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Langenskiöld Procedure for Madelung's Deformity: Case Series of Late Sequelae

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Abstract

Background: Although many authors advise to postpone surgical intervention until skeletal maturity is reached, the Langenskiöld procedure—entailing physiolysis and the release of Vickers' ligament—is a known procedure in case of Madelung's deformity in children. Although advised in textbooks and recent reviews, the results and late sequelae of this procedure have been poorly documented. The aim of this study is to portray the late effects of this procedure for three patients and emphasize a very careful patient selection. Methods: A retrospective study was performed of the medical records of three patients who underwent a Langenskiöld procedure with release of Vickers' ligament for Madelung's deformity of the wrist. They were recalled for function evaluation and their results are presented in the current study. Results: We report the long term results of three patients (five wrists) who underwent a Langenskiöld procedure with release of Vickers' ligament in detail. Mean follow-up was twelve years (range: 11 - 14 years). In all of the operated wrists severe pain recurred due to destruction of the distal radio-ulnar joint. Further salvaging surgeries were necessary, some with highly dissatisfying results. Conclusions: Based on our experience we advise not to perform this procedure until further studies have proven its effectiveness and clear indications. Other techniques are available for later correction of Madelung's deformity, with similar results on pain relief, wrist function and cosmetics. However, we will advocate conservative treatment until skeletal maturity is reached.

Keywords

Madelung's Deformity, Treatment, Langenskiöld, Physiolysis, Vickers' Ligament

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1. Introduction

The deformation of the wrist causing subluxation of the hand was by no means a novelty in the 1800s [1] [2]. It had been described, amongst others, by Dupuytren (1834) [3] and Malgaigne (1855) [4]. However, Otto Wilhelm Madelung [5] was the first to make a complete and accurate description, earning the honour of having the deformation named after him [1] [6].

Madelung's deformity arises when the palmar and ulnar part of the distal radial physis fuse prematurely [7]-[9]. Continued growth of the radial part, while the ulnar part is tethered, results in a shorter and dorsoradial bowed radius. The ulna continues to grow and surpasses the radiocarpal articular surface, luxating dorsally. The carpals realign in a pyramidal shape to accommodate themselves to the deformed radius and ulna [1] [7] [10]-[14]. Often Vickers' ligament [9], an anomalous thickened ligament between the lunate and radius, can be found both on magnetic resonance imaging and preoperatively [15]-[17].

Patients with Madelung's deformity generally present themselves between the ages of nine and fourteen [1] [2] [6] [11]-[13]. It occurs predominantly in females [10] and bilaterally [1] [2] [6] [8] [12] [13] [15] [19]. During the growth spurt the deformation gradually worsens until the physis has closed completely [15]. It can be one of the presenting clinical features of dyschondrosteosis, also known as Leri-Weill syndrome [8] [10] [13] [15]. Symptoms include wrist pain, fatigue during strenuous activities and a displeasing cosmetic aspect of the deformed wrist [7] [15] [18]. The range of motion (ROM) of the wrist and forearm is impaired [1] [11] [12] [19]. Pain usually subsides once skeletal maturity is reached, but may arise again due to incongruity of the distal radio-ulnar joint (DRUJ) [2] [12]. As the complaints are generally of a passing nature, they can be treated conservatively [11] [12]. However, several surgical methods with varying results have been designed to correct the deformity. These can be grossly divided into three groups: 1) correction of the radius (the bowing and volar tilt of the articular surface) [20]-[22]; 2) correction of the ulna (the length and luxation of the ulnar head and the DRUJ) [23]-[25]; 3) combined correction of radius and ulna [14] [19] [25]-[28]. These techniques are generally applied at skeletal maturity.

Because Madelung's deformity is caused by premature fusion of the physis, treatment should theoretically be possible by resection of the affected part, allowing the radius to continue to grow in a normal direction [29]. Vickers and Nielsen applied this in the so-called "Langenskiöld technique": resection of the physis with fat-interposition to prevent recurrence of a bony bridge combined with the resection of Vickers' ligament [9] [30] [31]. However, literature describing the results of this procedure is scarce. Aside from the original article by Vickers and Nielsen [9] of 1992, we found only one additional study describing the Langenskiöld procedure [32]. In this study three wrists were subjected to a physiolysis with a free fat graft according to Vickers, revealing an improved outcome in two out of three cases after a mean follow-up of only 1.7 years (range: 1 - 2.5 years). In our clinic we found unfavourable results after this procedure in the long term. It is our aim to present an overview of our experiences with the Langenskiöld procedure for Madelung's deformity and make others aware of the late sequela of this procedure.

2. Case Report

Three patients (five wrists) with Madelung's deformity who presented between 1998 and 2002 at the University Medical Centre Utrecht, the Netherlands, underwent the Langenskiöld procedure with resection of Vickers' ligament (**Table 1**). All interventions were performed by the same surgeons (A.S. and R.S). All patients underwent this procedure during childhood after careful international consultation. Aim was to relief pain and allow for correction of the deformity through growth. A thorough retrospective study of their records and current function evaluations—consisting of ROM, pinch strength, Jamar isometric grip strength and Disabilities of the Arm, Shoulder and Hand (DASH) scores—were performed (**Table 2-5**) [33]-[36].

Case 1

This woman (age fourteen) presented with increasing pain of the wrists, left more than right. ROM at presentation was 70-0-60˚ dorsal/palmar flexion and 70-0-10˚ pronation/supination bilaterally. The patient was operated on both wrists within the following year. At one year follow-up the patient was free of complaints. However, five years postoperatively she returned with complaints of pain bilaterally. X-rays showed a post-Langenskiöld state with destruction of the lunate fossa bilaterally. This was treated conservatively. At eight years postoperatively she returned with complaints of functional impairment predominantly on the left side with unchanged radiographic images (**Figure 1(A)** and **Figure 1(B)**). An ulnar shortening osteotomy was performed on

Table 1. Surgical description of the Langenskiöld procedure.

Surgical Description of the Langenskiöld Procedure
Incision distal over the tendons of the long digital extensor muscle. Separation of the subcutaneous tissue between the extensor tendons onto the distal radius. The distal radius is exposed subperiosteal on the ulnar side. An osteotomy is performed of the distal radius on the ulnar side up to the abnormal physis. The epiphysis is left in place. The bone just proximal is removed until normal physis is seen over the entire width. Vickers' ligament is identified and released. The spongiose part of the radius is sealed with bone-wax, except the part close to the physis. The physis is covered using a fat-interposition from the subcutaneous fat. Closure is achieved in layers and a forearm-cast applied post-operatively.

Table 2. Patient characteristics.

	Case 1	Case 2	Case 3
Sex	Female	Female	Female
Affected side	Bilateral	Bilateral	Bilateral
Dominant side	Right	Right	Left
Age at presentation (years)	14	11	12
Age at Langenskiöld procedure(years)	Right: 15 Left: 14	Right: 12 Left: 12	Right: 12 Left: -
Follow-up (years)	11	14	11
Re-intervention	Left: ulna shortening osteotomy	Right: excision exostosis epiphysis Sauve-Kapandji procedure removal osteosynthesis material Left: Sauve-Kapandji procedure excision exostosis ulna arthrodesis radius, scaphoid and lunate resection of the distal part of the scaphoid complete wrist arthrodesis removal osteosynthesis material removal anterior interosseal nerve STT arthrodesis and resection ulnar stump	Right: ulnar shortening osteotomy Left: ulnar shortening osteotomy

Table 3. Post-operative wrist function (average 12 years post-operative).

	Case 1		Case 2		Case 3	
	Right	Left	Right	Left	Right	Left
Dorsal flexion	30˚	60˚	40˚	10˚	50˚	60˚
Palmar flexion	40˚	50˚	70˚	10˚	40˚	50˚
Radial deviation	10˚	15˚	10˚	0˚	15˚	10˚
Ulnar deviation	20˚	15˚	10˚	0˚	20˚	20˚
Pronation	60˚	60˚	60˚	60˚	30˚	80˚
Supination	45˚	45˚	60˚	60˚	50˚	80˚

the left side. She was recalled at eleven years after the Langenskiöld procedure. ROM was impaired in flexion/extension and pronation/supination (**Table 3**). Isometric grip strength was considerably less than average (**Table 4**). However pinch strength was good (**Table 5**) and a DASH score of 0.83 revealed this patient currently experiences little pain or trouble by her reduced ROM.

Case 2

This eleven year old patient presented with severe pain of the wrists since several months, left more than right. Documented ROM at presentation was 20-0-50˚ dorsal/palmar flexion and 50-0-20˚ pronation/supination

Table 4. Jamar isometric grip strength*.

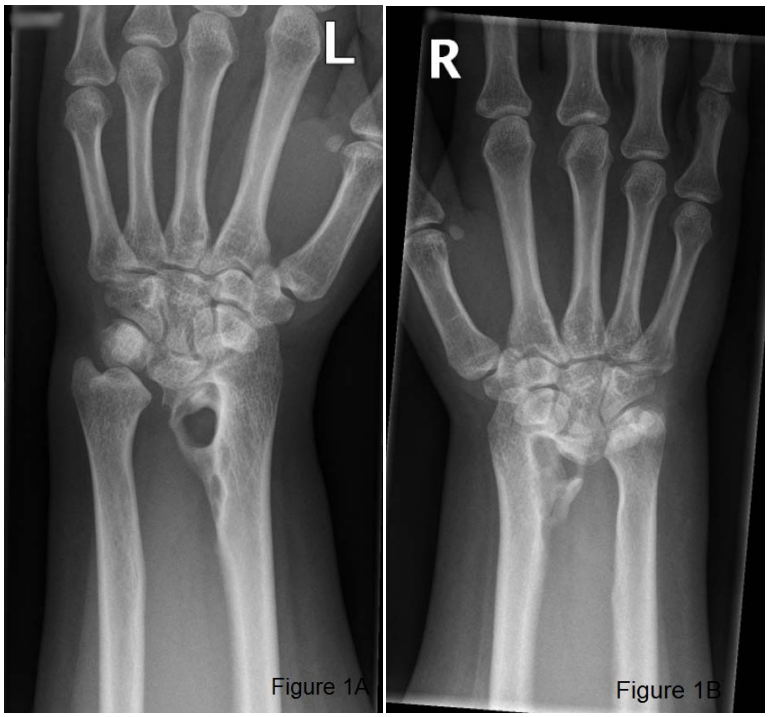
Side	Case 1		Case 2		Case 3	
	Right	Left	Right	Left	Right	Left
Score	13.33 kg (24.6 kg)	11.33 kg (22.4 kg)	0 kg (24.6 kg)	0 kg (22.4 kg)	14.67 kg (22.4 kg)	16.67 kg (24.6 kg)

*Isometric grip strength as measured with the Jamar dynamometer [34]-[36]; Reference value adjusted for sex and dominance.

Table 5. Pinch strength*.

Side	Case 1		Case 2		Case 3	
	Right	Left	Right	Left	Right	Left
Two-point	3.67 kg (3.6 kg)	4.00 kg (3.3 kg)	2.00 kg (3.6 kg)	2.00 kg (3.3 kg)	6.67 kg (3.3 kg)	5.67 kg (3.6 kg)
Three-point	5.00 kg (5.4 kg)	5.33 kg (4.6 kg)	2.33 kg (5.4 kg)	2.00 kg (4.6 kg)	7.00 kg (4.6 kg)	6.00 kg (5.4 kg)
Lateral	6.33 kg (4.4 kg)	7.00 kg (4.3 kg)	2.00 kg (4.4 kg)	2.33 kg (4.3 kg)	7.67 kg (4.3 kg)	6.33 kg (4.4 kg)

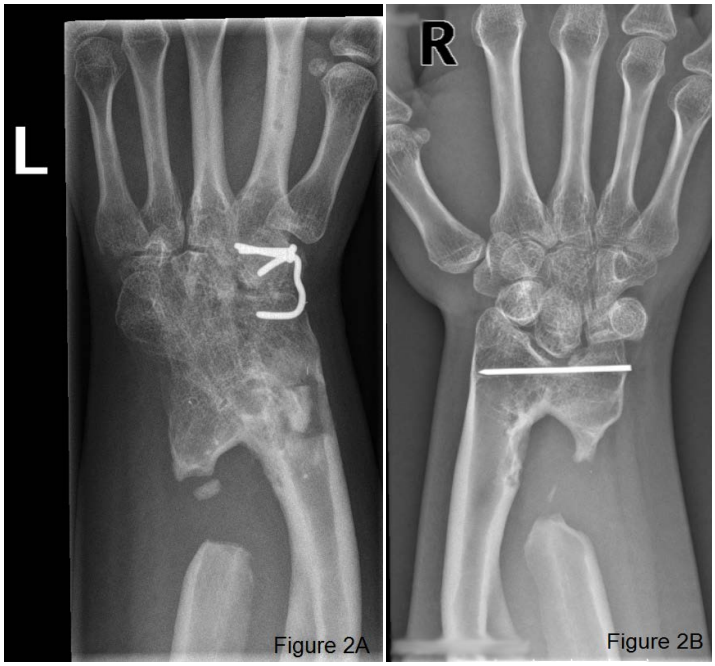
*Pinch strength: using a pinch gauge, strength of two-point pinch grip (thumb & index finger), three-point pinch (thumb, index & middle finger) and lateral pinch (thumb and lateral side index finger) is measured [34]-[36]; Reference values adjusted for sex and dominance.



(A) (B)

Figure 1. Conventional radiographic images 8 years post-operative. Destruction of the lunate fossa and proximal bone. The right wrist is more severely affected than the left. (A) Left wrist, AP view; (B) Right wrist, AP view.

bilaterally. After conservative treatment for half a year, pain increased and a Langenskiöld procedure was performed bilaterally. Within several months she complained of pain and restricted function. In the following years several operations were performed on both wrists (**Table 2**) due to painful arthritic changes (**Figure 2(A)** and **Figure 2(B)**). Eleven years after the initial Langenskiöld procedure she experiences severe pain of the left wrist and the left elbow, imaging revealing deformation of the proximal radial head. Function evaluation was performed at fourteen years post-Langenskiöld procedure (**Table 3-5**). A DASH score of 33.33 revealed she experiences trouble with actions requiring strength and is limited in her function.



(A) (B)

Figure 2. Conventional radiographic images. Left wrist: 11 years post-operative. Right wrist: 7 years post-operative. Bilaterally performed Sauve-Kapandji procedure. Complete arthrodesis of the carpals of the left wrist. (A) Left wrist, AP view; (B) Right wrist, AP view.

Case 3

This patient presented at the age of twelve years old, with a bilateral Madelung's deformity and complaints of pain predominantly on the right wrist. This was also the most affected side radiographically. Documented ROM at presentation was 70-0-60˚ dorsal/palmar flexion and 10-0-80˚ pronation/supination (right hand), 80-0-80˚ dorsal/palmar flexion and 90-0-70˚ pronation/supination (left hand). A Langenskiöld procedure was performed on the right side, and the left was treated conservatively. Six years post-operatively she returned with pain on both wrists. Due to our negative experiences in case 2, we chose to treat this conservatively with intensive physiotherapy. Interestingly, when performing McCarroll's measurements of the ulnar tilt, lunate subsidence and palmar carpal displacement [37]-[39] on the six year post-operative images (**Figure 3(A)** and **Figure 3(B)**), the deformity is still more severe on the operated wrist than the non-operated wrist (right: 43˚, 10mm and 31mm; left: 53˚, 9 mm and 24 mm). This could be interpreted that little to no correction had taken place after the Langenskiöld procedure. At ten years post-operative, the continuing pains of both wrists lead us to perform an ulnar shortening osteotomy bilaterally with good results in term of pain-relief. Function evaluation after eleven year is presented in **Table 3-5**. There was a DASH score of 20.0.

3. Discussion

In the present study we provide valuable long term follow-up outcome measurements after the Langenskiöld procedure. With the unsatisfactory results obtained after performance of this procedure, we would advocate a conservative treatment until skeletal maturity is reached.

Langenskiöld based his physiolysis technique on experiments performed on rabbits [30] [31]. They found that when a bony bridge in the physical plate is resected, recurrence can be prevented by interposition of autologous fat. The remaining physis was capable of partially regenerating the previously closed plate. Continued growth gradually corrected the deformity. This technique was successfully applied to the distal end of the femur and proximal and distal ends of the tibia in children [31]. However, the technique was applied to a physis which had closed prematurely due to trauma and was otherwise capable of normal growth. In Madelung's deformity the physis is not normal and therefore one could question the applicability of the theory. Operating the ulnar side of the radius might result in earlier DRUJ degeneration than other procedures or than naturally occurs in Made

(A) (B)

Figure 3. Conventional radiographic images 6 years post-operative. Left non-operated wrist compared to right operated wrist: similar degrees of deformity. DRUJ destruction in the operated wrist. (A) Left wrist, AP view; (B) Right wrist, AP view.

lung's deformity [12]. The concurring pain necessitates further surgeries on an already deformed wrist.

The procedure as applied to Madelung's deformity was described in detail by Vickers and Nielsen in 1992, documenting the results of eleven patients (fifteen wrists, age: 11-14.5 years, follow-up: 15 months - 12 years) [9]. After surgery all patients noted a decrease in pain and ROM improvement. However, only four patients were completely pain-free after the procedure and the ROM gained was gradually lost over time by some of their patients[9]. The lack of literature on the late effect of the Langenskiöld procedure in Madelung's deformity is striking. Some authors may mention having used the technique [20], and recently the results of two patients (three wrists) were reported with a follow-up of 1 - 2.5 years [32]. No additional articles record the long-term results. Still, this technique is advised as a procedure for young patients in Green's Operative Hand Surgery [8] and recent reviews concerning Madelung's deformity [6] [15].

Although no other studies report negative results of this procedure, rumour has it others have experienced similar problems and have stopped performing this procedure. Therefore we strongly encourage others to publish their results.

A plethora of other operative techniques have been described in literature to treat Madelung's deformity, each with their own objectives and results. McCarroll *et al*. advise a distal radial osteotomy combined with an ulnar shortening osteotomy [18]. Others prefer a dome osteotomy of the distal radius, combined with a form of ulnar resection for those with DRUJ osteoarthritis [8]. Small studies describe a complete arthroplasty of the wrist [40]. We have performed a fairly simple ulnar shortening osteotomy to patients' satisfaction. Most authors prudently advise conservative treatment [6] [8], and to reserve surgical intervention for extreme cases [6] [8] [12] [15].

In our opinion it would be advisable to remain conservative until skeletal maturity is reached, reserving sur-

gical intervention for those patients whose pain does not subside after the growth spurt [15] [19] [22]. Due to the dissatisfying long term results of the Langenskiöld procedure we no longer perform this intervention.

4. Conclusion

The Langenskiöld procedure, entailing physiolysis and the release of Vickers' ligament, is one of the surgical options for the treatment of Madelung's deformity in childhood. Although advised in textbooks and recent reviews, the results and late sequelae of this procedure have been poorly documented. Based on our own experiences and dissatisfying long term outcome after the Langenskiöld procedure, which is illustrated in three cases (five wrists) after a mean follow-up of twelve years in the current study, we advise not to perform this until further studies have proven its effectiveness and clear indications.

Informed Consent

We state that each patient was informed that data concerning the case would be submitted for publication and that each patient agreed by signing a consent form.

All authors state that there is no conflict of interest.

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