral palsy (CP) is a movement disorder caused by damage to the brain around birth. It causes muscle spasms and contractions that can lead to a condition called thumb-in-palm deformity, severely limiting hand function. Thumb-in-palm surgery is sometimes tried to improve ability to use the thumb. This review found no randomized controlled trials of thumb-in-palm surgery, but some other types of studies. Although some people with CP who had thumb-in-palm surgery experienced some improvement in movement, the improvement was generally modest. Trials are needed to show if thumb-in-palm surgery can improve function and quality of life for people with CP.

Background
Cerebral palsy is a collective term for a spectrum of motor disorders resulting from a non-progressive insult to the immature brain that occurs in the fetal or perinatal period (Blasco 1992). It includes motor impairments resulting from cerebral injury during infancy. It is relatively common, with a prevalence of 1-7/1,000 neonatal survivors (Nelson 1978).

Spastic cerebral palsy is the most common form of cerebral palsy. Spasticity or velocity dependent increase in tone is a consequence of the involvement of the pyramidal tract. Spasticity, in combination with imbalance of agonist-antagonist muscles, causes dynamic joint contractures which, left untreated, lead to static or fixed joint contractures. Growth of the spastic muscles fails to keep up with skeletal growth (Ziv 1984). These abnormal forces act on the vulnerable immature skeleton creating bony deformities. Overall function is affected.

The consequences can be burdensome and lifelong. The impact on the children, their families, the agencies responsible for their well-being, and society in general is substantial. The physically disabled have problems with mobility, accessibility, transportation, housing, integration with society, schooling and higher education, employment, and health care. The overall cost to society is significant (Rang 1989).

The management of spastic cerebral palsy is multi-disciplinary. It includes physiotherapy to maintain joint motion and muscle strength; casts and braces to obtain and maintain stretch or joint support; pharmacological and surgical methods to alleviate spas
ticity; and orthopedic surgery to correct joint contractures and bony deformities. Despite regular physiotherapy and pharmacological methods to reduce tone, spasticity may persist or recur, and joint contractures and bony deformities may develop in many children with cerebral palsy. Patients with cerebral palsy often have a thumb-in-palm deformity, which impairs the ability to use the thumb and thus severely limits hand function. On each attempt to make a fist the patient grasps his or her own thumb. On each attempt to pinch, the thumb fits in, under, or between the fingers, most frequently between the second and third digits. This uncontrollable pathologic motor automatism interferes with prehension, and can make the hand functionless.

Depending on the severity of the thumb-in-palm deformity, the thumb is adducted against the palm of the hand and flexed between the fingers, sometimes accompanied by a hyperextended metacarpophalangeal joint. (Matev 1991). Flexion-adduction contracture of the thumb is a result of the lack of normal balance between agonists and antagonists and failure of the normal reciprocal innervation in the spastic muscles. It is caused by a lifelong hypertonicity of the adductor pollicis muscle, the flexor pollicis brevis muscle and sometimes the flexor pollicis longus muscle, in combination with a paresis of the extensor pollicis longus muscle, the extensor pollicis brevis muscle and abductor pollicis longus muscle. This muscle imbalance, resulting from combinations of spasticity and/or weakness, produces the deformities observed. The severity of the deformity has been classified by House (House 1994). The classification depends on the static as well as dynamic components of each deformity. Type I describes a ‘simple’ metacarpal adduction contracture; type II a metacarpal adduction contracture and metacarpophalangeal flexion deformity; type III a metacarpal adduction contracture combined with metacarpophalangeal hyperextension deformity or instability; and type IV a metacarpal adduction contracture combined with metacarpophalangeal and interphalangeal flexion deformities or instability.

Because cerebral palsy is the result of a non-progressive brain defect (Bartram 1964; Maurer 2002), the option of surgical correction of the thumb-in-palm deformity, in addition to conservative bracing therapy, was proposed as early as 1843 (Little 1843). Of all surgical treatment in cerebral palsy, the treatment of the thumb is the most challenging, because treatment of the thumb has the most recurrences of all operative treatment in cerebral palsy and the result is unpredictable and discouraging (House 1981; Stoffel 1913). From the variety of operative procedures that have been described between 1938 (Burman 1938) and 1999 (Van Heest 1999), it is clear that there is no consensus on any algorithm for treating thumb-in-palm deformity.

**OBJECTIVES**

The purpose of this review was to examine the efficacy of surgical interventions for the thumb-in-palm deformity in spastic cerebral palsy; to review the selection criteria to treat thumb-in-palm deformity surgically; and to review the outcome assessment used in these trials.

**CRITERIA FOR CONSIDERING STUDIES FOR THIS REVIEW**

**Types of studies**

Randomized Controlled Trials

Clinical Controlled Trials

Any prospective study that compared pre-operative with post-operative outcome measures.

We included all languages.

**Types of participants**

Studies that concerned patients affected by spastic cerebral palsy with thumb-in-palm deformity. There was no restriction on age.

**Types of intervention**

Any surgical intervention that 1) described (or referenced) in adequate detail and performed in a consistent manner throughout the study; 2) aimed to correct the thumb-in-palm deformity; and 3) carried out pre-operative and post-operative assessment of one or more of the following outcomes, provided that subjects were followed for at least six months.

**Types of outcome measures**

The main outcomes of interest were:

1. rate of success of a patient keeping his thumb out of the palm at follow up;
2. functional improvement, including:
   - active and passive range of motion of the thumb;
   - grasp and release;
   - useful pinch;
   - overall capacity of the affected hand;
   - appearance of the hand (cosmetic improvement);
3. complications and side effects;
4. quality of life.

**SEARCH METHODS FOR IDENTIFICATION OF STUDIES**

See: Movement Disorders Group methods used in reviews.

See: Collaborative Review Group search strategy

We based our search strategy on the clinical question and then conducted an electronic search of the Cochrane Movement Disorders Group Specialized Register and the Cochrane Central Register of Controlled Trials (CENTRAL) (The Cochrane Library, Issue 4, 2003), MEDLINE (1966 to December 2004), EMBASE (1980 to December 2004) and CINAHL (1982 to December 2004)
using the Cochrane standard search strategy from 1966 in any language (Dickersin 1994). We explored the bibliography of retrieved articles in order to identify further trials.

In MEDLINE, we combined a subject specific search with all three levels of the optimal strategy for identifying randomized controlled trials (Dickersin 1994). The three levels included searching with MeSH-terms (Medical Subject Headings), searching by free text-words, and searching with Clinical Queries in a sensitive or specific way. We modified this strategy to search other databases (Clarke 2001). We used a Boolean operator (AND, OR or NOT) to combine terms.

Two authors identified trials that met the inclusion criteria individually by scanning records retrieved by the initial search. We retrieved full-text articles and two authors independently reviewed each to decide inclusion or exclusion. When necessary, the authors resolved differences of opinion by discussion.

METHODS OF THE REVIEW

Critical appraisal of eligible trials
Two authors independently evaluated the full text of the selected articles. Each author assessed the methodological quality of each trial, using the generic evaluation tool developed by the Cochrane Musculoskeletal Injuries Group (see Group details). When necessary, the authors resolved differences of opinion by discussion.

Data extraction and analysis
Two authors extracted data independently. Meta-analysis was not possible because of incompatible presentation of results, thus we compiled a descriptive summary of the results of the individual studies. We used a modification of the Cochrane Musculoskeletal Injuries Group quality assessment tool (see Group details) in the evaluation of the included studies. We have shown the scoring scheme for 12 aspects of trial validity, plus brief notes of coding guidelines for selected items, in Table 01. Although we did compile the scores of the individual items, we did this to gain an overall impression rather than for quantitative purposes.

DESCRIPTION OF STUDIES

Types of studies
We identified 14 studies that compared preoperative and postoperative outcomes as potentially eligible for inclusion in this review. We found no randomized clinical trials or controlled clinical trials. The studies with the best evidence available were prospective studies that compared pre- with post-operative assessment. Following assessment, we included nine studies: Dahlin 1998; Filler 1976; House 1981; Inglis 1970; Lee 1984; Nylander 1999; Rayan 1996; Roth 1993; Tonkin 2001.

Participants
The nine included studies treated 234 patients. Pre-operatively, the participants were in general reasonably comparable in all the included studies, although differences still existed. At surgery, their ages ranged from 4 to 48 years, with a median age of approximately 11 years. Fifty-five percent were male. Three studies described intelligence quotient (House 1981; Inglis 1970; Lee 1984). In those studies, intelligence scores ranged from 30 to 125, with the average intelligence quotient above 70.

Interventions
Twenty-four different interventions were performed aiming to 1) stabilize the first metacarpophalangeal joint; 2) weaken the spastic thumb adductors; and 3) augment thumb abduction and extension.

Outcome measures
All studies assessed whether the thumb had stayed out of the palm at follow-up. All studies also assessed hand function in some way. Further outcome measures varied among studies, including grip strength, stereognosis, range of motion, patient-subjective scores of appearance, expectations and quality of life, and various tests of thumb and hand function and dexterity.

METHODOLOGICAL QUALITY

General findings
The best evidence available was at the level of prospective studies that compared pre-operative with post-operative assessment. Randomization was not performed in any of the studies. The outcome assessor was not blinded in any study. Patient groups were heterogeneous, and the type of interventions varied considerably; A total of 24 different procedures to correct the thumb-in-palm deformity were described. In most cases, treatment consisted of a combination of these separate procedures. Furthermore, the evaluated intervention was not always performed in the same way. Consequently, it was impossible to compare the results among the different studies. Pooling of results was not possible, nor was judging the effectiveness of one particular surgical intervention.

The outcome measures varied considerably among the studies. Their relevance was not always clear, and they were not validated in most cases.

Included studies
Dahlin 1998
The authors described eligibility criteria. Because all patients with a spastic hand in cerebral palsy of the spastic hemiplegic type were eligible, the patients were considered as heterogeneous at baseline. Furthermore, the combination of chosen interventions varied among patients. However, the interventions were well described and the patients were evaluated identically pre-operatively and at six and eighteen months post-operatively. Patients were asked about their expectations of the surgery, their ability to carry out activities of daily living (patients were video recorded), and the
ability to make certain grips/grasps was assessed. Stereognosis was evaluated as well. Co-interventions were not avoided or comparable, because many different surgical procedures were performed. The results were presented as a number of patients who gained a certain function divided by the total number of patients. Statistically significant differences were tested by using the chi-squared test, Fisher exact test or Wilcoxon signed rank test with Bonferroni correction. Point estimates and measures of variability were presented for the primary outcome measure. The dropout rate was described, but not acceptable for every part of the assessment.

Fillar 1976
This study specified eligibility criteria, and there was little heterogeneity at baseline. It also listed indications and contra-indications. The authors mentioned but did not explicitly describe interventions and adjuvant therapies. Evaluation was similar pre- and post-operatively, but the presentation of the results per individual was not clear. All patients underwent capsulodesis of the metacarpophalangeal joint. In all patients but two, additional releases and transfers (five different procedures in 11 patients) were performed, so the co-interventions were not entirely comparable. Outcome measures were relevant: metacarpophalangeal joint extension was evaluated, as was hand function (pinch). The report described adverse (side) effects, but not dropout rate.

House 1981
This study defined eligibility criteria and listed indications for surgery. Selection of patients depended on a number of objective and subjective parameters: voluntary motor control and sensibility in the hand, type and extent of the cerebral palsy, availability of motors for transfer, and age and intelligence of the patient. However, selected patients showed some heterogeneity at baseline, as patients from all types classified by the House Classification (Background) were selected for surgery. The study described interventions adequately. Evaluation was performed pre- and post-operatively in the same way by the patient, the parents, the therapist, and the physician, using a functional classification system. This system differentiates nine functional categories from zero ("does not use") to eight ("uses hand completely independently"). The result of surgery in each patient was expressed as a "functional improvement score". This score was calculated from the ratio of the observed improvement to the predicted improvement. The predicted improvement was estimated from the quality of voluntary control and sensibility present pre-operatively. Scores were assigned to voluntary control and sensibility. The observed improvement was calculated by dividing the difference between pre-operative and post-operative functional classes by the number of functional classes that would have to be gained to have normal function. Co-interventions were not entirely avoided or comparable, because different combinations of multiple surgical interventions were performed. These interventions were performed based on three categories: step I - release of contractures; step II - augmentation of weak muscles; step III - skeletal stabilization. The study also described adverse effects; the withdrawal/dropout rate; and the follow-up period.

Inglis 1970
This study did not clearly describe eligibility criteria. The authors based decisions on the feasibility of corrective surgery primarily on the specific need for improvement in function expressed by the patient. When sufficient physiological and anatomical resources were available to achieve the needed improvement, surgical correction was considered. All patients had thumb-in-palm deformity, but the extent of the deformity and the patient's condition was not described, so it was not clear whether the selected patients were comparable at baseline. The authors described interventions and co-interventions clearly. Evaluation was performed in the same way pre- and post-operatively. The patients underwent different surgical procedures. Co-interventions were not completely avoided, nor were they comparable in all patients. The study assessed grasp and release, pinch, and over-all function, and independently rated each as good, fair or poor. The authors provided a clear definition of these different ratings, and stated that there were no deaths or immediate post-operative complications, and that no patient worsened following surgery. There were no dropouts.

Lee 1984
This study clearly described eligibility criteria. The authors undertook selection on the basis of Goldner's criteria (Matev 1991). Patients were comparable at baseline regarding the most important prognostic factors: the patient had to be old enough to co-operate with the surgeon and therapist, and sufficiently intelligent to cooperate with post-operative therapy and benefit from the anticipated functional improvement. All patients were mildly spastic hemiplegics and had the ability of voluntary extension of fingers in the neutral position of the wrist. The authors described experimental and control interventions. For functional evaluation, assessors evaluated the patients' ability to perform activities of daily living (that were listed in a table) pre- and post-operatively. Co-interventions were not avoided or comparable, since different additional procedures were performed. Although the study described functional evaluation and rated outcome measures as excellent, good and fair, it was not clear which activities had improved post-operatively, since there was no detailed description of the pre-operative assessed data. The authors described no adverse effects and recorded no dropouts.

Nylander 1999
The authors clearly specified eligibility criteria. Patients were similar at baseline, as patients were assessed as group 2B or 3 by the Zancolli classification system (Matev 1991). Furthermore, none of the patients had any marked involvement of the shoulder or severe disturbance of sensibility in the affected hand. The authors explicitly described selection of the interventions in a protocol, and evaluated patients in the same way pre- and post-operatively, according to a protocol developed by the Committee of Spastic Hand Evaluation and an additional score value. Although the
study attempted to avoid co-interventions as much as possible, surgical treatment options varied considerably among the patients. This was because of the varying extent of the deformities among patients and because some patients needed a second operation. Adverse effects and the dropout rate were not stated. Outcome measures were relevant, but the score for functional outcome was a mean score for total hand function, rather than a thumb-specific one. As a result, we were not able to undertake evaluation of specific sub-scores for the thumb (thumb opening, lateral pinch and pulp pinch e.g.). Statistical analysis was performed using the Student’s t-test for independent sample means.

Rayan 1996
This study specified eligibility criteria. Surgical indications were: inadequate thumb abduction-extension, a thumb that remained in the palm during release, caught in a clenched fist during grasp and a first web space angle less than 45°. Patients with poor hygiene and cognition, lack of motor control, severe deformity, and muscle contracture were surgical candidates, but not for the purpose of improving function. Patients were comparable at baseline, but no criteria were described to distinguish different degrees of severity of the deformity. They were rated as mild, moderate and severe. Evaluation included both subjective and objective parameters. Subjective evaluation included pre- and post-operative performance of 17 common activities of daily living provided by a questionnaire. Objective evaluation considered stereognosis, two-point discrimination, motor control, position of the thumb at rest and during grasp and release, the ability to grasp and release, the first web space angle, and the condition of the palmar skin. The latter two were not evaluated pre- and post-operatively. Although a modified extensor pollicis longus tendon rerouting was performed, it was always in combination with other surgical interventions. We did not consider the outcome measures entirely relevant, as the number of gained activities did not include many activities that need thumb function specifically (opening a jar, buttoning a shirt and zipping trousers). Adverse effects (recurrence of the thumb-in-palm deformity) were mentioned, but not described in detail. There were no dropouts.

Roth 1993
The study listed eligibility criteria, not specifically for thumb-in-palm deformity, but considered the whole upper limb. All the patients could move their arms voluntarily, and none of them had athetosis or were cognitively unable to follow rehabilitation training. The authors explicitly described interventions and adjuvant therapies. A single surgeon performed all procedures and each patient underwent a more or less comparable set of surgical procedures. A modified version of House’s nine point functional rating scale was used to assess hand function (House 1981). Inter-observer reliability for the modified House functional classification was 0.89 - 0.92, indicating that the modifications did not interfere with the reliability of the scale. There were no complications related to the surgery. There were no dropouts.

Tonkin 2001
This study specified eligibility criteria, but not in detail. Patients had thumb-in-palm deformity of all three different classes (House 1981), and 30 of 32 patients had spasticity. Both hemiplegic and quadriplegic patients were included. Patients were examined to decide which surgical interventions were required. This study explicitly described the assessment pre- and post-operatively using the same functional assessment system performed by the same team. Five criteria were chosen to assess functional ability: eating (ability to hold a knife and fork and cut up food), dressing (pulling up trousers and doing up buttons), tying shoelaces, riding a bicycle (ability to grasp and release) and playing sports. This study also documented hand sensibility, classification type (by House) and passive range of motion. All patients received different interventions and co-interventions that were not always comparable. Therefore it was not possible to relate the outcomes of this study to one particular surgical intervention or combination of surgical interventions and compare the interventions that were used. Functional improvement and mean active position of thumb joints were given, but data were only presented as averages. The variability of the results was not presented. The study mentioned adverse effects, but did not describe them in detail. There were no dropouts.

Excluded studies
Goldner 1990
Selected patients showed large heterogeneity regarding the most important prognostic indications at baseline. Not all the patients in the study had spastic cerebral palsy. Twenty patients had athetosis, which is not included in this review.

Hoffer 1983
Evaluation was not primarily pre- and post-operatively, but comparison between three different groups (patients with total adductor release, patients with partial adductor release, and patients who were not operated on), was based on pre-operative electromyography study. This was not included in this review. Outcome measures were considered not relevant.

Keats 1965
Patients were not evaluated identical pre- and post-operatively.

Tonkin 2002
Data in this manuscript were of the same patients as used in the included study of Tonkin 2001. For that reason, this manuscript was excluded for the review. However, it provided useful additional information on the inclusion and exclusion criteria of the patients and of the surgical techniques, and this information was used for the current review.

Van Heest 1999
This study does not explicitly evaluate surgical interventions of thumb-in-palm deformity, but surgical interventions of deformities in upper limb cerebral palsy in general. Information of specific...
function of the thumb was lacking. Patients were not considered homogeneous at baseline.

RESULTS

Selection criteria
There was no consensus on the selection criteria for eligibility for surgical treatment. The expected significance of the severity of thumb-in-palm, age, intelligence, sensibility and voluntary muscle control for surgical outcome differed among the studies, but none of the studies provided evidence-based support for selection criteria. Selection criteria for the studies evaluated depended upon the goals of the treatment. Some voluntary control appeared to be necessary to achieve functional improvement, so the thumbs with the best pre-operative function were expected to have the best result. By comparison, surgery undertaken for hygienic and cosmetic reasons allows thumb-in-palm deformities of all severities in all patients for an adequate result.

Outcome measures
There is no standardized method to evaluate the pre- and post-operative data and most of the assessment methods were not validated. It was impossible to compare outcomes among the studies. It was also not possible to make judgments about the effectiveness of one particular surgical intervention, because different surgical interventions and co-interventions were used within most studies, and the outcome measures also varied considerably.

Efficacy of surgery
Dahlin 1998 corrected the thumb-in-palm deformity completely in 31 of the 36 patients. At six months postoperatively various grips and grasps had improved significantly (p < 0.02). Furthermore, stereognosis improved significantly (p < 0.05). The improved function of the thumb increased the overall grip ability. The authors ascribed the improvement of stereognosis to functional cerebral reorganization induced by a modified afferent inflow. The study did not describe complications or side effects, and indicated no significant improvement in activities of daily living. However, 95% of the patients reported that all or several of their expectations had been fulfilled by the surgery. Improvement of appearance was not described.

Treatment in Fillter 1976 consisted of capsulodesis of the metacarpophalangeal joint of the thumb, leaving the joint in 30° to 35° of flexion. To evaluate functional improvement, the degree of extension and flexion of the metacarpophalangeal joint of the thumb was measured, and key pinch was assessed. Pre-operatively, all 13 children had between 29 and 42° of hyperextension. Post-operatively, 11 of the 13 patients had a position of flexion of between 20 and 33°. No patient had recurrence of hyperextension of the metacarpophalangeal joint, although two patients showed no improvement. Key pinch had improved in all. Each patient had improved flexion of the interphalangeal joint of the thumb after operation, but the interphalangeal joint went into some hyperextension in five out of 13 patients. The study suggested that improvement of flexion resulted in a better functional pinch between the side of the index finger and the broad pulp of the thumb. Furthermore, appearance of the hand had noticeably improved. The study did not describe improvement in quality of life.

In House 1981, function improved in 56 consecutive selected patients. Half the patients improved by three or more functional classes, and all improved at least one class. Adverse effects were described adequately. One patient suffered loss in pinch force. This was assumed to be a result of over-lengthening that had resulted from a combination of immobilization of the arm and with the thumb in abduction and extension and the wrist also in extension. Another patient had an apparent rupture of the flexor pollicis longus at the site of the lengthening six weeks postoperatively. The study did not describe improvement in quality of life or cosmetic improvement.

In Inglis 1970, grasp was the most useful functional improvement, because four out of five patients gained grasp. Furthermore, one out of five patients gained pulp-to-pulp pinch, three out of five patients key pinch; all retaining a mobile thumb. The study stated that the number of patients with spastic paralysis who benefit from surgery is small compared to the total number of patients with cerebral palsy or to patients who benefit from surgery of lower extremities. No conclusion was drawn on the effectiveness of the surgical procedures performed. The study did not describe complications or side effects specifically, although it did state that no patient was worse following surgery. All patients were rated improved in appearance. The study did not describe the effect on quality of life.

Lee 1984 achieved active abduction and extension in 13 of the 14 brachioradialis tendon transfers. All but one patient reported restoration or improvement of opposition and pinch grip. Although the study stated that activities of daily living were evaluated, the authors did not present the results of this evaluation. Furthermore, the functional improvements presented were the result of a combination of multiple operations. Therefore, we were unable to draw any conclusions from the results. The study did not describe complications or side effects, but noted that the appearance of the thumb and hand improved in all patients.

Nylander 1999 reported that dysfunction of the arm was significantly reduced six months after the reconstructive surgery, and that improvements remained essentially unchanged at the later follow-up. The mean pre-operative score was 21.5 points; at six months it was 26.5 points (P < 0.001). The mean score at the 4.5-year follow-up was 2.5 points. The mean score at final assessment was 27.5 points, which was not significantly different from the six-month score. In five of the 27 patients, functional swan-neck deformities of the metacarpophalangeal joint recurred after the first operation and required a second operation. Twenty-one patients subjectively rated their hand function as improved. The study did
not describe improvement in quality of life or appearance, but 22 patients stated that they would undergo the same procedure again. Rayan 1996 showed that during grasp and release and at rest, the thumb remained out of the palm in all patients after surgery. In 12 patients, the surgery improved muscle balance around the thumb, but active abduction and pinch against the index were not achieved in every patient. Seven patients were able to actively extend and abduct the thumb. Patients with mild deformity had consistently satisfactory outcomes and better objective functional results than those with severe deformities and poor cognition. Patients with satisfactory functional results exhibited adequate motor control. They stated that for functional improvement adequate cognition, sensibility and sufficient motor control were required. Otherwise, only improvement of hygiene and appearance is achieved. Voluntary motor control needs to be present, but even with sensory defect functional improvement can be achieved. Low intelligence and sensory deficit are not considered contraindications for surgery. In severe cases the best results were with extensor pollicis longus rerouting and metacarpophalangeal fusion, and a combined aductor pollicis and first dorsal interosseous release. When extensor pollicis longus rerouting was the only operation on the thumb, less web-space correction was obtained as compared to those who had additional procedures. Quality of life had improved, since patients and caretakers were all uniformly satisfied about the activities of daily living after surgery. This was true even in two patients who were considered objectively to have unsatisfactory results. Cosmetic improvement was not described.

In Roth 1993, 14 children had thumb-in-palm deformity prior to surgery and all maintained the thumb out of the palm after surgery. Three patients gained active abduction, but this was not a statistically significant improvement. After surgery there was a significant increase in grasp and release function of the hand. This allowed the hand to actively assist the other hand rather than act as a helper. There was no correlation between hand sensibility and outcome of surgery in this study. The authors concluded that impaired vision and poor sensation together are contra-indications to surgery, but did substantiate this conclusion. The study did not describe complications or side effects. Quality of life and the appearance of the hand were considered improved, since the position of the hand improved. Participants recorded both improved functional performance and high parental satisfaction.

Of 32 patients enrolled in Tonkin 2001, 15 reported improvement in eating, five in tying shoe-laces, 12 in dressing, five in riding a bicycle, and 10 in playing sports. Although the average improvement in metacarpal abduction was 19º, this was considered functionally significant when it brought the thumb ray out of the plane of the palm. In the 18 patients who underwent sesamoid capsulodesis, the position of the metacarpophalangeal joint improved from an average of 30º hyperextension preoperatively to 15º flexion postoperatively. The thumb maintained to stay out of the palm in 29 patients, and 26 patients gained lateral pinch. No patient improved from dependant to independent functioning. A side-effect was that although extensor pollicis longus to extensor pollicis brevis transfer supplemented metacarpophalangeal joint extension, it decreased extensor activity to the interphalangeal joint of the thumb. In five patients who had severe deformities, the primary goal was to improve cosmesis and hygiene. All five reported improved appearance, and two reported establishment of lateral pinch.

**DISCUSSION**

We undertook a Cochrane Systematic Review in order to review the efficacy of surgical interventions for the thumb-in-palm deformity in patients with cerebral palsy; to review the selection criteria used to define thumb-in-palm deformity; and to review the outcome assessment used in these trials.

**Methodological quality of the review**

The studies included in the review were of a disappointingly poor methodological quality, because all studies failed to meet one or more validity criteria. This indicated a high risk of bias. There was no randomization performed in any of the studies. As a matter of fact, there was no comparison of groups at all! As a result, it was impossible to perform analysis of either treatment allocation concealment or intention to treat. Additionally, the outcome assessors in the studies were never blinded and performance bias may have occurred in all studies. As such, the poor quality of all studies constituted grounds for exclusion. The studies with the best evidence available were observational studies, prospective studies that compared pre-operative with post-operative assessment. We decided to accept those studies for inclusion because evidence from non-randomized studies may supply information on the types of participant or outcomes that are of relevance to the review. Such evidence does pose problems and threats to validity as unexpected biases may occur and invalidate the conclusions (Clarke 2001).

None of the included studies described approval from an ethical committee or obtained written informed consent of the patients.

**Findings of the review and mechanisms of findings**

**Selection criteria**

All patients in the studies had thumb-in-palm deformity caused by spastic cerebral palsy. However, criteria on the type and extent of the deformity may be needed because they may co-determine the outcome (Rayan 1996). Similarly, quality of voluntary muscle control, hand sensibility, age and intelligence quotient of the patient may be co-determinants (Rayan 1996). Finally, the goal of the surgical reconstruction should be clearly stated, as it determines the rating of success. Surgical reconstruction may be indicated to improve function, to facilitate hygiene, or improve appearance (Tonkin 2002). There is no consensus on the relative importance of the several co-determinants. Unfortunately, these co-determinants were poorly presented in most studies and, therefore, patients may have been non-comparable among the studies.
and heterogeneous within the studies. As a result, the relative importance of the various co-determinants as selection criteria could not be studied well. None of the indications presented below are validated, but are merely a reflection of the expert-opinion of the several surgeons.

Type and extent of the deformity
Classification with a validated system is helpful in the decision as to what surgical intervention should be performed in every specific case. Since none of the studies used the same classification system for thumb-in-palm deformity and some studies did not classify the type of deformity at all, there is no consensus about the selection of patients based on the type and severity of thumb-in-palm deformity. Different classification systems were used. Zancolli and Goldner designed a classification system for the cerebral palsied hand in general, which does not concern the thumb (Matev 1991). House's classification is specialized for thumb-in-palm deformity (House 1981; House 1994). Tonkin designed a modified version (Tonkin 2001; Tonkin 2002) of House's classification. None of these classifications systems have been validated. Moreover, the classification of patients has so far not led to agreement on the strategy for treatment.

Sensibility
Although normal sensibility is important for good hand function, it was more or less agreed that this is not an absolute prerequisite (House 1981; Matev 1991; Roth 1993). As long as there is a reasonable muscle balance and useful voluntary control, the patient can compensate, at least in part, for any sensory deficit in the hand by visual control (House 1981; Roth 1993). As a consequence, impaired sensibility of the hand is a considerable disability when there is associated visual impairment (Roth 1993; Tonkin 2002).

Voluntary motor control
Several authors stated that for the purpose of functional improvement, voluntary muscle control needs to be present. Even with impaired sensation, functional improvement could be achieved when voluntary motor control is sufficient (House 1981; Rayan 1996; Roth 1993). Muscles that fire in phase with good control of contraction and relaxation seem to be the most effective candidates for tendon transfers (Tonkin 2002).

Age
There is no consensus on the best age for surgery. Ideally, the patients need to be old enough to understand the reasons for surgery and to co-operate during postoperative rehabilitation (Tonkin 2002). However, within the studied age group older patients did not seem to have better or worse results than younger patients.

Intelligence
One study stated that cognition and motivation were important determinants for the outcome of the tendon transfers (Rayan 1996). However, the same authors do not consider low intelligence and sensory deficit contraindications for surgery (Rayan 1996). If cognition and motivation were absent, surgical treatment would help to improve hygiene and appearance. Moreover, intelligence does not seem to play a very important role in predicting results of another study (House 1981). As such, although there is no consensus about the exact level of cognition as an inclusion criteria for surgery, it is generally recommended that patients are trainable (Matev 1991; Tonkin 2002).

Outcome measures
Since there was a great variety in the outcome measures used in the included studies, it is not possible to recommend a set of outcome measures based on the included studies. The great variety of the assessment methods among the studies may be explained by a lack of a ‘gold standard’ to evaluate function. Most of the assessment methods were not validated. All studies showed that the thumb remained out of the palm at follow-up; this appeared to be an adequate and simple assessment. To evaluate the improvements of thumb function, pinch grip could be a valid test. For testing dexterity and ADL-function, there is no widely used tool.

Efficacy of the surgery
Surgical techniques are designed to decrease the strength of the deforming forces, augment the weakened muscles and stabilize unstable joints. Overall the results of surgery in the studies appeared to be good. Surgery does seem to be an adequate choice for treating thumb-in-palm deformity to achieve a significant, but modest, improvement in range of motion, pinch grip and hand function. However, as the results were based on empirical evidence only, the success of the surgery may be explained by selection bias, inadequate concealment of allocation and lack of double blinding that have resulted in over-estimates of the effects of treatment (RevMan 2000). Furthermore, because within all the studies the patients underwent different surgical procedures, it was not possible to compare the results of the interventions separately. This was also stated in two of the included studies (Lee 1984; Rayan 1996).

The clinical experience is that, of all corrections in cerebral palsy, the correction of the thumb-in-palm deformity is especially difficult and the problem often recurs (House 1981; Stoffel 1913). However, this experience was not reflected in the included studies; all studies showed significant improvement, with few recurrences. This could indicate that the time to follow-up was too short in the included studies. None of the included studies discussed the apparent limited duration of the effect of the operation.

Limitations and strengths of the present review
We presented all the available studies on surgical treatment of the thumb-in-palm deformity that compared surgical outcome to baseline, using a variety of outcome measures. No controlled studies of surgical treatment for the thumb-in-palm deformity have been conducted that compare the outcomes of different surgical techniques. Studies with best evidence available were prospective studies that compared pre-operative with post-operative assessment. As a consequence, heterogeneity among the selected studies was present and bias occurred. It was not possible to pool the
results and prepare a meta-analysis. As such, no evidence-based clinical decision can be made based on the present review.

**AUTHORS’ CONCLUSIONS**

**Implications for practice**

Because the methodological quality of the studies is poor, it is impossible to provide reliable conclusions based on the currently available data of this systematic review. However, each individual study reported improvement of hand function. These positive results were modest and no patient had perfect hand function postoperatively. We do not know whether another combination of treatments would have resulted in an even better outcome for a patient. None of the interventions showed dramatic improvement, but it appears that any of the proposed surgical interventions is better than none.

The included studies all recommend that successful surgical treatment requires accurate, detailed preliminary assessment, careful selection and performance of the surgical interventions and a postoperative program of protective splinting that is coordinated with guided physical therapy. Selection criteria include some voluntary motor control and the cognitive ability and motivation to follow a rehabilitation program. Age, sensibility and the severity of the deformity do not appear to be strict selection criteria.

When the thumb-in-palm deformity is corrected, the range of motion of the thumb improves and it is possible that the patient’s ability to perform activities of daily living improves.

There is a lack of validated assessment methods. The Melbourne Assessment of Unilateral Upper Limb Function is a promising tool that has been developed recently. A study in 2001 concluded that it is a reliable tool for measuring the quality of unilateral upper-limb movement in children with cerebral palsy (Randall 2001).

**Implications for research**

This systematic review has demonstrated the need for further research on the surgical treatment of thumb-in-palm deformity in patients with cerebral palsy. It has revealed the need for better methodological quality as well. Therefore, randomized clinical trials or controlled clinical trials should be conducted. It is important to establish the relative (dis)advantages of the various procedures and to know what techniques may provide a beneficial outcome, given the preoperative condition of the patient and his or her hand. Therefore, selected groups of patients need to be homogeneous. Although selection criteria are often specifically stated, there is still controversy about the influences of age, intelligence and sensibility for overall result, and clearly, such criteria are not evidence-based. The specific effect of these factors on the outcome of surgical treatment should be investigated. Furthermore, a validated classification system may help to determine the type and extent of the cerebral palsied hand. Classification based on the House classification that is specialized for thumb-in-palm deformity can help to select patient-‘tailored’ surgical interventions. The effectiveness of particular surgical interventions may vary among different levels of thumb-in-palm deformity. Within the trials, investigators should focus on one particular surgical intervention, or a set of interventions, to avoid bias of co-interventions that hinder comparison of experimental groups. We also recommend that studies are designed to investigate different procedures that aim for the same functional goal. As such, we recommend studying the efficacy of: different techniques of performing rerouting of EPL muscle for increasing thumb abduction; capsulodesis versus arthrodesis to stabilize the first metacarpal joint; augmentation of the (rerouted) EPL with the brachioradialis muscle versus the PL muscle, versus FDS IV muscle versus no augmentation at all; augmentation of rerouted EPL muscle versus augmentation of APL muscle; adductor pollicis release versus adductor pollicis release + release of FPB muscle.

As inter-individual differences among patients are large in this specific patient population, large numbers of patients have to be studied, and possible subgroups have to be recognized. Because the recurrence rate of thumb-in-palm deformity is considered notoriously high, patients should be followed for a longer period than three years. Furthermore, the outcome assessment should be standardized: the Melbourne Assessment of Unilateral Upper Limb Function may be a reliable tool for measuring the quality of upper-limb movement.

**POTENTIAL CONFLICT OF INTEREST**

None known.

**SOURCES OF SUPPORT**

**External sources of support**

- No sources of support supplied

**Internal sources of support**

- Dept plastic, reconstructive and hand surgery, AMC NETHERLANDS
References to studies included in this review

Dahlin 1998 (published data only)

Filler 1976 (published data only)

House 1981 (published data only)

Inglis 1970 (published data only)

Lee 1984 (published data only)

Nylander 1999 (published data only)

Rayan 1996 (published data only)

Roth 1993 (published data only)

Tonkin 2001 (published data only)

References to studies excluded from this review

Goldner 1990

Hoffer 1983

Keats 1965

Tonkin 2002

Van Heest 1999

Additional references

Bartram 1964

Blasco 1992

Bburman 1938

Clarke 2001

Dickersin 1994

House 1994

Little 1843

Matev 1991

Maurer 2002

Nelson 1978

Randall 2001

Rang 1989
Table 1: Characteristics of included studies

<table>
<thead>
<tr>
<th>Study</th>
<th>Methods</th>
</tr>
</thead>
<tbody>
<tr>
<td>Stoffel 1913</td>
<td>Preoperative versus 6 and 18 months postoperative assessment of different grips and grasps, supination and range of motion (ROM), stereognosis, expectations, the ability to carry out activities of ADL</td>
</tr>
<tr>
<td>Van Heest 1999</td>
<td>36 patients</td>
</tr>
<tr>
<td>Ziv 1984</td>
<td>36 patients</td>
</tr>
</tbody>
</table>

*Indicates the major publication for the study
Characteristics of included studies (Continued)

**Interventions**

Several combinations of the following interventions in different patients:
- Division of the insertion of the AP and a partial release of the 1st DIO muscle together with a Z-plasty of the web space.
- Transfer of the BR to the APL tendon
- Transfer of BR to EPB tendon
- Transfer of BR to EPL
- Transfer of ECU to APL
- Transfer of FCR to EPB
- Transfer of EI to APB
- Transfer of EI to EPL
- Transfer of EI to EPB
- Transfer of EDQ to EPB
- Transfer of EDQ to EPL
- Transfer of PL to EPB
- Transfer of PL to APL
- Transfer of EI to EPL
- Transfer of FPL to EPL
- Tenodesis EPB
- FPL lengthening
- EPL rerouting
- Capsulodesis of MCP I joint
- Arthrodesis of MCP I joint

**Outcomes**

A significant improvement of the various grips and grasps postoperatively (P < 0.02). Significant improvement in stereognosis. In 31 of the 36 patients the thumb-in-palm deformity was completely corrected. The improved function of the thumb increased the overall grip ability. Stereognosis improved. The improvement was considered due to functional cerebral reorganization induced by the modified afferent inflow. Complications and side-effects were not described. Activities of daily living did not improve significantly. 95% of the patients had all or several of their expectations fulfilled.

**Notes**

Allocation concealment D
### Study House 1981

**Methods**
Pre- and 2 to 12 years post-operatively evaluation by the patient, the parents, the therapist, and the physician of nine functional categories (from "does not use" to "spontaneous use").

**Participants**
- 56 patients
- Age range: 4 - 20 yrs.
- Mean age: 11.8 yrs.
- Boy/girl ratio: 36/20
- IQ range: 30 - 119. Mean IQ: 77.5

**Interventions**
- z-plasty of the first web space, recession of the 1st DIO muscle, and lengthening of the AP.
- Transfer of PL, BR, or FCR to EPL, APL, or EPB
- Arthrodesis of the MCP joint and occasionally of IP joint

**Outcomes**
Improved function in 56 consecutively selected patients was established. One-half of the patients improved by three or more functional classes and all were improved by at least one class. In one patient there was loss of pinch power due to overlenghtening of the thumb in the postoperative cast. Another patient had an apparent rupture of the flexor pollicis longus at the site of the lengthening six weeks postoperatively.

**Notes**
Allocation concealment: D

### Study Inglis 1970

**Methods**
Prospective study with pre- and postoperative assessment of
- grasp
- pinch.

**Participants**
- 28 patients
- Age range: 9 - 48 yrs.
- Mean age: 11.8 yrs.
- Boy/girl ratio: 13/15

**Interventions**
A surgical plan was developed for every individual patient out of the following options:
- recession of the origin of the adductor pollicis
- release of the insertion of the adductor pollicis and the first dorsal interosseus muscle
- Arthrodesis of the MCP I joint
- FPL lengthening
- Transfer of FCR to APL
- APL rerouting
- Transfer of FCR to EPL
- EPL rerouting

**Outcomes**
All retained a mobile thumb. Twenty-three out of 28 patients gained grasp. Twelve gained pinch grip. Complications and side-effects were not described, but no patient was worse following surgery. All patients improved in appearance.

**Notes**
Allocation concealment: D

### Study Lee 1984

**Methods**
Pre- and 3 to 10 years. (average 6.5 years) postoperative evaluation of anatomy, function, and cosmesis.
- pinch
- ROM
- evaluation of the ability to perform activities of daily living

**Participants**
- 14 patients
Characteristics of included studies (Continued)

Age range: 5 - 14 yrs. Mean age: 7.5 yrs.
Boy/girl ratio: 6/8
IQ: above 70

Interventions
- Transfer of BR to EPL and/or APL

Outcomes
Active abduction and extension was achieved in 13 of the 14 patients. Opposition and pinch grip was restored or improved in all patients except in one. Although the study stated that activities of daily living were evaluated, results of this evaluation were not described. The appearance of the thumb and hand improved in all patients.

Notes
Allocation concealment D

Study Nylander 1999

Methods
Prospective study with preoperative and a mean of 4.5 years postoperative assessment of function by a custom score-system.

Participants
24 children
Age range: 6 - 19 yrs. Mean age: 10 yrs.

Interventions
- Lengthening of Adductor pollicis
- Lengthening of the FPL.
- Transfer of FDS IV or PL to EPL or EPB
- Shortening of the APL

Outcomes
Dysfunction of the arm was significantly reduced six months after the reconstructive surgery and the improvements remained essentially unchanged at the later follow-up. The mean pre-operative score was 21.5 points and at six months it was 26.5 points (P < 0.001). The mean score at 4.5 year follow-up was 2.5 points. The mean score at final assessment was 27.5 points. This was not significantly different from the six month score. In five of the 27 patients functional swan-neck deformities of the metacarpophalangeal joint remained after the first operation and required a second operation.

22 patients stated that they would undergo the same procedure again, because they benefited form the surgery. And 21 patients subjectively rated their hand function as improved.

Notes
Allocation concealment D

Study Rayan 1996

Methods
A subjective and an objective evaluation were performed including pre- and post-operative performance of 17 common activities of daily living provided by a questionnaire. Objective evaluation considered stereognosis, two-point discrimination, motor control, position of the thumb at rest and during grasp and release, the ability to grasp and release, the first web space angle, and the condition of the palmar skin.

Participants
14 patients (15 hands). Age range: 6 - 30 yrs. Mean age: 12.2 yrs.
Boy/girl ratio: 6/8

Interventions
A modified EPL- rerouting was performed in combinations with several surgical procedures.

Outcomes
The thumb remained out of the palm in all patients. In twelve patients, the surgery improved muscle balance around the thumb, but active abduction and pinch against the index were not achieved in all. Seven patients were able to actively extend and abduct the thumb. Patients with mild deformity had consistently satisfactory outcome and better objective functional results than those with severe deformities and poor cognition. Patients with satisfactory functional results exhibited adequate motor control. Low intelligence and sensory deficit are not considered contraindications for surgery. In severe cases the best results were with extensor pollicis longus rerouting and metacarpophalangeal fusion, and a combined adductor pollicis and first dorsal interosseous release. When extensor pollicis longus rerouting was the only operation on the thumb, less web-space correction was obtained as compared to those who had additional procedures.
### Characteristics of included studies (Continued)

**Notes**
- Quality of life improved.
- Allocation concealment: D

<table>
<thead>
<tr>
<th>Study</th>
<th>Methods</th>
<th>Participants</th>
<th>Interventions</th>
<th>Outcomes</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Roth 1993</strong></td>
<td>Patients were evaluated pre- and at a mean of 2.6 years post-operatively. A modified version of House's 9 point functional rating scale was used (House 1981). Active and passive range of motion, strength, type of grasp and release patterns and bimanual use of the hands were assessed.</td>
<td>17 children Age range: 3.3 - 15.5 yrs. Mean age: 8.3 yrs. Boy/girl ratio: 11/6 IQ range: 64 - 125. Mean IQ: 99.8</td>
<td>All patients had transfer of BR to APL and EPB. Further procedures varied among patients</td>
<td>All maintained the thumb out of the palm at follow-up. Three patients gained active abduction. Grasp and release function of the hand increased significantly. There was no correlation between hand sensibility and outcome of surgery. Quality of life and the appearance of the hand improved.</td>
</tr>
<tr>
<td><strong>Tonkin 2001</strong></td>
<td>Prospective study with preoperative and postoperative assessment of functional tasks.</td>
<td>32 patients (33 thumbs) Age range: - Mean age: approx. 10 yrs. Boy/girl ratio: 15/17</td>
<td>Surgical techniques included releases, tendon transfers and joint stabilization.</td>
<td>Of 32 patients, The thumb maintained out of the palm in 29 patients and lateral pinch was established in 26 patients. Eating had improved in fifteen patients, tying shoe-laces in five, dressing in twelve, riding a bicycle in five, and playing sports in ten. Although the average improvement in metacarpal abduction was only 19º, this was functionally significant as it brought the thumb ray out of the plane of the palm. In the eighteen patients who underwent sesamoid capsulodesis, the position of the metacarpophalangeal joint was improved from an average of 30º hyperextension preoperatively to 15º flexion postoperatively. No patient improved from dependant to independent functioning. A side-effect was that although extensor pollicis longus to extensor pollicis brevis transfer supplemented metacarpophalangeal joint extension, it decreased extensor activity to the interphalangeal joint of the thumb. In five patients who had severe deformities the primarily goal was to improve cosmesis and hygiene. Appearance was improved in all five of them and lateral pinch was established in two.</td>
</tr>
</tbody>
</table>

**Notes**
- Allocation concealment: D

### Characteristics of excluded studies

- **Goldner 1990** Selected patients were not similar at baseline regarding the most important prognostic indications. Not all the patients in the study had spastic cerebral palsy. Twenty patients had athetosis.
- **Hoffer 1983** Evaluation was not primarily pre- and post-operatively, but comparison between three different groups (patients with total adductor release, patients with partial adductor release, and patients who were not operated on), was based...
Characteristics of excluded studies (Continued)

on pre-operative electromyography study. This was not included in this review. Outcome measures were considered not relevant.

Keats 1965 Patients were not evaluated identical pre- and post-operatively.

Tonkin 2002 Data in this manuscript were of the same patients as used in the included study of Tonkin 2001. For that reason, this manuscript was excluded for the review. However, it provided useful information about the inclusion and exclusion criteria of the patients and of surgical techniques. This information was included in the review.

Van Heest 1999 This study does not explicitly evaluate surgical interventions of thumb-in-palm deformity, but surgical interventions of deformities in upper limb cerebral palsy in general. Information of specific function of the thumb was lacking. Patients were not considered similar at baseline.

ADDITIONAL TABLES

Table 01. Methodological quality assessment scheme

<table>
<thead>
<tr>
<th>items</th>
<th>scores</th>
<th>notes</th>
</tr>
</thead>
<tbody>
<tr>
<td>(1) Was the assigned treatment adequately concealed prior to allocation?</td>
<td>3 = method did not allow disclosure of assignment. 1 = small but possible chance of disclosure of assignment or unclear. 0 = quasi-randomised or open list/tables.</td>
<td>Cochrane code (see Handbook): Clearly Yes = A; Not sure = B; Clearly No = C.</td>
</tr>
<tr>
<td>(2) Were the outcomes of trial participants who withdrew described and included in the analysis (intention to treat)?</td>
<td>3 = withdrawals well described and accounted for in analysis. 1 = withdrawals described and analysis not possible, or probably no withdrawals. 0 = no mention, inadequate mention, or obvious differences and no adjustment.</td>
<td></td>
</tr>
<tr>
<td>(3) Were the outcome assessors blinded to treatment status?</td>
<td>3 = effective action taken to blind assessors. 1 = small or moderate chance of unblinding of assessors, or some blinding of outcomes attempted. 0 = not mentioned or not possible.</td>
<td></td>
</tr>
<tr>
<td>(4) Were important baseline characteristics reported and comparable?</td>
<td>3 = good comparability of groups, or confounding adjusted for in analysis. 1 = confounding small, mentioned but not adjusted for, or comparability reported in text without confirmatory data. 0 = large potential for confounding, or not discussed.</td>
<td></td>
</tr>
<tr>
<td>The principal confounders considered were age, gender, type of fracture, type of treatment, existing co-morbidities (arthritis), prior functional and mental status, and complications.</td>
<td></td>
<td></td>
</tr>
<tr>
<td>(5) Were the participants blind to assignment status after allocation?</td>
<td>3 = effective action taken to blind participants. 1 = small or moderate chance of unblinding of participants. 0 = not possible, or not mentioned (unless double-blind), or possible but not done.</td>
<td></td>
</tr>
<tr>
<td>(6) Were the treatment providers blind to assignment status?</td>
<td>3 = effective action taken to blind treatment providers. 1 = small or moderate chance of unblinding of treatment providers.</td>
<td></td>
</tr>
</tbody>
</table>
Table 01. Methodological quality assessment scheme  (Continued)

<table>
<thead>
<tr>
<th>items</th>
<th>scores</th>
<th>notes</th>
</tr>
</thead>
<tbody>
<tr>
<td>(7) Were care programmes, other than the trial options, identical?</td>
<td>3 = care programmes clearly identical. 1 = clear but trivial differences, or some evidence of comparability. 0 = not mentioned or clear and important differences in care programmes.</td>
<td>Examples of clinically important differences in other interventions were: differences in treatment intervention (e.g. surgery, plaster cast; duration of immobilisation), differences in call back times for assessment, clinician experience and speciality.</td>
</tr>
<tr>
<td>(8) Were the inclusion and exclusion criteria for entry clearly defined?</td>
<td>3 = clearly defined (including type of treatment). 1 = inadequately defined. 0 = not defined.</td>
<td></td>
</tr>
<tr>
<td>(9) Were the interventions clearly defined (including who provided the care)?</td>
<td>3 = clearly defined interventions are applied with a standardised protocol and care providers identified. 1 = clearly defined interventions are applied but the application protocol is not standardised or care providers identified. 0 = intervention and/or application protocol are poorly or not defined.</td>
<td></td>
</tr>
<tr>
<td>(10) Were the outcome measures used clearly defined?</td>
<td>3 = clearly defined. 1 = inadequately defined. 0 = not defined.</td>
<td></td>
</tr>
<tr>
<td>(11) Were the outcome measures clinically useful - with adequate accuracy, precision and considerations of observer variation - including active follow-up?</td>
<td>3 = optimal. 1 = adequate. 0 = not defined, not adequate.</td>
<td></td>
</tr>
<tr>
<td>(12) Was the timing (e.g. duration of surveillance) clinically appropriate?</td>
<td>3 = optimal. (&gt; 1 year) 1 = adequate. (6 months - 1 year) 0 = not defined, not adequate. (&lt; 6 months)</td>
<td></td>
</tr>
</tbody>
</table>

**GRAPHS AND OTHER TABLES**

This review has no analyses.

**COVER SHEET**

**Title**  Surgical treatment for the thumb-in-palm deformity in patients with cerebral palsy

**Authors**  Smeulders M, Coester A, Kreulen M

**Contribution of author(s)**  M. Smeulders searched the databases, review the studies and wrote the protocol and review. A. Coester searched the databases and wrote part of the protocol. M. Kreulen reviewed the studies and wrote part of the review.