Local control of extra-abdominal desmoid tumors: systematic review and meta-analysis

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CRD summary
This review concluded that surgery alone for extra-abdominal desmoid tumours had a consistently high rate of local recurrence, addition of radiation therapy improved local control and limited data suggest benefits of both cytotoxic and non-cytotoxic chemotherapy in achieving stable disease. These conclusions were based on non-randomised studies that were mostly uncontrolled and retrospective. Their reliability is uncertain.

Authors' objectives
To assess the local failure and response rates of surgery, radiation therapy and chemotherapy in the treatment of extra-abdominal desmoid tumours.

Searching
MEDLINE, EMBASE, CINAHL and Cochrane databases were searched up to January 2011. Search terms were reported. Only studies reported in English were eligible for inclusion.

Study selection
Studies that assessed a method for treatment of patients with extra-abdominal desmoid tumours were eligible for inclusion. Studies could be randomised controlled trials (RCTs), other controlled designs or observational studies. Studies needed to report a clinical outcome such as recurrence rate for each treatment group.

Included studies mostly included a mixture of patients with primary and recurrent disease. Radiation doses varied from 22 to 75Gy. Surgery was reported as being primary excision, wide excision, complete excision, marginal or intrallesional. Systemic therapies included a range of cytotoxic and non-cytotoxic treatments. There was no information on the characteristics of participants in the included studies.

Two reviewers independently assessed studies for inclusion and disagreements were resolved by a third reviewer.

Assessment of study quality
Study quality was assessed independently by two reviewers using the nine-point Newcastle-Ottawa scale for cohort studies. Studies that scored 7 points or higher were considered to be of high quality, those that scored 5 or 6 to be moderate quality and those that scored 4 or less to be low quality. Disagreements were resolved through discussion.

Data extraction
Data were extracted to enable calculation of odds ratios with 95% confidence intervals. The authors did not state how many reviewers performed the data extraction.

Methods of synthesis
Pooled odds ratios with 95% confidence intervals were calculated for comparative studies using a fixed-effect model. A weighted pooled proportion was calculated for single group studies for either response rate or failure rate. Heterogeneity was assessed using the Cochran Q and quantified using the I² statistic. Publication bias was assessed using a funnel plot.

Results of the review
Thirty-five studies were included in the review. No RCTs were identified. Seven retrospective studies assessed radiation therapy. Eighteen studies (two had a prospective design) assessed surgical therapy. Fourteen studies (three prospective) assessed a combination of methods. Nine studies (six prospective) assessed systemic therapy; five of these assessed cytotoxic agents. Follow-up ranged from nine months up to 28 years. The mean quality assessment score was 7.23, which indicated high quality of evidence.

Radiation therapy alone: The weighted pooled proportion for local failure rate was 0.22 (95% CI 0.16 to 0.28; I²=15%).
Surgery alone: The weighted pooled proportion for local failure rate was 0.35 (95% CI 0.26 to 0.44; I² = 90%).

Combined surgery and radiation therapy: The weighted pooled proportion for local failure rate was 0.28 (95% CI 0.18 to 0.39; I² = 84%). Comparative studies showed lower failure rates for combination therapy compared to both radiation alone (OR 0.69, 95% CI 0.38 to 1.23; four studies; I² = 20%) and to surgery alone (OR 0.69, 95% CI 0.47 to 1.00; eight studies; I² = 0%).

Systemic therapy: The weighted pooled proportion for response rate was 0.91 (95% CI 0.86 to 0.96; I² = 0%) for non-cytotoxic therapies and 0.52 (95% CI 0.39 to 0.65; I² = 29%) for cytotoxic therapies.

There was no evidence of publication bias.

Authors' conclusions
Surgery alone for extra-abdominal desmoid tumours had a consistently high rate of local recurrence; addition of radiation therapy improved local control. However, limited data suggest the benefits of both cytotoxic and non-cytotoxic chemotherapy in achieving stable disease.

CRD commentary
The review had a clear question supported by inclusive criteria for selecting studies. The search was reasonably extensive. The authors reported using an appropriate means of assessing the non-randomised and often uncontrolled studies that were located. They also reported using review methods that are designed to reduce the possibility of reviewer bias and error during study selection and the assessment of study quality; it was unclear whether this was also the case for data extraction.

The synthesis involved pooling studies that appeared to differ in treatment techniques and follow-up duration; statistical heterogeneity appeared to be low in most analyses.

The authors’ conclusions reflect the evidence of the included studies. However, the evidence was based on non-randomised and largely uncontrolled and retrospective studies. The authors acknowledged this limitation and it should be regarded as a source of uncertainty about the reliability of the conclusions.

Implications of the review for practice and research
Practice: The authors stated that overall there appeared to be a role for systemic therapy (and specifically non-cytotoxic chemotherapy) in the treatment of extra-abdominal desmoid tumours.

Research: The authors stated a need for high quality randomised controlled trials of radiation therapy and chemotherapy in the treatment of desmoid tumours.

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