Comprehensive review of procedures for total colonic aganglionosis

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CRD summary
The conclusion that operative techniques for treating total colonic aganglionosis were comparable in terms of mortality, morbidity, enterocolitis and functional outcome should be regarded with some caution, due to a lack of good-quality evidence and presence of heterogeneity in the study findings and methodological weaknesses in the review.

Authors' objectives
To compare the effectiveness of different operative treatments for total colonic aganglionosis.

Searching
MEDLINE (1950 to 2007) and The Cochrane Library were searched for studies in English. Search terms were reported.

Study selection
Studies of single operative interventions for total colonic aganglionosis were eligible for inclusion if they reported operative outcomes (mortality, morbidity, enterocolitis or functional outcome) after definitive treatment. Studies of total colonic aganglionosis with extensive small bowel involvement and studies of sub-total colonic aganglionosis were excluded.

The following interventions were included in the review: total colectomy with endorectal pull-through with ileorectal or ileoanal anastomosis (endorectal pull-through only); endorectal pull-through with ileorectal anastomosis and ileocolostomy using the entire aganglionic colon (ileocolostomy entire colon) and the left descending colon (ileocolostomy left colon); endorectal pull-through with a right ascending colon on-lay patch (right colon patch); and total proctocolectomy with ileoanal pouch anastomosis.

The mean age of participants having pull-through procedures was 15.3 months (+/- 8.4 months). Measures of functional outcomes varied across studies and included continence, number of stools daily and stool consistency. Outcomes in the review included operative outcomes and surgical complications. Duration of follow up ranged from one month to 11 years; mean follow up was 3.9 years (where reported).

A single reviewer evaluated abstracts for inclusion.

Assessment of study quality
The authors did not state that they assessed validity.

Data extraction
Cumulative event rates for each outcome were extracted for each study. The authors did not state how many reviewers performed the data extraction.

Methods of synthesis
Studies were combined after logit transformation using fixed-effect models to calculate pooled event rates for each type of intervention, with 95% confidence intervals (CIs). Where the event rate was nil or one, statistical adjustment was made to permit calculation of the standard error of the logit. Heterogeneity was assessed using the $I^2$ statistic. Overall event rates across studies were also calculated.

Results of the review
Nineteen retrospective studies were included (n=161).

Morbidity
The mean overall morbidity rate was 44 per cent +/- 32.9 per cent (18 studies). Event rates and heterogeneity ($I^2$) for each intervention were: for endorectal pull-through only 0.255 (95% CI: 0.142, 0.414; four studies, n=63, $I^2$=73.9%);
for ileocolostomy of the entire colon 0.327 (95% CI: 0.162, 0.549, two studies, n=24, $I^2=8.2\%$); for ileocolostomy of the left colon 0.637 (95% CI: 0.416, 0.812, five studies, n=41, $I^2=56.6\%$); for right colon patch 0.302 (95% CI: 0.143, 0.528, five studies, n=23, $I^2=0\%$); and for ileal pouch anastomosis 0.100 (95% CI: 0.014, 0.467, one study, n=10).

Other outcomes
Data for other outcomes were unsuitable for meta-analysis. Across all studies the mean mortality rate was 1.9 per cent (19 studies) and mean enterocolitis rate was 22.6 +/- 13.5 per cent (seven studies). Most children were continent with an average of three to five stools daily (19 studies). Complications of comparable severity were noted for all types of intervention, the most frequent complication being diarrhoea with or without perineal excoriation.

Authors’ conclusions
Operative techniques for treating total colonic aganglionosis were comparable with respect to mortality, morbidity, enterocolitis and functional outcomes.

CRD commentary
The objectives and inclusion criteria of the review were clear. Some relevant sources were searched for studies, but few databases were searched, the search was limited to studies in English and there was apparently no specific attempt to retrieve unpublished studies. Thus, it is possible that studies were missed and that the review was subject to language and publication biases. Studies were selected by a single reviewer, which increased the risk of bias and error. The process of data extraction was not reported. It did not appear that study validity was assessed. These factors make it difficult to assess the reliability of the findings presented.

The statistical techniques used to combine data and assess for heterogeneity appeared suitable; heterogeneity and other potential biases (such as small sample sizes) were acknowledged in the text. In view of the lack of good-quality evidence, the heterogeneity in study findings and the methodological weaknesses in the review, the authors’ conclusions should be regarded with some caution.

Implications of the review for practice and research
Practice: The authors stated that choice of operative technique for total colonic aganglionosis should be made according to the familiarity and expertise of the surgeon and the overall experience of the surgical centre.

Research: The authors stated that long-term studies of functional outcomes after definitive treatment of total colonic aganglionosis were required.

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Record Status
This is a critical abstract of a systematic review that meets the criteria for inclusion on DARE. Each critical abstract contains a brief summary of the review methods, results and conclusions followed by a detailed critical assessment on the reliability of the review and the conclusions drawn.