

Characterisation of paediatric craniopharyngiomas in a single centre study – analysis of factors affecting recurrence rates

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Introduction

Craniopharyngioma

- A rare benign pituitary tumour arising from remnants of Rathke's Pouch (Fig.1).
- Constitutes 1-3% of all brain tumours.¹
- Recurrence rates can be as high as 90%.²

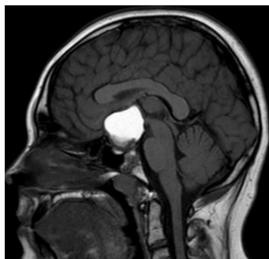


Fig. 1 – A craniopharyngioma MRI with contrast.
Source: OCDEM

Previous studies have suggested a range of factors that may affect recurrence, in particular the initial treatment modality:

- Significantly lower recurrence rates have been reported after:
 - Gross Total Resection (GTR) compared to Subtotal Resection (STR).³⁻⁵
 - STR with radiotherapy compared to STR alone.³⁻⁵
 - The role of growth hormone (GH) replacement in recurrence is contended. GH is mitogenic and anti-apoptotic, but has been associated with lower recurrence rates in paediatric populations.^{6,7}

However, evidence is limited, particularly in paediatric cases.

Project Aims:

- To characterise the population of patients with paediatric craniopharyngiomas.
- To investigate the association between different treatment modalities and subsequent recurrence rates of paediatric craniopharyngiomas.

This was achieved using historic data from the Oxford Centre of Diabetes, Endocrinology and Metabolism (OCDEM).

Materials and Methods

A retrospective single-centre study of cases at OCDEM was conducted. Data was obtained from records of 21 patients currently under follow up with craniopharyngiomas presenting in childhood (under 18 years).

Results

Clinical characteristics of the patient population are outlined in Table 1. 90% were followed up for more than eight years (median 15 years, range 5-31 years). The association of some of these characteristics with recurrence rate was calculated (Fisher's exact test) (Table 1).

Table 1: Clinical characteristics of the study population

Variable (no. of cases)		No. that Number (%)	recurred (%)	P value
Recurrence (21)	Yes	8 (38)		
	No	13 (62)		
Time to recurrence, years (7)	Median (range)	5.5 (2-19)		
Length of follow up at time of study, years (21)	Median (range)	15 (5-31)		
Age at time of surgery, years (21)	Median (range)	9 (3-18)		
Sex (21)	Male	13 (62)	7 (54)	0.085
	Female	8 (38)	1 (13)	
Operation (21)	Subtotal resection	16 (76)	5 (31)	0.595
	Aspiration	2 (10)	1 (50)	
	Fenestration	3 (14)	2 (67)	
Radiotherapy (20)	Yes	16 (80)	4 (25)	0.101
	No	4 (20)	3 (75)	
Radiation dose (16)	50Gy	9 (56)	3 (33)	0.585
	54Gy	7 (44)	1 (14)	
Growth hormone replacement (20)	Yes	18 (90)	7 (39)	1.000
	No	2 (10)	1 (50)	
Tumour histology (21)	Adamantinomous	21 (100)	8 (38)	1.000
	Papillary	0	0 (0)	
Endocrinopathies (21)	Panhypopituitism	21 (100)	8 (38)	1.000
	Single deficiency	0	0 (0)	
Secondary Tumours (21)	Yes	1 (5)	1 (100)	0.381
	No	20 (95)	7 (35)	

Surgical intervention

No significant association was found between any of the three surgical procedures undergone by these patients and recurrence rates (p=0.595) (Fig. 2).

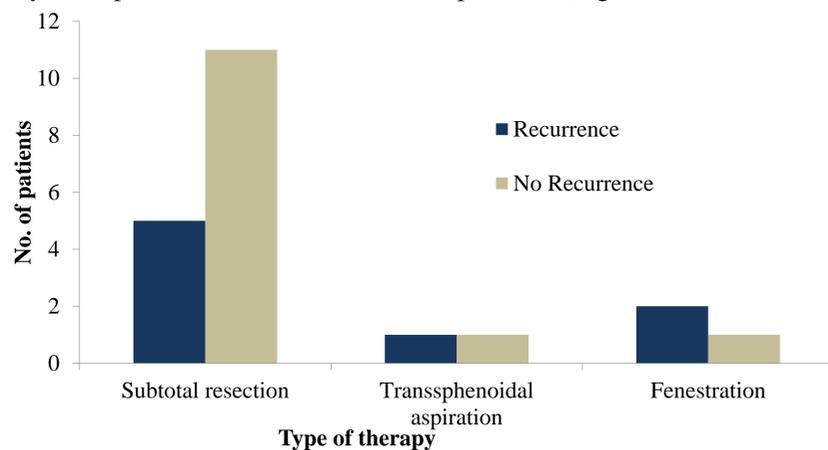


Fig. 2: The effect of different surgical treatments on the number of patients with or without craniopharyngioma recurrence. Fisher's Exact Test with Freeman-Halton extension: p=0.595.

Radiotherapy

Although there was a trend towards reduced recurrence rate with adjuvant radiotherapy, no significant association was found (p=0.101) (Fig. 3).

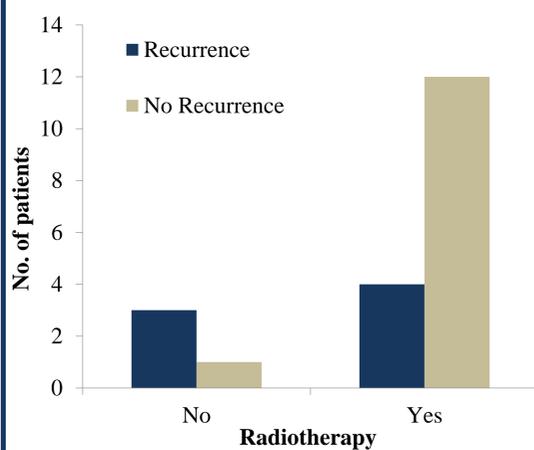


Fig. 3: The effect of radiotherapy on the number of patients with or without craniopharyngioma recurrence. Fisher's Exact Test : p=0.101.

Growth hormone replacement therapy

No significant association between growth hormone replacement after surgical treatment and recurrence rate was found (p=1.000) (Fig.4).

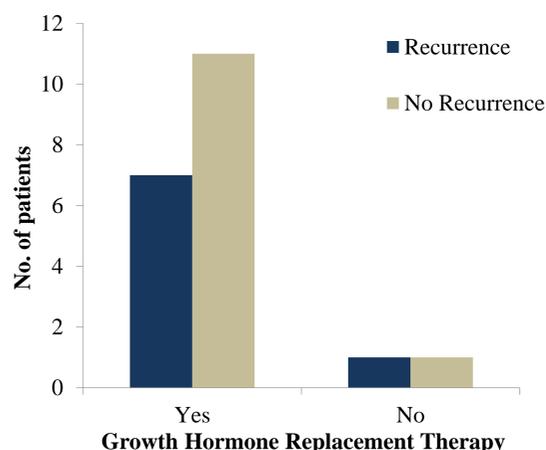


Fig 4: The effect of growth hormone replacement therapy on the number of patients with or without craniopharyngioma recurrence. Fisher's Exact Test : p=1.000.

Discussion

Conclusions

- A database of clinical characteristics of paediatric craniopharyngiomas patients at OCDEM has been constructed (Table 1).
- No statistically significant association was found between any treatment modality and recurrence rate (Figs. 2-4), in contrast to previous studies.³⁻⁶
- A substantially smaller proportion of patients who had had radiotherapy suffered recurrence compared to those who had had no radiotherapy, although this did not reach statistical significance (Fig.3).

Limitations

- The relatively small sample size.
- The potential selection bias of a retrospective study design.
- All surgical interventions studied left residual disease (no patient had GTR), which could explain the similar levels of recurrence.
- Analysis of recurrence rates did not take into account length of follow-up, which further work could assess.

Potential future work:

- A multi-centre analysis of paediatric populations, examining whether larger numbers show effects which this study is too small to demonstrate.
- Further analysis of this patient population to investigate the impact of different treatments on time to recurrence and on other long term complications including cerebrovascular disease and secondary tumours.

Understanding recurrence is vital to improve treatments decisions and patient quality of life.

Acknowledgements

Many thanks to Dr Jafar-Mohammadi for coordinating and supervising the project, and to everyone at OCDEM who provided teaching and support.

If you would like to contribute to this work, please contact miranda.rogers@some.ox.ac.uk.

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