



Research Article

ASSESSMENT OF QUALITY OF LIFE IN CHILDREN WHO HAVE UNDERGONE HYPOSPADIAS REPAIR

¹Parveen Kumar, ²Simmi K Ratan, ³Satish Kumar Aggarwal, ⁴Monica Juneja
⁵Jugal Kishore and ⁶Prashant Srivastava

¹Department of Pediatric Surgery, Chacha Nehru Bal Chikitsalya, New Delhi-110031, India

²Department of Pediatric Surgery, Maulana Azad Medical College, New Delhi-110002, India

³Department of Pediatric Surgery, Ganga Ram Hospital, New Delhi-110060, India

⁴Department of Pediatrics, Maulana Azad Medical College, New Delhi-110002, India

⁵Department of Community Medicine, Vardhman Mahavir Medical College, New Delhi-110029, India

⁶Department of Psychiatry, Kalpana Chawla Govt Medical College, Karnal, Haryana – 132001, India

ARTICLE INFO

Article History:

Received 10th March, 2020

Received in revised form 2nd

April, 2020

Accepted 26th May, 2020

Published online 28th June, 2020

Key words:

Quality of life; hypospadias; cosmetic assessment

ABSTRACT

Background/Introduction: The quality of life is a major concern in children who have undergone hypospadias repair; however there is paucity of data.

AIMS: To compare quality of life in children who have undergone hypospadias repair, as interpreted by child and parents. Also, to correlate the quality of life assessment with different types of hypospadias, surgical techniques and between staged and single repairs.

Material and Methods: An observational cross-sectional study was done at a single center from November 2014 to September 2016 in which 50 children, who had undergone hypospadias repair, were enrolled. Quality of life assessment was done with the help of direct questionnaire from patients and parents, using proforma of Paediatric Quality of Life Inventory™ (PedsQL)™ 4.0 Generic Core Scale- Standard that assesses 4 domains namely, physical (8 items), psychological (5 items), social (5 items) and school functioning (5 items). The data was analyzed using SAS 9.2 statistical software.

The age ranged from 2 years to 17 years at the time of enrollment. The follow up period post-surgery ranged from 6 months to 8.4 years. The study groups were divided based on location of meatus and as those undergone single stage repair, into distal penile hypospadias (DPH)[n=28], mid penile hypospadias (MPH)[n=11] and proximal penile hypospadias (PPH)[n=11]. 44/50 patients underwent single stage repair (88%), of which TIP was the most common approach (86.36%).

Results: Both parents and subjects enjoyed good quality of life (> 92% domain scores). DPH had better quality of life in comparison to MPH and PPH. Quality of life was better in single stage repairs irrespective of TIP and non-TIP repair.

Conclusions: The hypospadiacs and their parents were satisfied with cosmetic outcome with different types of repairs and enjoyed good quality of life.

Copyright©2020 Parveen Kumar et al. This is an open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

INTRODUCTION

The cosmetic and psychological aspects of hypospadias repair greatly affect the quality of life in children who undergoes hypospadias repair as well as that of family members in the long run and therefore, cannot be ignored.^{1,2,3} There is paucity of literature addressing this issue.⁴ The quality of life in family of hypospadias is not only affected by cosmetic aspects but also by the number of hospital admissions and follow up visits required. The occasional references to this aspect, in the existing English literature for hypospadias, relate to the adults.⁵⁻⁸

*Corresponding author: Parveen Kumar

Department of Pediatric Surgery, Chacha Nehru Bal Chikitsalya, New Delhi-110031, India

The quality of life accounts for adaptation of human beings to various conditions or circumstances in life. It measures people's individual perceptions about their position relative to other people and relative to their own expectations.⁹ The physical and mental health are a prerequisite to good QOL but do not account for it entirely.

The WHOQOL-BREF instrument for assessment of quality of life was developed for cross-cultural comparisons of QOL and is validated for people, aged 18 years and older. It comprises 24 items, resulting in four domains (physical health, psychological, social relationships, and environment) with three to eight items per domain.¹⁰ The health-related quality of life (HRQoL), a generic measure is determined as a functional status weighted by the emotional reaction. Functional status

items tell about the incidence of physical, psychological and social problems on different subscales. For all scales, higher scores indicate a better HRQOL.

METHODOLOGY

This was an observational cross-sectional study done at pediatric surgery department of a tertiary care hospital, from November 2014 to September 2016. A total of 139 subjects (age range 2 to 17 years) who had undergone repair for varying severity of isolated hypospadias between February 2006 to March 2016 were contacted telephonically or by postal mode for this study. An inclusion criterion of completion of at least 6 months after last repair was set. Subjects with disorders of sexual differentiation or associated syndromes or other urological association were excluded from the study. Out of 139, 89 patients either did not fulfill recruitment criteria or did not respond to communication. Hence, 50 subjects were included in the study.

Quality of life assessment was done by child psychologist using proforma of Paediatric Quality of Life Inventory™ (PedsQL)™ 4.0 Generic Core Scale- Standard.^{11,12,13} The permission was sought from the MAPI Institute, Lyon, France prior to use of the questionnaire. This scale assessed the QOL of children in 4 domains with total 23 items, namely, physical functioning (8 items), psychological/emotional functioning (5 items), social functioning (5 items), school functioning (5 items). PedsQL items assess how much of a problem a particular variable among all above stated has been for patients during a certain period (the standard recall period is the past one month). Suitable module (Hindi or English version) were given to the child depending on his age and comprehension and parent's proxy report were used wherever child was not able to answer the questionnaire. [Samples attached] For computing PedsQL score, the guidelines provided by PedsQL were used. Items were reversed scored and linearly transformed to a 0-100 scale as follows: 0=100, 1=75, 2=50, 3=25, 4=0. If more than 50 % of the items in the scales of a domain were missing, then scale scores were not computed. Mean scores were computed by summing of the items over the number of items answered. The scores were compared in relation to severity of hypospadias, stages of repair and results of assessment by subject and their parents.

The data was fed in computer on MS excel and subjected to descriptive statistics to estimate means and standard deviation. The comparison between levels in each variable was done using t-test in One-way ANOVA. The relationship among the variables was determined using Pearson's correlation. All the data analyses were performed using SAS 9.2 statistical software. Ethical clearance for the study was obtained from the Institutional Ethical Committee.

RESULTS

50 patients qualified for the study. The distribution of our study subjects was as (age was calculated depending on the completed number of years): Age groups 2-4 years (n=9), 5-7 years (n=13), 8-12 years (n=22) and 13-18 years (n=6).

Follow up period ranged from 6 months to 8.4 years (mean follow up of 37.48 months and median 28 months). Out of 50 patients, 44 had undergone single stage repair and 6 had staged

repairs. Among the former, 86.36 % (n=38) had TIP repair and rest 13.64 % (n=6) had non-TIP operations (1 Thierschduplayurethroplasty, 2 Duckett tubed prepuccial flap urethroplasty, 3 had Duckett prepuccial island onlay flap urethroplasty).

The study group had subjects with distal penile hypospadias (DPH)[n=28], mid penile hypospadias (MPH)[n=11] and proximal penile hypospadias (PPH)[n=11].

PedsQL assesses the QOL of children in 4 domains as mentioned in methodology. Mean scores for PQL (Pediatric Quality of Life) given by parent were as follows: overall score 96.50 ±4.74, physical domain 97.31±4.28, emotional domain 96.8±6.53, social domain 98±5.62 and school domain 92.86±13.23. Similarly mean scores for child PQL overall score was 95.64±5.12, physical domain 96.79±5.01, emotional domain 93.65±8.52, social domain 96.95±7.89 and school domain 94.5±11.00. Parent and child overall PQL scores were equally correlated well with physical, emotional, social and school health.

PQL scores given by parent and child did not differ among DPH, MPH and PPH, except for PQL social domain score by parent which were significantly higher for DPH and MPH in comparison to PPH patients.

Few other observations emerged when the data was compared for single/staged repairs. The PQL score given by parent was statistically higher for single stage repair (P value 0.04) in comparison to staged repair. This did include difference of emotional domain of score. There was no difference for PedsQOL score given by parent or patient for either TIP or non-TIP type of repair.

DISCUSSION

With increasing success rate of hypospadias surgery, psychosexual, psychosocial issues and quality of life are the major concerns for treating surgeons, parents and patients. In past, assessment of cosmetic appearance was generally domain of surgeon; whereas parents and patients were generally asked about the functional outcome. However, of late, the views of parents and patients are also been given due importance.

We chose Ped QOL generic core scale for its wider coverage of age range of our study subjects and its simplicity. It has high reliability and validity, with high internal consistency, taking less than 5 minutes to complete. It also has advantage of patient- report age range 5 to 18 years and proxy-report age range of 2 to 18 years and high parallelity in patient and proxy reports. Our assessment was limited to psychosocial aspects and physical quality of life only as our subjects were younger. They were not enquired into psychosexual function.

We analysed the quality of life in these children. Bracka 1999 stated that hypospadias patients grew up with fewer psychological problems than their predecessors.¹⁴ Mureau *et al* (1997) did not report any differences in self-reported psychosocial adjustment between 116 boys with operated hypospadias (9-18 years) and 88 age-matched healthy control subjects on several standardized measures (DPQ-J: Junior Dutch Personality Questionnaire, SAS-C: Social Anxiety Scale for Children, YSR: Youth Self-Report).¹⁵ Conversely, the

findings of Purschke and Standke (1993) suggest that the 47 children with hypospadias (5–13 years) had more behavior problems than the 23 children of the same age, who had been subjected to a variety of mild operations for other medical problems.¹⁶ In 1989, an American research group (Sandberg *et al.*) found more behavior problems and lower social competence in 69 children with hypospadias (6–10 years) compared to the corresponding norms of 300 healthy children between the ages of 4 and 16 years.¹⁷ Twelve years later, the same researchers (Sandberg *et al.*, 2001) published a study with a slightly better methodological standard that involved a larger sample of hypospadias patients (N=175) as well as a control group of healthy school boys (N=333).¹⁸ They showed that the parents of hypospadias patients (6–10 years) considered their children to be less socially competent than the parents of the comparison subjects (6–10 years). They reported that the parent's satisfaction with the appearance of the patients' genitals positively correlated with their academic achievement though not so with their behavior problems. With regard to sexual inhibitions, both Mondaini *et al.* (2002)¹⁹ and Mureau *et al.* (1995a)²⁰ found that children and adolescents with hypospadias reported more sexual inhibitions than the healthy age-matched controls. They were not only more afraid of being rejected by a potential partner due to genital appearance but also prone to hide genitals in public lavatories.^{19,20} Schonbucher *et al.* also found in their study that boys with hypospadias suffered from negative genital appraisal and sexual inhibitions.²¹

It is very interesting to note that the period of follow up post hypospadias surgery has been always a matter of debate. It has been criticized that the follow-up periods are too short to draw any affirmative conclusions on outcome and long term issues and complications.²² On the other hand, Snodgrass *et al.* believe that mostly complications occur shortly after operation, sufficing a follow up period of 6 months.²³ However, Spinoit *et al.* in their study reinforced the need of mandatory long term follow up to extract the true complication rates.²⁴ They showed that about 75 % of patients would have good long-term outcome without further complications but only about half of remaining 25 % presented in first year of follow up for re-operation. In our series, the patients had a follow up period ranging from 6 months to 8.4 years and we are of the opinion that long term follow up of these patients is a must.

Schönbucher *et al.* in their study on health related quality of life and psychological adjustment of children and adolescents with hypospadias concluded that in comparison with control subjects, self-reported HRQOL scores were lower among hypospadiacs.²¹ A China based case control study showed that the adults after hypospadias repair had higher incidence of anxiety and depression. There was also a correlation between the severity of symptoms and age at operation and penile size.⁵ Vandendriessche *et al.* in their small study on adolescents following hypospadias repair showed that the overall social, psychosocial, and sexual development is normal although body image and genital perception is impaired.⁶ Our study showed ≥ 92 % satisfaction with various domains of QOL in both parents and child. Our study groups had more younger age, so comparison to adult data is not possible. Also, parents rated most domains better as compared to children except school

domain. This domain included question about school attendance and performance. Emotional domain rating of study subjects did not differ with types of hypospadias but surprisingly mean values were higher for PPH though not statistically different from those of DPH and MPH. One reason could be that most of our study subjects were younger and must not have been subjected to peer ridicule before repair. This might be because as per general norm in our study population, most parents would not send children to school before completion of all stages of hypospadias repair. Also, parental norms allow lesser effecting on children. Hence there is agreement in early age for hypospadias surgery. The true comparison to our study may not be possible as our patients hail from different cultures and socioeconomic strata and it may cause skewed results.

In our study, minor complications like meatal stenosis were managed well on OPD or home basis. Also, there were no strictures, so quality of life may be compromised. From available data, it was inferred that the main concern for parent while assessing PedsQOL was school performance (lowest mean school score 92.86 in comparison to means of other 3 domains) whereas for patient themselves, it was emotional parameters (lowest mean emotional score 93.65 in comparison to means of other 3 domains).

In a multicenter cross-sectional clinical evaluation study about quality of life in adults with disorders/differences of sex development (DSD) compared to healthy European populations, QOL was similar in psychological, slightly worse in physical health, and slightly better in environment. In social relationships, QOL was significantly poorer compared to healthy and non-healthy reference populations.²⁵ An Indonesian population study on DSD patient's quality of life found out that children with DSD reported more problems in social functioning and had less positive moods.²⁶ In our study, only children were included and that too with isolated hypospadias. The syndromic and DSD conditions were excluded criteria as they have chronic physical and mental diseases both related and unrelated to the specific DSD diagnosis and is likely to impact on quality of life.

In our study, the QOL of parents and child inter-correlated. It may be stated that the emotional and psychological outlook of parents has great bearing on similar aspects of child. These may have subsequent effect over psycho-sexual aspects and needs longer follow up. However, parents rated school performance domain with poor scores and children rated emotional domain poorer.

Limitations: The children should be followed up in adolescence to assess psychosexual aspects resultant to hypospadias repair. For better comparisons, the inclusion criteria should include equal numbers of DPH and PPH and also control population. A large number of patients did not respond to the questionnaire. This may have led to a selection bias, as it is possible that the ones with a poorer QOL chose not to respond.

CONCLUSION

Our study concludes that both hypospadiacs and their parents were satisfied with cosmetic outcome with different types of repairs employed and they also enjoyed good quality of life especially in subjects having isolated hypospadias. The awareness for public must be done for getting early surgery before sending to school.

References

1. Springer A. Assessment of outcome in hypospadias surgery—a review. *Frontiers in pediatrics*. 2014;2.
2. Hadidi AT. Assessment of outcome. In: Hadidi AT, Azmy AF, editors. *Hypospadias Surgery*. Heidelberg: Springer. 2004; 90–2.
3. Mureau MA, Slijper FM, Slob AK, Verhulst FC, Nijman RJ. Satisfaction with penile appearance after hypospadias surgery: the patient and surgeon view. *The Journal of urology*. 1996;155(2):703-6.
4. Sinko K, Jagsch R, Precht V, Watzinger F, Hollmann K, Baumann A. Evaluation of esthetic, functional, and quality-of-life outcome in adult cleft lip and palate patients. *The Cleft Palate-Craniofacial Journal*. 2005;42(4):355-61.
5. Wang WW, Deng CH, Chen LW, Zhao LY, Mo JC, Tu XA. Psychosexual adjustment and age factors in 130 men undergone hypospadias surgery in a Chinese hospital. *Andrologia*. 2010;42(6):384-8.
6. Vandendriessche S, Baeyens D, Van Hoecke E, Indekeu A, Hoebeke P. Body image and sexuality in adolescents after hypospadias surgery. *Journal of pediatric urology*. 2010;6(1):54-9.
7. Schönbacher VB, Weber DM, Landolt MA. Psychosocial adjustment, health-related quality of life, and psychosexual development of boys with hypospadias: a systematic review. *Journal of pediatric psychology*. 2008;33(5):520-35.
8. Rynja SP, de Jong TP, Bosch JL, de Kort LM. Functional, cosmetic and psychosexual results in adult men who underwent hypospadias correction in childhood. *Journal of pediatric urology*. 2011;7(5):504-15.
9. WHOQOL Group. The World Health Organization quality of life assessment (WHOQOL): position paper from the World Health Organization. *SocSciMed*. 1995;41:1403–9.
10. WHOQOL Group. Development of the World Health Organization WHOQOL-BREF quality of life assessment. *Psychol Med*. 1998;28:551–8.
11. Merriman LS, Arlen AM, Broecker BH, Smith EA, Kirsch AJ, Elmore JM. The GMS hypospadias score: assessment of inter-observer reliability and correlation with post-operative complications. *Journal of pediatric urology*. 2013;9(6):707-12.
12. Wouters G, Rynja S, Van Schaijk M, Kok ET, de Jong TP, de Kort LM. Long Term Follow Up Into Adulthood of Boys Operated for Hypospadias: A Study Into Functional And Cosmetic Results. *Journal of Pediatric Urology*. 2009;5:S56.
13. Rynja SP, de Kort LM, de Jong TP. Urinary, sexual, and cosmetic results after puberty in hypospadias repair: current results and trends. *Current opinion in urology*. 2012;22(6):453-6.
14. Bracka A. Sexuality after hypospadias repair. *BJU international*. 1999;83(S3):29-33.
15. Mureau MA, Slijper FM, Slob AK, Verhulst FC. Psychosocial functioning of children, adolescents, and adults following hypospadias surgery: a comparative study. *Journal of Pediatric Psychology*. 1997;22(3):371-87.
16. Purschke C, Standke M. Psychische Besonderheiten bei Knaben mit Hypospadien. *Pädiatr Grenzgeb*. 1993;31:175-85.
17. Sandberg DE, Meyer-Bahlburg HF, Aranoff GS, Sconzo JM, Hensle TW. Boys with hypospadias: a survey of behavioral difficulties. *Journal of Pediatric Psychology*. 1989;14(4):491-514.
18. Sandberg DE, Meyer-Bahlburg HF, Hensle TW, Levitt SB, Kogan SJ, Reda EF. Psychosocial adaptation of middle childhood boys with hypospadias after genital surgery. *Journal of pediatric psychology*. 2001;26(8):465-75.
19. Mondaini N, Ponchiotti R, Bonafè M, Biscioni S, Di Loro F, Agostini P, Salvestrini F, Rizzo M. Hypospadias: incidence and effects on psychosexual development as evaluated with the Minnesota Multiphasic Personality Inventory test in a sample of 11,649 young Italian men. *Urologia internationalis*. 2002;68 (2):81-5.
20. Mureau MA, Slijper FM, Nijman RJ, van der Meulen JC, Verhulst FC, Slob AK. Psychosexual adjustment of children and adolescents after different types of hypospadias surgery: a norm-related study. *The Journal of urology*. 1995;154(5):1902-7.
21. Schönbacher VB, Landolt MA, Gobet R, Weber DM. "Health-related quality of life and psychological adjustment of children and adolescents with hypospadias." *The Journal of paediatrics*. 2008; 152.6:865-872.
22. González R, Ludwikowski B, Silay MS, Sirin H, Tepeler A, Karatag T, et al. "Snodgraft" technique for the treatment of primary distal hypospadias: pushing the envelope. *J Urol*. 2012; 188:938–42.
23. Snodgrass W, Macedo A, Hoebeke P, Mouriquand PDE, et al. Hypospadias dilemmas: a round table. *J Pediatr Urol*. 2011;7:145–57.
24. Spinoit AF, Poelaert F, Groen LA, Van Laecke E, Hoebeke P. Hypospadias repair at a tertiary care center: long-term followup is mandatory to determine the real complication rate. *J Urol*. 2013;189:2276–81.
25. Rapp M, Mueller-Godeffroy E, Lee P, Roehle R, Kreukels BP, Köhler B, et al. Multicentre cross-sectional clinical evaluation study about quality of life in adults with disorders/differences of sex development (DSD) compared to country specific reference populations (dsd-LIFE). *Health and quality of life outcomes*. 2018;16(1):54.
26. Ediati A, Verrips GH, Juniarto AZ, Faradz SM, Drop SL, Dessens AB. Quality of life in late-treated patients with disorders of sex development: Insights for patient-centered care. *Frontiers in pediatrics*. 2019;6:434.