



LUND UNIVERSITY

Congenital thumb anomalies and the consequences for daily life patients' long-term experience after corrective surgery. A qualitative study

Carlsson, I. K.; Dahlin, L. B.; Rosberg, H. E.

Published in:
Disability and Rehabilitation

DOI:
[10.1080/09638288.2016.1243159](https://doi.org/10.1080/09638288.2016.1243159)

2018

Document Version:
Peer reviewed version (aka post-print)

[Link to publication](#)

Citation for published version (APA):
Carlsson, I. K., Dahlin, L. B., & Rosberg, H. E. (2018). Congenital thumb anomalies and the consequences for daily life: patients' long-term experience after corrective surgery. A qualitative study. *Disability and Rehabilitation*, 40(1), 69-75. <https://doi.org/10.1080/09638288.2016.1243159>

Total number of authors:
3

General rights

Unless other specific re-use rights are stated the following general rights apply:
Copyright and moral rights for the publications made accessible in the public portal are retained by the authors and/or other copyright owners and it is a condition of accessing publications that users recognise and abide by the legal requirements associated with these rights.

- Users may download and print one copy of any publication from the public portal for the purpose of private study or research.
- You may not further distribute the material or use it for any profit-making activity or commercial gain
- You may freely distribute the URL identifying the publication in the public portal

Read more about Creative commons licenses: <https://creativecommons.org/licenses/>

Take down policy

If you believe that this document breaches copyright please contact us providing details, and we will remove access to the work immediately and investigate your claim.

LUND UNIVERSITY

PO Box 117
221 00 Lund
+46 46-222 00 00

1
2 Congenital thumb anomalies and the consequences for daily life: patients'
3 long-term experience after corrective surgery. A qualitative study.

4
5 Running Title: Congenital thumb anomalies – consequences for daily life.

6
7 Carlsson IK, Dahlin LB, Rosberg H-E

8 Dept Translational Medicine – Hand Surgery

9 Skane University Hospital, Lund University, Sweden

10
11
12
13 Correspondence to

14 Ingela Carlsson

15 Dept Translational Medicine – Hand Surgery

16 Skane University Hospital, Lund University

17 S-205 02 Malmö, Sweden

18 Phone +46-40-33 65 71, Fax +46-40-33 70 65

19 e-mail: ingela.k.carlsson@hotmail.com

20
21
22 Keywords: Thumb hypoplasia/aplasia, thumb duplication, pollicisation, hand
23 disfigurement, adaptation, qualitative research

ABSTRACT

Purpose: The aim of the study was to explore patients' long-term experience of a congenital hand problem, and the consequences for daily life.

Method: Fifteen participants with a median age 24 years (17-55), born with thumb hypoplasia/aplasia or thumb duplication were interviewed using a semi-structured interview guide. The interviews were subjected to qualitative content analysis.

Results: Although the mobility and strength in the thumb/hand(s) varied within the group, hand function was generally described as good. Compensatory strategies were used to overcome practical obstacles. The emotional reactions to being visibly different from peers in early life varied from total acceptance and a sense of pride in being special, to deep distress and social withdrawal. Support from parents, teachers and others was important in facing emotional challenges and practical consequences.

Conclusion: The present study highlights the importance of healthcare professionals addressing appearance-related concerns which may have long-term emotional and social consequences for patients born with a thumb anomaly.

46 **Implications for Rehabilitation**

- 47 · Appearance related concerns and need for emotional support should be fully
48 considered throughout the rehabilitation process to prevent distress and social
49 withdrawal
- 50 · Effective problem-solving strategies, such as compensation, change in occupational
51 performance and support from others may reduce activity limitations and participation
52 restriction.

53

INTRODUCTION

Congenital malformations in the upper extremities, with an overall reported incidence of 21.5/10 000 live births [1], may have serious consequences for the individual patient.

A variety of such malformations involve the thumb, causing possible functional disturbances of varying severity as the thumb is crucial for proper hand function.

Thumb hypoplasia/aplasia, with a “non-functioning”, thumb represents a surgical challenge and usually requires a pollicisation procedure, while thumb duplication poses minor surgical and functional problems in the hand.

Earlier reports have mainly focused on quantitative research, such as technical issues, functional outcome and subjective assessment of appearance, in both thumb hypoplasia and thumb duplication [2-5]. However, long-term satisfaction and well-being in patients with e.g. thumb hypoplasia/aplasia and thumb duplication may depend on several other factors such as emotional stress related to functional limitations or the experience of being visibly different [6]. Such emotional stress throughout childhood has been reported to affect a majority of children with congenital hand differences and may not always be recognized by parents or caregivers [6]. Access to effective coping mechanisms to counteract stress factors when facing challenges in daily life may also vary. It is therefore important to further illuminate the long-term consequences in daily life for this patient group.

A qualitative approach with open questions may deepen the understanding of how patients perceive and handle such consequences in daily life [6,7]. Our aim was to explore the patient’s experience of a congenital hand problem, focusing on thumb hypoplasia/aplasia and thumb duplication, and the long-term consequences for daily life, including emotional and social aspects as well as impact on activity and

participation. We were also interested in personal qualities and the support needed to achieve positive adaptation.

METHOD

Design and Participants

A qualitative descriptive method with an inductive approach was used to reach a deeper understanding about the consequences for the individual of being born with a congenital hand problem. In total, 15 individuals were included and interviewed, seven with a thumb hypoplasia and eight with a duplicated thumb. The sample size was determined by the role of redundancy, meaning that the last four interviews provided little new information [7].

All participants were patients admitted to the Hand Surgery Clinic, Skane University Hospital, between the years 1978-1998 who could be interviewed (i.e. had no serious mental, cognitive, and or linguistic impairments). Twenty-two patients were eligible for inclusion. Seven patients were not included as they declined participation due to lack of time or living too far away or could not be reached. The thumb duplications included were of the Wassel IV [8] and the thumb aplasia was Type IIIB, IV or V [9,10] and all had had previous surgery. Five of the patients with thumb hypoplasia had undergone a pollicisation procedure, two had tendon transfers and one had only removal of the rudimentary thumbs [11]. The patients with thumb duplication had surgery as described elsewhere for this type [12].

The median age was 24 years (17-55). All patients except one had finished high school, four were in secondary education and three were completing their university studies.

One patient was unemployed and seven patients worked in a variety of professions, such as sales, economy, project management, professional driving, restaurant work and healthcare professions. Six participants lived with their parents, five had a partner and four had a family with children. Seven men and eight women participated. Table 1. Prior to the interview the participants completed the Swedish versions of several self-report questionnaires; the QuickDASH [13], the condensed 13-item Sense of Coherence (SOC) [14], the acute version of the Short Form 36 (SF-36) questionnaire [15], the Cold Intolerance Symptom Severity (CISS) questionnaire [16,17] and seven single questions (numeric rating scale 0-100, 0 representing no problem and 100 worst possible problem) concerning pain at rest, pain during motion without load, pain with load, grip function, fine motor skill, weakness and appearance of the hand. The interviewer also measured grip strength, key pinch and pinch strength [18,19]. Table 2.

Procedure and ethics

Written information was sent to the participants emphasizing the voluntary nature of the study. The first author then contacted them and arranged an interview time for those who agreed to participate. Written consent was obtained in conjunction with the interview and all participants were informed about how the data would be analysed and were assured of confidentiality. The collection of all data was conducted by the first author not previously involved with the participants care. The study was performed in accordance with the ethical guidelines stated in the Helsinki Declaration and the Swedish Act Concerning the Ethical Review of Research Involving Humans (SFS:2003:460) and approved by the local ethics committee of Lund University (Dnr 2009/339). All interviews were performed and tape-recorded by the first author in a

quiet room at the clinic and lasted between 20 and 66 minutes. The interview started with a repetition of the aim of the study. A semi-structured interview guide with open questions was then used and the participants were asked to describe their overall thumb/hand function, pain, appearance, emotional and social consequences, personal qualities, support from others and the impact on activity and participation. Follow-up questions were asked such as: How did you experience that? How did you handle that? Can you describe that in more detail? A secretary, marking nonverbal expressions, then transcribed all the interviews verbatim. All transcripts were checked for accuracy by the first author, who also translated the quotations from Swedish into English. The translations were verified by the last author.

Data analysis

The text was read and reread by the first and last author and subjected to qualitative content analysis [20,7]. The analysis started with a naive reading of each interview to gain a general impression of the content. Meaning units, described as words or sentences related to each other through their content and related to the aim of the study, were then identified. The impression of the text was discussed and the selected meaning units compared. The meaning units were shortened into codes (labels for the meaning units) and then grouped into categories. Within each category similar statements were analysed critically and questioned, then read and compared until a reasonable interpretation was reached. The categories were then discussed with the second author and adjustments were made to ensure that the categories covered all aspects in the text. Finally, the categories were compared with the text and with each other. The second author read seven randomly selected interviews and reviewed the different codes and

categories. Concerning the authors' pre-understanding, the first author is an experienced occupational therapist specialized in hand rehabilitation; the second and last authors are experienced hand surgeons. All three authors work in a specialized unit. Both the first and the second authors are experienced in qualitative research methodology [21-24].

RESULTS

Consequences for hand function.

Even though the mobility in the affected thumb(s) varied among participants they described normal sensibility and overall good hand function. *"I have not been limited – there is just one finger less..." (Participant 12)*. However, situations requiring fine motor skills and dexterity could be troublesome, e.g. when undoing a necklace or putting on earrings. Participants who had undergone a pollicisation procedure also described a sense of weakness and tiredness requiring the use of modified grip patterns using other fingers or the unaffected side or both hands. Depending on the muscle strength, those born with duplicated thumbs used similar habitual strategies. *"It works well; I was born with the problem so I have learned to compensate by using both hands when needed"*. (Participant 10).

With age, hand function and dealing with everyday challenges were considered increasingly important compared to a visibly different appearance.

Pain experience

Pain at rest was uncommon, but hitting the base of the thumb or carrying something heavy could trigger pain. This was described as an increasing pain, a sense of strain

rather than actual pain or as if the lack of muscles enabled the pain to continue deep inside, triggering numbness and an ache in the bones. *“The whole hand explodes and then it feels a little numb...”* (Participant 1). The pain could be unbearable, like having a knife or a nail in the hand and, if severe, affecting sleep. A premonition of pain was also connected to locking within the wrist, or present in the unaffected hand when it was overused and when the other hand worked mainly as a support hand. Compensatory grip patterns and use of the whole body, technical aids or simply enduring the pain were strategies used. *“I squeeze my hand but the hand doesn’t listen. I try to massage the hand to alleviate the pain.”* (Participant 9). For those with bilateral anomalies and constant pain in both hands the possibility of using an unaffected side to compensate was lost, which caused distress and worries for the future. Re-education and occupational changes was therefore necessary. *“In the morning I go straight to the medicine (paracetamol) and in the evening and four times daily. I can’t cope. It is frightening.”* (Participant 14). Exposure to cold caused pain, stiffness, numbness and a change in skin colour for those affected and was experienced as a numb feeling deep inside, as if the thumb was lost and had been cut off.

Consequences for activity and participation

When grip function, fine motor skills and weakness were affected the participants described fruitful strategies for overcoming challenges, and engagement in activities and participation in society were realizable. In school, writing could cause pain in the thumb and required the use of specially adapted pencils or breaks. This was especially apparent under time pressure and in examinations, both at younger ages and during university education. Various activities in physical education (gymnastics), such as

198 hand-, basket-, or volley-ball, push-ups, cartwheels, turning upside down/handstands,
199 grasping a bar, could trigger pain or cramps or were impossible for some because of
200 reduced strength. In textile handicraft the ability to handle a needle and thread required
201 dexterity skills and small needles were sometimes difficult to grip because of reduced
202 strength. Supportive and understanding teachers were important in finding solutions or
203 alternative activities.

204 Activities at home, such as holding on to a broom or vacuum cleaner, opening a jar,
205 peeling potatoes, using cutlery or scissors, holding a glass or unbuttoning a pair of jeans
206 could, cause pain. A modified grip, use of the other hand or both hands, technical aids
207 or asking for help were strategies used to overcome obstacles.

208 Several leisure activities were possible to pursue and caused no problems at all. *"I play*
209 *the French horn without any problems since you don't use the thumb while playing."*
210 *(Participant 10). "I play the trumpet – I hold it with my left hand and play with the other*
211 *hand."* *(Participant 11). "I have learned to use my right hand in a different way when I*
212 *play with the X-box, because it's developed for five-fingered hands."* *(Participant 1).*

213 However, there was also examples of the opposite, e.g. bowling put too much strain on
214 the thumb, ball sports was painful when hitting the ball, playing goal keeper in soccer
215 required too much strength and the desire to play a violin had to be abandoned in favour
216 of an accordion where a modified grip pattern enabled the person to make music.

217 *"It is not easy to go to a fitness centre to work up your strength – they don't understand.*
218 *You are at the level of minus ten and need help to arrive at zero and the exercises are*
219 *too heavy..." (Participant 14).*

220 The career choices for those of working age were not at all or only to some extent
221 influenced by activity limitations. An altered performance, re-education or change of

222 work was alternatives followed. *“I am studying to become a chef and I can do*
223 *everything that the others do and I do it as well as them.”* (Participant 1). *“I had my*
224 *own tricks when turning a patient around in the bed – I rolled up the draw sheet a lot*
225 *underneath my hand and then I used my whole body and pulled.”* (Participant 7).

226

227 **Appearance - emotional consequences and social acceptance**

228 The views on the aesthetics of the thumb varied from a feeling of looking nice and
229 almost normal to ugly, scarred and misconfigured. The latter was especially
230 troublesome in childhood/adolescence, but eased during high school. *“Why don’t I look*
231 *like everybody else?”* (Participant 3). Appearance-related concerns were expressed both
232 by those with minor or more visible disfigurements. The choice to preserve a four-
233 fingered hand and not proceed with a pollicisation procedure arose from the wish to
234 look “whole/complete”, pretending that there was a thumb hidden in the palm and
235 thereby looking like everyone else. It was all about hiding and, in the worst scenario,
236 even disappearing as a person. Habitual solutions were to hide the hand in a pocket,
237 wear long-sleeved sweaters or pull jacket sleeves down. Responding to questions in the
238 classroom by raising one’s hand was avoided because then the deformity became
239 visible. This could affect grades in early school years. *“...sometimes I greet people and*
240 *then I hide my hand quickly so no one can tell I’m different.”* (Participant 9). *“Socially,*
241 *it affects me when shaking hands, because my hand is smaller and a little weird, but no*
242 *one notices, it’s all in my head.”* (Participant 1). *“It has become a habit that I feel*
243 *ashamed of it – everybody else has ten fingers.”* (Participant 13).

244 Intrusive or hurtful comments, such as your thumb looks like an “ape’s thumb”, what a
245 “weird thumb” you have or simply being called “CP” (person with cerebral palsy) were

246 difficult to handle as a child. The appearance of the hand gave rise to verbal teasing or
 247 even bullying and caused insecurity, distress and social withdrawal.

248 *“Schoolmates in primary school commented my thumb saying - You can only count to*
 249 *nine and things like that, and it makes you think – What is wrong with me?”*
 250 *(Participant 12).*

251 *“ I wake up, looking at my hand, thinking – what a monster I am, remembering how I*
 252 *was bullied in school and called a four-fingered freak. I had to rename myself E.T. to*
 253 *save my psyche/soul... “I was very lonely, had no friends so I played with an imaginary*
 254 *friend – in one respect I had two kinds of personalities...” (Participant 9).* Choosing
 255 friends carefully before opening up was a lesson learned and comparing yourself with
 256 someone less fortunate or playing down the importance of appearance were helpful
 257 when dealing with emotional consequences. *“My father and I met another patient in*
 258 *rehab who had the same problems in both hands and for me that was like, shit, I could*
 259 *have had it in both hands. I was lucky, it could have been worse; I still have one hand*
 260 *that works.” (Participant 1).*

261 Others experienced the opposite, feeling rather cool and special because of the thumb(s)
 262 or having a hand closer to normal. Schoolmates also thought it was interesting and
 263 exciting being born with e.g. duplicated thumbs. Hiding the hand was considered to
 264 draw more attention to it. To “have ones place” by being physically stronger/bigger
 265 instilled some fear “in” schoolmates and counteracted possible teasing or bullying.
 266 Peers rarely made negative comments.

267 *“My mates thought it was a cool thing – it made me proud.” (Participant 4).* *“I have*
 268 *never thought about it as a misconfiguration in the hand because it has always been*
 269 *gone.” (Participant 6).*

270 Consequently, the emotional reactions to appearance varied from total acceptance and a
271 sense of pride in being special to deep distress and social exclusion. The bullying
272 stopped in secondary school, but earlier memories could create emotional scars resulting
273 in habitual hiding of the hand even as adults and especially among strangers. However,
274 comments referring to the smaller number of fingers on the hand could be used as
275 motivation to achieve excellent results in mathematics and with age this type of remark
276 was also considered mentally strengthening. As adults it was easier to deal with
277 whispered comments with confidence: *“Talk to me instead”*, and suddenly a new insight
278 emerged – *“I decided to put jewellery on my fingers – why should I hide myself*
279 *anymore.” (Participant 14).*

280

281 Approaching the other sex could be scary, especially during adolescence, even if treated
282 with respect and empathy, but with time it also became natural since being born with
283 e.g. duplicated thumbs was part of that person’s life story and was eventually accepted
284 internally. Initially, there were examples of a pattern of concealment of the disfigured
285 hand and a fear of being rejected, but the total acceptance and emotional support from
286 partners/spouses seemed to bolster confidence and self-esteem. *“When it comes to boys*
287 *I think a lot about, is he going to continue talking to me when he knows about the hand,*
288 *how will he react – it worries me a little, however, most guys seem to accept it.”*
289 *(Participant 12).*

290 *“A girl I met held my hand and felt that something was different but she didn’t care, she*
291 *just squeezed my hand tight, calmed my fear, accepted it, and then I felt that my self-*
292 *confidence came back to me knowing that girls could like me despite my hand.”*
293 *(Participant 9).*

294

295 **Support and personal characteristics**

296 The support from parents, grandparents, relatives and teachers was important when
297 facing emotional challenges and practical consequences, although support was not
298 considered a necessity for everyone. *“My father’s support has meant a great deal to me.
299 I told him what kind of problems I had and then we solved them together.”* (Participant
300 9). *“I have not felt the need for support – I have managed on my own.”* (Participant 2).
301 Teachers or parents gave helpful hints about compensatory measures, such as adapted
302 pencils, and how to tie shoelaces with the middle and ring finger. In school it could be
303 helpful to receive copies or assistance with writing especially after surgery when
304 strength was reduced and dexterity impaired, although it was also important for personal
305 development to manage by one’s self and not be receiving help all the time. Caring
306 parents spoke about playing down the importance of appearance, looking at the bigger
307 picture, looking outside the box and other words of wisdom.
308 *“The gym teacher knew about my hand and he was very helpful and understanding, he
309 took me aside and talked, not in front of everybody else because that is embarrassing
310 and he encouraged me to tell him if I had any problems and he would support me. I
311 didn’t feel any pressure or fear in gymnastics, I thought it was fun, it was my favourite
312 lesson.”* (Participant 1).

313 When dealing with practical and emotional challenges it was important to be self-
314 confident, positive, persistent, hard-bitten, creative, motivated and patient. To ignore
315 negative comments from schoolmates and to follow one’s own path was a fruitful
316 strategy. The strength to set limits when insulted was something that gradually
317 developed with time. Humour could be a redemptive strategy in uncomfortable

situations. *"I try to be funny/hilarious especially in uncomfortable situations; to joke about it helps..." (Participant 1).* All the experiences through life were considered meaningful in retrospect and were described as *"...an enormous resource that I can use to help other people."* (Participant 9).

It was important for healthcare professionals to consider the psychological aspects of feeling different from others and the need to talk about it. Since the surgical procedures were usually completed at an early age, our respondents stressed the importance of talking directly to the child in a calm and reassuring way. Information about realistic expectations concerning appearance-related improvements was also cited as important as well as informative leaflets that would be useful in school.

Advice to future patients

Based on their own experiences a wealth of advice to future patients was expressed: find your own way and go forward; learn to live with it, try to accept it and make the best of it; don't let it hinder you; do the things that feel best for you and not what pleases others; be yourself and nobody else; set your own goals; if you can't manage, don't be afraid to ask for help; live a normal life and don't worry about the future. *"Your hand is just like an ordinary hand, only a little weaker - you should feel special. If someone tells you that you can't then try to prove that you actually can manage, it's all about proving that you want it and having support from family..."* (Participant 13).

DISCUSSION

Patients' long-term experiences of being born with a congenital malformation in the upper extremity (i.e. thumb hypoplasia/aplasia or thumb duplication) were highlighted in this study. Although the mobility and strength in the thumb/hand(s) varied within the group, hand function was generally described as good. Compensatory strategies were frequently used to overcome practical obstacles caused by impaired fine motor skills/dexterity, weakness or pain. The emotional reactions to being visibly different from peers during early life varied from total acceptance and a sense of pride in being special, to deep distress and social withdrawal. The support from parents, teachers and others was important when facing emotional challenges and practical consequences.

The most striking finding in our study was the long-term psychosocial effect of being visibly different, although it was not a problem for all respondents. Appearance is important for an individual's self-concept and affects the way we are perceived and treated by peers, teachers and others [25]. The severity or extent of a disfigurement is not always related to the degree of emotional distress. A person's perception of how noticeable their difference is to others is a more relevant predictor, particularly if the face and hands are involved [26,27]. Hands are difficult to conceal, always on show both to the child and to those with whom the child interacts. Hiding the affected hand(s) in pockets or wearing long-sleeved sweaters/jackets was a strategy frequently resorted to by the respondents and in the worst scenario it even brought about a wish to disappear as a person. This habitual pattern was used both in school and in other social contexts, especially during childhood and adolescence, but for some it extended even

into adulthood, despite the mildness of the visible difference. Andersson et al [28] pointed out that children with milder disabilities may be at a higher risk of concealing their hand and this may contribute to a poor self-concept. One explanation for this might be that the hands appear closer to normal and therefore less support or sympathy is received compared to children with severe visible deformities. It is impossible for these children to hide the disability, they are likely to experience negative reactions more consistently and the process of acceptance and effective ways of coping may therefore start earlier on. This may lead to comparable self-concepts and a self-esteem as high as that of healthy children [27-29]. The negative consequences, including social withdrawal, for some of our participants is in line with earlier reports of lower social functioning among patients with congenital hand differences, especially in older children when negative self-sense and stigma may be perceived [29].

The support given by parents or close relatives was described and highly valued, however, being able to talk to someone else about the psychological aspects of feeling different from others was also expressed. According to Lukash, 2002, children cannot adequately express their feelings about physical issues that may affect them emotionally [30]. Being exposed to teasing at the vulnerable age of six or seven, when acceptance from peers is of the utmost importance, may be overwhelming for the child [31]. At this age children may not have access to the problem-solving skills needed for effective coping and the child's reaction to negative comments may trigger and reinforce patterns of bullying [31]. The opportunity to receive professional help to deal with negative comments from peers and thereby bolster one's self-esteem may have facilitated positive coping skills in our participants.

390 Maddern et al pointed out that a child's self-concept, including perceptions, beliefs,
391 feelings, attitudes and values about themselves, is more likely to be healthily maintained
392 if parents and teachers adopt a positive attitude towards the disfigurement [32]. It is also
393 well known that parental responses to a child's physical problem play a significant role
394 in their social well-being [31]. If comments or questions about the child's disfigurement
395 cause the parents distress, the child may feel that this is too serious a matter for their
396 parents to discuss, control and contain [31]. This was not an issue covered in depth with
397 our respondents, but an overall impression was that the support and security obtained
398 from parents, relatives and teachers was important for them to play down the
399 importance of appearance, look at the bigger picture and look outside the box. This
400 support facilitated emotion-, and problem-based coping mechanisms.

401 Some of our participants did not express any appearance-related concerns. Appearance
402 was not an issue since the hand did not differ much from normal, or even if that was the
403 case they expressed a strong self-image and described a sense of pride in being special.
404 Negative comments from peers or others rarely occurred and social interaction was
405 therefore less of a problem. Hiding the hand was considered counterproductive, drawing
406 even more attention. To minimize the importance of appearance, by e.g. comparison
407 with patients with a more visible disfigurement, had a positive affect on well-being and
408 constituted a strategy found in other studied patient groups [33]. Whether the stronger
409 resilience within these respondents is because of a personality trait enabling them to
410 access effective coping strategies, or access to social networks and other factors
411 strengthened their coping even more remains to be explored. But, as pointed out by
412 Joachim, 2000, the state of being stigmatized is more likely to depend on how the
413 individual chooses to see her/himself and not on the degree of disability [34].

In addition to its functional importance, the hand also has a psychological and social meaning and is an instrument for sensory exploration and intimate contact [30]. During adolescence appearance becomes increasingly salient. The impact of a malformation of the hand, burn injury, skin disorder or cleft lip on romantic relationships and on the enjoyment of an intimate relationship may be an additional concern in a challenging period of life but is rarely studied [35]. However, a fear of being rejected was expressed by some of our respondents but the acceptance received from partners instilled confidence and self-esteem.

Although some impact on activity and participation was described, due to reduced strength, pain and grip function, the participants managed to compensate or overcome many practical obstacles by using effective problem-solving skills. Furthermore, an inner strength and support system facilitated effective problem- and emotion-based coping, as described by other groups of patients with hand injuries [22,36].

Methodological considerations

In qualitative research, the findings are evaluated in terms of trustworthiness, which includes establishing credibility, dependability, confirmability and transferability [20,7]. Purposive sampling was used to provide variation in age, gender and cause of congenital thumb anomaly. The interviews varied in length and depth, but were rich in detail. The last four interviews provided little new information which ensured saturation of data [20]. To strengthen dependability, three authors independently read the text and engaged in in-depth discussions to arrive at a reasonable interpretation. Representative quotations from the transcribed text are given to make the interpretation of the text visible to the reader. Constantly confirming and clarifying information during the

interviews ensured confirmability. All the researchers participated in the analysis in order to reduce the risk of over interpretation of the results due to the authors pre-understanding of the phenomena in focus. Furthermore, the method used consistently throughout the research process was analysis focused on the text, limiting the risk of predetermined interpretation. The transferability of the findings may be limited to patients experiencing appearance related concerns because of diseases or trauma in the upper extremity.

Implications for provision of care

This study shows that the impact of a congenital hand anomaly on daily life varies. This applies both to dealing with practical challenges and emotional consequences. Access to support and effective coping strategies for those affected is vital. It is important to note that healthcare professionals need training in how to deal with appearance-related concerns. A pathological approach that uses negative terminology, such as defect, deformity, abnormality and disfigurement, may exacerbate the difficulties experienced by those seeking help [33,37]. A normalising approach for patients who are visibly different may instead empower them and promote adaptive behaviour in social situations. Some of our respondents indicated a need for professional assistance when dealing with emotional consequences. The opportunity to meet other patients and share experiences as well as having access to informative leaflets that would be useful in school were also mentioned. Maddern et al show that cognitive behavioural therapy (CBT), including social skills and problem-solving strategies, represents an effective therapy for children with appearance-related problems due to congenital abnormalities such as cleft lip and palate, burns or other forms of trauma. A reduction in the frequency

of teasing and in the degree of distress it caused both in the classroom and in the playground was seen at a six-month follow-up [32].

In conclusion

This study allows a deeper understanding of how being born with a visibly different or missing thumb (thumb hypoplasia/aplasia and thumb duplication) may influence daily life. The findings emphasize the importance for healthcare professionals addressing appearance-related concerns, which may cause long-term emotional distress and social consequences.

Acknowledgment

The authors particularly thank the patients who shared their narratives.

Declaration of interest

The study was supported by grants from the Swedish Medical Research Council, Lund University, Region Skåne, i.e ALF-grants; Health-Care Academy at Skane University Hospital; HKH Kronprinsessan Lovisas Förening för Barnsjukvård; Fanny Ekdahls stiftelse för Pediatrisk forskning; Kockska Stiftelsen and Svenska Läkaresällskapets fonder. The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

ORCID

I. K. Carlsson <http://orcid.org/0000-0002-2746-8346>

L. B. Dahlin <http://orcid.org/0000-0003-1334-3099>

H.-E. Rosberg <http://orcid.org/0000-0001-9229-2319>

REFERENCES

1. Ekblom AG, Laurell T and Arner M. Epidemiology of congenital upper limb anomalies in 562 children born in 1997 to 2007: A total population study from Stockholm, Sweden. *J Hand Surg* 2010;35:1742-54.
2. Aliu O, Netscher DT, Staines KG, Thornby J and Armenta A. A 5-year interval evaluation of function after pollicization for congenital thumb aplasia using multiple outcome measures. *Plast Reconstr Surg* 2008;122:198-205.
3. Netscher DT, Aliu O, Sandvall BK et al. Functional outcomes of children with index pollicizations for thumb deficiency. *J Hand Surg Am* 2013;38:250-7.
4. Patel AU, Tonkin MA, Smith BJ, Alshehri AH and Lawson RD. Factors affecting surgical results of Wassel type IV thumb duplications. *J Hand Surg Eur* Vol 2013; 39:934-43.
5. Staines KG, Majzoub R, Thornby J and Netscher DT. Functional outcome for children with thumb aplasia undergoing pollicization. *Plast Reconstr Surg* 2005;116:1314-23.
6. Franzblau LE, Chung KC, Carlozzi N, Chin AY, Nellans KW and Waljee JF. Coping with congenital hand differences. *Plast Reconstr Surg* 2015;135:1067-75.
7. Berg BL. *Qualitative research methods for the social sciences*. 5th Edn. Boston: Pearson Education Inc, Allyn and Bacon, 2004;269-70.
8. Wassel HD. The results of surgery for polydactyly of the thumb. A review. *Clin Orthop Rel Res* 1969;64:175-93.

- 511 9. Kozin SH. Upper-extremity congenital anomalies. J Bone Joint Surg Am Vol
512 2003;85: 1564-76.
- 513 10. Manske PR, McCarroll HR, Jr. and James M. Type III-A hypoplastic thumb. J
514 Hand Surg 1995;20:246-53.
- 515 11. Kozin SH. Pollicization: The concept, technical details, and outcome. Clin
516 Orthop Surg 2012;4:18-35.
- 517 12. Tonkin MA. Thumb duplication: Concepts and techniques. Clin Orthop Surg
518 2012;4:1-17.
- 519 13. Gummesson C, Ward MM and Atroshi I. The shortened disabilities of the arm,
520 shoulder and hand questionnaire (QuickDASH): Validity and reliability based
521 on responses within the full-length DASH. BMC Musculoskelet Disord
522 2006;7:44.
- 523 14. Eriksson M and Lindstrom B. Validity of Antonovsky's sense of coherence
524 scale: A systematic review. J Epidemiol Community Health 2005;59:460-6.
- 525 15. Sullivan M, Karlsson J and Ware JE, Jr. The Swedish SF-36 Health Survey-I.
526 Evaluation of data quality, scaling assumptions, reliability and construct validity
527 across general populations in Sweden. Soc Sci Med 1995;41:1349-58.
- 528 16. Carlsson I, Cederlund R, Hoglund P, Lundborg G and Rosen B. Hand injuries
529 and cold sensitivity: Reliability and validity of cold sensitivity questionnaires.
530 Disabil Rehabil 2008;30:1920-8.
- 531 17. Irwin MS, Gilbert SE, Terenghi G, Smith RW and Green CJ. Cold intolerance
532 following peripheral nerve injury. Natural history and factors predicting severity
533 of symptoms. J Hand Surg [Br] 1997;22:308-16.

18. Mathiowetz V, Kashman N, Volland G, Weber K, Dowe M and Rogers S. Grip and pinch strength: Normative data for adults. Arch Phys Med Rehabil 1985;66:69-74.
19. Mathiowetz V, Weber K, Volland G and Kashman N. Reliability and validity of grip and pinch strength evaluations. J Hand Surg [Am] 1984;9:222-6.
20. Graneheim UH and Lundman B. Qualitative content analysis in nursing research: Concepts, procedures and measures to achieve trustworthiness. Nurse Educ Today 2004;24:105-12.
21. Carlsson IK, Edberg AK, Wann-Hansson C. Hand-injured patients' experiences of cold sensitivity and the consequences and adaptation for daily life: a qualitative study. J Hand Ther. 2010;23:53-61.
22. Cederlund R, Thoren-Jonsson AL, Dahlin LB. Coping strategies in daily occupations 3 months after a severe or major hand injury. Occup Ther Int. 2010;17:1-9.
23. Chemnitz A, Dahlin LB, Carlsson IK. Consequences and adaptation in daily life - patients' experiences three decades after a nerve injury sustained in adolescence. BMC Musculoskelet Disord. 2013;14:252.
24. Wijk U, Carlsson I. Forearm amputees' views of prosthesis use and sensory feedback. J Hand Ther. 2015;28:269-77.
25. Dion K, Berscheid E and Walster E. What is beautiful is good. J Pers Soc Psychol 1972;24:285-90.
26. Blakeney P, Meyer W 3rd, Robert R, Desai M, Wolf S and Herndon D. Long-term psychosocial adaptation of children who survive burns involving 80% or greater total body surface area. J Trauma 1998;44:625-32.

- 558 27. Rumsey N and Harcourt D. Body image and disfigurement: Issues and
559 interventions. *Body image* 2004;1:83-97.
- 560 28. Andersson GB, Gillberg C, Fernell E, Johansson M and Nachemson A. Children
561 with surgically corrected hand deformities and upper limb deficiencies: Self-
562 concept and psychological well-being. *J Hand Surg Eur Vol* 2011;36:795-801.
- 563 29. Ardon MS, Janssen WG, Hovius SE, Stam HJ and Selles RW. Low impact of
564 congenital hand differences on health-related quality of life. *Arch Phys Med*
565 *Rehabil* 2012;93:351-7.
- 566 30. Lukash FN. Children's art as a helpful index of anxiety and self-esteem with
567 plastic surgery. *Plast Reconstr Surg* 2002;109:1777-86.
- 568 31. Bradbury E. The psychological and social impact of disfigurement to the hand in
569 children and adolescents. *Dev Neurorehabil* 2007;10:143-8.
- 570 32. Maddern LH, Cadogan JC and Emerson MP. 'Outlook': A psychological service
571 for children with a different appearance. *Clin Child Psychol Psychiatry*
572 2006;11:431-43.
- 573 33. Egan K, Harcourt D, Rumsey N and Appearance Research Collaboration. A
574 qualitative study of the experiences of people who identify themselves as having
575 adjusted positively to a visible difference. *J Health Psychol* 2011;16:739-49.
- 576 34. Joachim G and Acorn S. Stigma of visible and invisible chronic conditions. *J*
577 *Adv Nurs* 2000;32:243-8.
- 578 35. Griffiths C, Williamson H and Rumsey N. The romantic experiences of
579 adolescents with a visible difference: Exploring concerns, protective factors and
580 support needs. *J Health Psychol* 2012;17:1053-64.

- 581 36. Gustafsson M, Persson LO and Amilon A. A qualitative study of coping in the
582 early stage of acute traumatic hand injury. J Clin Nurs 2002;11:594-602.
- 583 37. Murray CE, Kelley-Soderholm EL and Murray TL. Strengths, challenges, and
584 relational processes in families of children with congenital upper limb
585 differences. Families, Systems & Health 2007;25:276-92.
- 586

587 Table 1. Characteristics of participants

Participant	Gender	Age group (years)	Type of congenital hand problem
1	M	21-25	Thumb hypoplasia
2	M	26-30	Thumb duplication
3	F	36-40	Thumb duplication
4	F	21-25	Thumb duplication
5	F	21-25	Thumb duplication
6	M	21-25	Thumb duplication
7	F	26-30	Thumb duplication
8	M	16-20	Thumb duplication
9	M	26-30	Thumb hypoplasia
10	F	36-40	Thumb hypoplasia
11	M	26-30	Thumb duplication
12	F	16-20	Thumb hypoplasia
13	M	16-20	Thumb hypoplasia
14	F	51-55	Thumb hypoplasia
15	F	16-20	Thumb hypoplasia

588

589

590 Table 2. Characteristics of participants with congenital hand problems
 591 (n=15).

Outcome measures	Median (range)
Quick DASH	4.5 (0-55)
Sense of Coherence (SOC)	74 (48-85)
Physical Functioning*	95 (75-100)
Role Physical*	100 (0-100)
Bodily Pain*	84 (10-100)
General Health*	82 (25-100)
Vitality*	75 (0-100)
Social Functioning*	100 (38-100)
Role Emotional*	100 (0-100)
Mental Health*	84 (44-100)
Cold Intolerance Symptom Severity	15 (4-58)
Pain at rest**	0 (0-100)
Pain on motion without load**	0 (0-100)
Pain on load**	20 (0-100)
Grip function**	30 (0-80)
Fine motor skill**	20 (0-90)
Weakness**	30 (0-100)
Appearance of the hand**	60 (10-100)

Grip strength (Jamar dynamometer)***	28 (10-54)
Key pinch strength*** (n=14)	6.2 (1-12)
Pinch strength*** (n=13)	6 (0-14)

592

Values are in median (range)

593

* Subscales in Short Form -36 questionnaire [15]

594

** Numeric rating scale range (0-100). 0 represents no problem, 100 represents worst possible problem

595

*** Average kg (range) [18,19]

596

597

598