Surgical management of craniopharyngiomas in children: meta-analysis and comparison of transcranial and transsphenoidal approaches

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CRD summary
This review found that directly comparing outcomes after transcranial and transsphenoidal surgery in children with brain tumours may not be valid because of baseline differences in the groups undergoing these surgical procedures. Methodological flaws in the review and the unknown quality of the included studies means the reliability of authors' cautious conclusions is unclear.

Authors' objectives
To determine whether comparisons between transcranial and transsphenoidal surgery in paediatric patients with craniopharyngiomas (brain tumours that develop in the pituitary gland) are valid.

Searching
PubMed and EMBASE were searched from January 1990 to January 2010 for relevant studies in English; search terms were reported. Reference list selected articles of interest were checked to identify additional studies.

Study selection
Studies that examined transcranial surgery (in more than 20 patients) and transsphenoidal surgery (in more than 10 patients) in patients younger than 21 years old with craniopharyngioma were eligible for inclusion. Studies that reported on planned limited resection and radiation therapy, cyst aspiration, intracavity treatments, biopsies or cerebrospinal fluid diversion procedures, single functional domains were excluded; as were those that reported combined results of craniopharyngioma resection with other pathologies.

For the patients who received transcranial surgery, visual status deficiencies were recorded for 53.5% of the patients (where reported); the presence or absence of hydrocephalus and increased intracranial pressure was reported in just over half of the patients. The mean tumour size (where reported) for the group having transcranial surgery was 5.2cm; across another set of studies the median or range of tumour size was 3 to 6cm. Tumour locations, although inconsistently reported, were suprasellar or predominantly suprasellar craniopharyngiomas.

Patients who received transsphenoidal surgery presented with visual field and visual acuity deficits (68.5%), presence or absence of hydrocephalus (26.5%), and increased intracranial pressure (37%). The mean tumour size (where reported) was 2.5cm; tumours located were intrasellar craniopharyngiomas and intrasellar tumours with suprasellar extension.

The outcomes evaluated were survival, rate of gross total resection, tumour recurrence, surgical mortality, visual outcomes, neurological deterioration obesity and/or hyperphagia, diabetes insipidus, incidence of cerebrospinal fluid leaks, and meningitis.

The authors did not state how many reviewers performed the study selection

Assessment of study quality
The authors did not state they assessed methodological quality

Data extraction
Data were extracted on the outcomes as reported and presented as percentages of patients with complete data reported.

The authors did not state how many reviewers performed the data extraction.

Methods of synthesis
Weighted averages were calculated to determine the conglomerate value for all the included studies. The Fisher exact tests were used to compare proportions between the transcranial and transsphenoidal treatment groups.
Results of the review
Sixty-one studies (3,328 patients) were included in the review, comprising 48 studies of 2,955 patients (sample sizes ranging from 22 to 309 patients) who had transcranial surgery and 13 studies of 373 patients (sample sizes ranging from 10 to 76 patients) who underwent transsphenoidal surgery.

Transcranial surgery: At mean follow-up time of 5.9 years after transcranial surgery, 90.3% of the patients were still alive. Gross total resection was attained in 60.9% of operations. Perioperative deaths occurred in 2.6% of surgeries. Neurological deterioration was recorded in 9.4% of patients. Diabetes insipidus occurred in 69.1% of patients post-surgery; obesity/dysphagia was experienced by 32.2% of patients. Disease recurrence after gross total resection was 17%. There were improvements in the visual status of 47.7% of patients who had preoperative deficits and 13% of patients experienced new visual deficits after surgery.

Transsphenoidal surgery: At mean follow-up of 5.5 years after transsphenoidal surgery, 93.9% of patients were alive. Gross tumour resection was attained in 72.1% of operations. Disease recurrence after surgery occurred in 8% of patients. Perioperative deaths occurred in 1.3% of operations. Neurological deterioration occurred in 3.1% of patients. After treatment, diabetes insipidus was present in 36% of cases; 32.1% of patients had post-operative obesity of hyperphagia. Visual status improved in 85.5% of patients.

A comparison of transcranial and transsphenoidal groups prior to surgery showed that the transcranial group had less visual loss, a higher proportion of hydrocephalus, increased intracranial pressure, larger tumours and more suprasellar disease. After surgery, the patients in the transcranial surgery group had lower rates of gross total resection, more frequent tumour recurrence, higher neurological morbidity, more frequent diabetes insipidus, less visual improvements and greater deterioration in vision. There were no differences in the incidence of obesity or hyperphagia. There were non-significant trends towards less perioperative mortality and improved overall survival with transsphenoidal surgery.

Authors' conclusions
Directly comparing outcomes after transcranial and transsphenoidal surgery in paediatric patients with craniopharyngiomas may not be valid because of important differences in in populations treated with each surgery type.

CRD commentary
The review addressed a clear question and criteria for the inclusion of studies in the review were stipulated and reproducible. Two appropriate databases were searched for relevant studies, but there were no attempts to identify unpublished material. This meant that there was some risk of publication bias. The restriction of the review to articles in English language meant that there was some risk of language bias. No steps were taken at any stage of the review process to minimise errors or bias in the conduct of the review.

There was no assessment of the methodological quality of the included studies, which meant it was difficult to judge the reliability of the results. Combining the results may not have been appropriate because the results of non controlled studies were associated with a number of potential biases.

Methodological flaws in the review and the unknown quality of the included studies means the reliability of the authors' cautious conclusions is unclear.

Implications of the review for practice and research
Practice: The authors stated that the decision on which treatment approach and the planned aggressiveness of the resection should be determined on an individual basis after considering the imaging findings and the wishes of the patient and family. Both approaches could be combined to completely resect intrasellar and suprasellar portions. Transsphenoidal approaches should be considered with subtotal resection goals and planned adjuvant radiotherapy, which may spare children the potential morbidity of formal craniotomy.

Research: The authors stated that future research was required to determine the role of radiosurgery, fractionated radiotherapy, intracavitary therapies, and open surgery in the management of craniopharyngiomas of different sizes and disparate locations. Improved outcome measures and prospective reporting were required to determine the role of aggressive surgery for paediatric craniopharyngiomas.
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