
Surgical management of aortic root disease in Marfan syndrome: a systematic review and meta-analysis

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CRD summary

The authors concluded that valve-sparing root replacement may represent a valuable option for patients with Marfan syndrome with aortic aneurysm. The reliability of the authors' conclusions is uncertain due to review weaknesses that included potential publication bias, reviewer error and bias in data extraction, unclear quality of included studies and small study sample sizes.

Authors' objectives

To compare results of total root replacement versus valve-sparing aortic root replacement in Marfan syndrome patients.

Searching

PubMed, EMBASE and The Cochrane Library were searched for papers published between January 1966 and February 2010. Search terms were reported. It appeared that only studies published in English were eligible for inclusion. Reference lists of relevant articles were handsearched.

Study selection

Observational studies (sample size of 30 or more) that reported on valve-related morbidity and mortality after total root replacement and/or valve-sparing root replacement in patients with Marfan syndrome were eligible for inclusion. Outcome measures were re-intervention on the aortic valve, thromboembolic event and endocarditis.

Data on study settings was not reported. Surgical techniques evaluated included composite valve graft, composite valve graft or homograft, remodelling and reimplantation or remodelling. Mean age of patients ranged between 29.2 and 41.7 years.

Two reviewers independently assessed studies for inclusion; disagreements were resolved by negotiation.

Assessment of study quality

The authors did not state that they assessed study quality.

Data extraction

Data were extracted on total root replacement and valve-sparing root replacement. Where the total number of patient years was unreported it was computed by multiplying the number of hospital survivors with the mean follow-up duration.

The authors did not state how many reviewers extracted data.

Methods of synthesis

Pooled occurrence rates of valve-related complications were calculated using fixed-effect meta-analysis (where there was no heterogeneity) or random-effects meta-analysis (where there was heterogeneity). Heterogeneity was assessed using Cochran's Q and I². Publication bias was assessed using a funnel plot and Egger's test. Subgroup analysis explored the effects of patient age, urgent indication and valve sparing technique (reimplantation versus remodelling) on pooled results. The potential influence of study follow-up on results was explored using meta-regression analysis.

Results of the review

Eleven studies (1,385 participants) were included. Sample sizes ranged between 43 and 625. Mean follow-up time ranged between 1.58 and 9.5 years. Seven studies assessed total root replacement and four assessed valve-sparing root replacement.

The re-intervention rate was lower in total root replacement compared to valve-sparing root replacement (0.3% per

year, 95% CI 0.1 to 0.5 versus 1.3% per year, 95% CI 0.3 to 2.2; $p=0.02$). There was evidence of statistical heterogeneity for this analysis (total root replacement $I^2=24\%$, valve-sparing root replacement $I^2=71\%$).

Thromboembolic events rate was higher in total root replacement compared to valve-sparing root replacement (0.7% per year, 95% CI 0.5 to 0.9 versus 0.3% per year, 95% CI 0.1 to 0.6; $p=0.01$). There was no evidence of statistical heterogeneity ($I^2=0\%$ for both groups).

Compared to remodelling, reimplantation was associated with reduced rates of reimplantation among patients who underwent valve-sparing root replacement (0.7% per year versus 2.4% per year; $p=0.02$).

There were no significant differences between total root replacement and valve-sparing root replacement for endocarditis rate or composite valve related events. There was some evidence of statistical heterogeneity for these comparisons except for the analysis of valve-sparing root replacement for endocarditis rate.

Meta-regression analysis of differences in follow-up durations among studies for re-intervention rates after valve-sparing root replacement found that the highest rates were recorded in the studies with small sample size and short-follow-up.

There was no association between mean age and urgent indication on valve-related complication rates.

There was evidence for publication bias for re-intervention and endocarditis outcomes.

Authors' conclusions

Valve-sparing root replacement may represent a valuable option for patients with Marfan syndrome with aortic aneurysm. The technique should be used with caution in patients with valve characteristics at risk for decreased durability.

CRD commentary

The review question was broadly stated. It appeared that only studies published in English were included and this may mean that some relevant studies were missed. Study selection was conducted in duplicate, which minimised potential for reviewer error and bias; it was unclear whether similar processes were used in data extraction so the possibility of error and bias could not be excluded. Quality of the included studies was unclear as there was no validity assessment. Appropriate methods were used to explore heterogeneity and combine study results. Most studies enrolled few patients.

The reliability of the authors' conclusions is uncertain due to review weaknesses that included potential publication bias, reviewer error and bias in data extraction, unclear quality of included studies and small study sample sizes.

Implications of the review for practice and research

Practice: The authors stated that reimplantation should be considered for patients with Marfan syndrome as it was associated with an acceptable re-intervention rate. This technique needed to be used cautiously in patients with valve characteristics at risk for decreased durability. This surgery should be performed in centres with considerable experience in valve-sparing techniques.

Research: The authors stated that there was on-going non-randomised prospective international registry study comparing valve-sparing with total root replacement in patients with Marfan syndrome.

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