

Letter to the Editor: Sexual Function and Quality of Life relates to functional outcome in adult male patients with Hirschsprung's

Dear Editor,

We have read with great interest the recent publication by Dr Trinidad and the group of Dr Frischer at Cincinnati Children's describing Sexual Function and its relationship to Bowel Dysfunction in patients with Anorectal Malformation (ARM) and Hirschsprung's Disease (HSCR)¹. The authors should be congratulated for completing this study with the use of objective instruments which allows for comparison of results to other series and other conditions.

We published in this journal our own institutional series of long-term function and quality of life outcomes in HSCR specifically²; we were able to capture information regarding sexual function and fertility on both male and female adult patients albeit in a less detailed manner than the present paper – we utilised the simplified Erectile Hardness Scale (EHS; score 0-4³) and Sexual Quality of Life – Male (SQOL-M; score out of 66⁴) questionnaire in addition to simple questions relating to orgasm and ejaculation. Bowel function was assessed with the Bowel Function Score (BFS; $\leq 12/20$ considered poor function⁵). Our results were initially published with a view to demonstrate apparent issues in female HSCR patients and therefore our analysis of factors in male patients was somewhat lacking⁶. We have taken this opportunity and recent publication to re-analyse our data further and produce similar results to those seen by the team in Cincinnati.

The responding adult male population of 98 patients had a median age of 30 years (range 18-40), 80% had short segment disease and 79% had undergone an open Duhamel pull-through. Sexual dysfunction among the 94 patients with complete data was infrequent, however "any ED" (taken as EHS < 4) was reported at a rate not statistically different to the current series (17/98 vs 2/12; $p=1.0$). There was infrequent reporting of "any Issues" surrounding sexual climax (9/94; 9.6%) and ejaculation (4/94; 4.3%). When analysing correlations between sexual quality of life, erectile dysfunction and bowel function we saw a similar relationship to that observed by the present series, with an apparent link between bowel function and sexual quality of life as well as observing a poorer BFS in those patients with ED (see Figure).

It may be of additional interest that the fertility outcomes in this patient cohort appear to be relatively normal. Thirty-six patients had attempted to father children, 30 (83%) doing so successfully and an additional 3 with assisted-reproduction services (IVF success rate 3/6; 50%). Information regarding affected offspring was not sought.

Clearly both of these published datasets may present some concerns regarding reporting bias within the dataset (our own data for this demographic 130/164 contacted (79%) – reflecting differences in population distribution and central record keeping; 98/130 completed (75%)) and in reality the direction in which this might bias the results is quite unknown. Nevertheless, we are pleased to be able to provide reinforcing data to underline the message presented by our colleagues, we hope that these data prove useful for counselling and long-term assessment of this patient group and we congratulate our colleagues once more for their work.

Joe Davidson (University College London and Great Ormond Street Hospital)

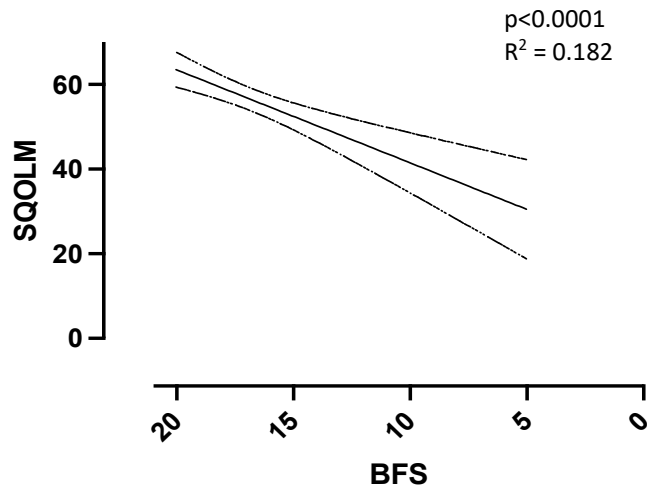
Simon Eaton (University College London)

Mikko Pakarinen (Helsinki University Children's Hospital)

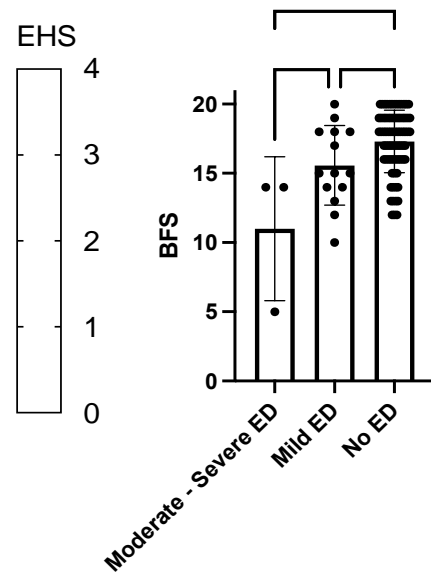
Joe Curry (Great Ormond Street Hospital)

Figure – Sexual Quality of Life (SQOLM) vs. Bowel Function Score (BFS) with linear regression analysis; symbols represent individual responses with colour reflecting corresponding EHS. BFS in patients with mild and moderate-severe Erectile Dysfunction; comparison with 1-way ANOVA and multiple comparisons; * $p < 0.05$, ** $p < 0.001$, * $p < 0.0001$.**

Sexual Quality of Life vs Bowel Function Score



ED vs BFS



References

1. Trinidad, S. *et al.* Long-Term Male Sexual Function and Fecal Incontinence Outcomes for Adult Patients With Hirschsprung Disease or Anorectal Malformation. *J Pediatr Surg* (2023) doi:10.1016/j.jpedsurg.2023.04.006.
2. Davidson, J. R. *et al.* Long-term surgical and patient-reported outcomes of Hirschsprung's Disease. *J Pediatr Surg* (2021) doi:10.1016/j.jpedsurg.2021.01.043.
3. Mulhall, J. P., Goldstein, I., Bushmakina, A. G., Cappelleri, J. C. & Hvidsten, K. Validation of the Erection Hardness Score. *J Sex Med* **4**, 1626–1634 (2007).
4. Abraham, L., Symonds, T. & Morris, M. F. Psychometric Validation of a Sexual Quality of Life Questionnaire for Use in Men with Premature Ejaculation or Erectile Dysfunction. *J Sex Med* **5**, 595–601 (2008).
5. Rintala, R. J. & Lindahl, H. Is normal bowel function possible after repair of intermediate and high anorectal malformations? *J Pediatr Surg* **30**, 491–4 (1995).
6. Davidson, J. R. *et al.* Sexual function, quality of life, and fertility in women who had surgery for neonatal Hirschsprung's disease. *British Journal of Surgery* **108**, e79–e80 (2021).