General language abilities following management of childhood supratentorial tumour: Part I

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SUMMARY

Background. *To date few studies have investigated the impact of management for supratentorial tumour on the language abilities of children. In reporting children with brain tumour as part of a larger cohort of various aetiologies of brain injury, such studies have failed to differentiate between the causes of acquired childhood language disorders, or specifically report associated information relating to site and treatment.*

Material and methods. *The present study examined the general language abilities of six children managed for supratentorial tumour, using a comprehensive standardized general language assessment battery, including receptive and expressive components, receptive vocabulary, and naming.*

Results. *At a group level, children managed for supratentorial tumour performed below an individually matched control group in the area of general expressive language. However, at an individual case level it was revealed that only two cases exhibited specific language deficits. Reduced performance in the area of expressive language and syntax was evident in the language profile of one child treated surgically for a left parietal astrocytoma, while a child treated surgically for an optic nerve glioma demonstrated difficulties in receptive semantic abilities. The remaining four cases with similar treatments and locations demonstrated intact general language abilities.*

Conclusions. *Factors such as site, long-term presence of tumour prior to diagnosis, young age at diagnosis, and variations in time post treatment were considered to have contributed to the findings. The need for long-term monitoring of language abilities post treatment as well as larger group sizes and the investigation of higher-level abilities was highlighted.*

INTRODUCTION

The supratentorial region of the brain encompasses the intracranial structures of the brain above the tentorium cerebelli, and accounts for the location of approximately 40% of brain tumours in children (Becker & Jay 1990). Although brain tumours have been recognised as a cause of acquired language disorders in children for over three decades, until recently little information has been documented as to the language abilities of children who experience this condition, with few studies systematically investigating language disorders associated with brain tumour in children (Murdoch & Hudson 1999a). Additionally, studies that have been documented have rarely differentiated between the causes of acquired childhood language disorders (traumatic brain injury, tumour, infarct, haematoma), or reported specific and particular information such as location, treatment details, and associated

symptoms, now considered necessary to determining prognosis of language disorders caused by tumours, tumour sites and treatment modalities.

Several researchers have reported the presence of intellectual impairment in children specifically treated for supratentorial tumour (e.g. Danoff et al. 1982; Ellenberg et al. 1987; Jannoun & Bloom 1990; Mostow et al. 1991; Pierre-Kahn et al. 1983). Considering the disruption to higher functions of intelligence, it could reasonably be expected that children treated for supratentorial tumour would also demonstrate deficits in language function. A study by Mostow et al. (1991) examined the long-term survivors of central nervous system tumours of childhood and adolescents. Compared with children who had normal intellectual functioning, these authors noted that survivors considered intellectually incompetent were more likely to have had a supratentorial tumour. It was also reported that supratentorial tumours were more likely to result in deficits of higher functions involving intellect, memory and emotion than infratentorial tumours (Mostow et al. 1991). These same authors reported that overall, patients surviving childhood supratentorial tumour were at greater risk for reduced quality of life than those who had experienced an infratentorial tumour.

In a study by Danoff et al. (1982), intellectual impairment was again noted in a group of children treated for supratentorial tumour (inclusive of one patient with a pinealoma, three patients with optic nerve gliomas, and two with hypothalamic gliomas). It was suggested that the amount of radiation administered did not affect intelligence, as all six patients received local field treatment only, therefore indicating that the supratentorial location of the tumour was responsible for these findings. In their overall study of 38 children with brain tumour, Danoff et al. (1982) observed that patients with tumour involvement of either the hypothalamus or the thalamus had an increased incidence of intellectual impairment.

In contrast to these findings, a study by Mulhern et al. (1992) found no significant difference in cognitive ability between a group of children with a posterior fossa tumour and those with a tumour in the cerebral hemispheres (inclusive of the frontal, temporal, parietal, and occipital lobes, and the lateral ventricles). Mulhern et al. (1992) also observed that those children who had received local radiation for posterior fossa tumour did not differ significantly from children receiving local radiation for third ventricle tumour (inclusive of the pineal gland, hypothalamus, midbrain area, thalamus, optic chiasm, pituitary gland, and the suprasellar area) or from those receiving local radiation for tumours of the cerebral hemispheres (Ellenberg et al. 1987). Additionally, no differences were found between children receiving local radiation or no radiation for tumours in the third ventricle region, suggesting the importance of location. Despite this, Mulhern et al. (1992) acknowledged that the absence of a significant effect of tumour location was in contrast to findings reported by other authors (e.g. Ellenberg et al. 1987, Jannoun & Bloom 1990). However, Mulhern et al. (1992) suggested that insufficient patient numbers and a 30-60% increased variability among IQ scores compared with normative data may have contributed to failure to detect some differences between groups.

In describing the sequelae of intracranial ependymomas, Pierre-Kahn et al. (1983) reported that neurological sequelae were either absent or mild in 64% of children, with four of the eleven cases demonstrating a supratentorial location. More significant neurological sequelae, however, were reportedly present in six cases, two supratentorial and four infratentorial. The remaining case, a posterior fossa ependymoma, was noted by the authors to demonstrate total disability. As a total group, two-thirds of the children had an IQ less than 90.9, although Pierre-Kahn et al. (1983) did not specify outcomes according to tumour location. Speech, writing and spatial orientation deficits were reportedly present in eleven of the eighteen children examined, and only 25% of the fifteen school-aged children had a normal academic level.

Although such studies have specifically examined nonverbal intellectual abilities in children following management for supratentorial tumour, limited information relating to the impact on language post surgery and following subsequent adjuvant therapies is available. An early study conducted by Alajouanine and Lhermitte (1965) documented the language abilities of children aged between six and fifteen years with left hemispheric cerebral lesions resulting from varied aetiologies (including two participants with brain tumour) who had been diagnosed with acquired aphasia. Language deficits characterised by a reduction in the expressive area of language ability, including both oral and written modalities and gestural expression, were noted in all 32 children, in addition to a noted simplification in

syntax. Simplification of syntax has been reported to be a common finding amongst children with brain injury inclusive of the cerebral hemispheres, in addition to naming and word-finding problems and word fluency (Carrow-Woolfolk & Lynch 1982, Cooper & Flowers 1987). Other findings in these populations have been documented to include auditory comprehension and receptive vocabulary difficulties, as well as reduced performance in word, sentence, and paragraph comprehension (Carrow-Woolfolk & Lynch 1982, Cooper & Flowers 1987, Hécaen 1976).

It is noted, however, that although overall language deficits – inclusive of the aforementioned specific findings of simplified syntax, reduced oral expression, naming and word-finding difficulties, reduced auditory comprehension, and reading and writing deficits - have been associated with childhood brain trauma and/or lesions localised to the left cerebral hemisphere (Alajouanine & Lhermitte 1965, Satz & Bullard-Bates 1981, VanDongen et al. 1985), high variability in the language patterns across individuals is noted. Authors report that no particular type or group of language deficits are characteristic of such patient groups. Similarly, the severity of language impairment ranges between participants exhibiting no deficits to significant difficulties (Alajouanine & Lhermitte 1965, Carrow-Woolfolk & Lynch 1982, Cooper & Flowers 1987, Hécaen 1976, Satz & Bullard-Bates 1981, VanDongen et al. 1985).

More recently, in one of the only known reports investigating language outcomes in children following supratentorial tumour, van Lieshout et al. (1990) suggested that normal thalamic functioning had a major role in verbal memory processing. These researchers documented the case of a child who presented with language and memory disturbances as a result of radiotherapy treatment for a thalamic tumour. In particular, the case described by van Lieshout et al. (1990) evidenced a significant impairment of auditory short-term memory, auditory word comprehension, nonword repetition, and syntactic abilities, with mild disturbances of word fluency and naming. However, the most prominent feature was considered to be an auditory-verbal short-term memory deficit, with all verbal deficits appearing related to or impacted by this area (van Lieshout et al. 1990).

In another recent study, Sands, van Gorp and Finlay (1998) reported the language abilities of ten children treated with chemotherapy for brain tumour. Tumour types included pineoblastoma, glioblastoma, ependymoma, PNET, and medulloblastoma. While tumour location was not specified, the nature and pathological underpinnings of these various tumour types with tendencies for specific locations indicated that both supratentorial and posterior fossa locations were accounted for in this group. As a group, however, it was noted that the most affected area of language was in expressive skills, with impaired expressive picture vocabulary and naming skills reported (Sands et al. 1998).

Due to findings of language deficits in children with cerebral damage resulting from various forms of brain injury, together with reports of intellectual dysfunction in patients treated for supratentorial tumour, it is clear that the occurrence of language disturbances is not unlikely in children treated for supratentorial tumour. Yet few recent studies have specifically examined the language abilities of this population of children in the context of specific supratentorial tumour location, associated presenting symptoms, and treatments employed. It was therefore the aim of the present study to document the general language abilities of a group of children managed for supratentorial tumour, in order to closely examine the possible presence and nature of resulting language deficits that may emerge following tumour in this region.

METHODOLOGY

Participants
Six participants were included in the present study, three female and three male, ranging in age from three years ten months to fourteen years one month (mean age = 9.25 years, standard deviation = 3.70 years), who had been diagnosed with a tumour in the supratentorial region of the brain and had completed any treatment at least six months prior to involvement. Three of the participants (Cases 1, 2, and 3) were managed at the Mater Children's Hospital in Brisbane, Australia, while the remaining three participants (Cases 4, 5, and 6) were managed at the Royal Children's Hospital, Brisbane, Australia. All participants managed for supratentorial tumour were recruited through the Haematology/Oncology departments in both hospitals. Biographical details of these six participants are summarised in Table 1. Six control participants individually matched for age and gender (mean age = 9.36 years, standard

deviation = 3.76 years) were also included in the study. All control participants had no history of cancer, acquired brain injury, epileptic activity or seizures, or had a known history of speech/language difficulties. All twelve participants spoke English as their only language.

Table 1. Biographical data of participants treated for supratentorial tumour

Case	Gender	Age* at assessment	Age* at diagnosis	Time post treatment	Tumour type	Tumour location	Treatment	Extent of surgery	Total radiation dosage	Chemoth
1	F	7;4	3;1	4;2	juvenile pilocytic astrocytoma	left parietal lobe	S	near-total		
2	M	7;7	6;2	0;6	germ cell	pineal region	R, C		72Gy	
3	M	11;1	10;2	0;9	(a) LG glioma (b) optic nerve glioma	(a) upper tectal plate (b) optic nerve	S	endoscopic 3rd ventriculostomy		Cisplatin cyclophc bleomyci amifostin
4	F	11;7	5;8	5;11	pilocytic astrocytoma	optic nerve	S	subtotal		
5	M	14;1	10;7	3;6	ganglioglioma	left frontal lobe	S	total		
6	F	3;10	2;8	1;2**	hamartoma	hypothalamus	none			

Note: *Age and time presented in years, months; ** = age since diagnosis; S = surgery; R = radiotherapy; C = chem Gy = grays LG = low-grade.

Procedure

All six participants in the study and their parents were provided with both verbal and written (all parents and those participants of sufficient age) information describing the nature of the study and the assessments to be administered. Consent was obtained from both the parental guardian and the participant to undergo involvement in the study. Language testing was conducted in a quiet environment with limited distractions. In each case, testing was conducted over a number of sessions to reduce the influence of fatigue.

The general language abilities of each of the six cases and their individually matched peers were assessed in order to determine a comprehensive profile of general language ability. The following three tests which reflect a measure of general language skills were administered:
– either the Clinical Evaluation of Language Fundamentals – Third Edition (CELF-3) (Semel et al. 1995) or the Clinical Evaluation of Language Fundamentals – Preschool (CELF-Preschool) (Wiig et al. 1992);
– the Peabody Picture Vocabulary Test - Third Edition (PPVT-III) (Dunn & Dunn 1997);
– the Hundred Pictures Naming Test (HPNT) (Fisher & Glenister 1992).

The age of each participant at the time of testing determined whether they were administered the CELF-3 or the CELF-Preschool. Five participants (Cases 1, 2, 3, 4 and 5) and their matched controls were administered the CELF-3 (for children aged over six years of age). Specifically, Cases 1 and 2 were administered the 6-8 years version of the CELF-3 and Cases 9, 10 and 11 completed the 9+ years of age version. The remaining participant, Case 6, and the individually matched control participant completed the CELF-Preschool. Due to visual difficulties experienced by Case 4, all assessment materials were enlarged for this case according to specified visual requirements, as well as consultation with the child's parents and the child herself.

RESULTS

Group Analysis

Homogeneity of variance was tested across all parameters using Levene's Test for Equality of Variance, and was found to be non-significant (p > 0.05). However, due to the small group numbers, conservative analysis using non-parametric statistics was conducted. Consequently, Mann-Whitney U tests were employed to determine the presence of statistically significant discrepancies across all

parameters of the general language test battery between a group of six subjects managed for supratentorial tumour and their age- and gender-matched controls. These results are summarised in Table 2.
A variation in age level across participants required the use of both current versions of the Clinical Evaluation of Language Fundamentals series (CELF-3 and CELF-Preschool) as well as varied age level subtest completion

Table 2. Supratentorial tumour and control group analysis: Means (M), standard deviations (SD), and Mann Whitney U comparisons for the CELF-3/Preschool, PPVT-III, and HPNT

Parameter	Supratentorial group (n = 6)		Control group (n = 6)		Mann-Whitney U	Asymp. sig. (2-tailed)
	M	SD	M	SD		
CELF-3/Preschool						
Receptive Language Score	111.17	14.95	125.17	14.88	8.5	0.13
Expressive Language Score	101.00	13.25	122.83	12.17	3.5	0.02*
Total Language Score	105.00	13.56	124.67	11.91	4.5	0.03*
PPVT-III	105.33	16.37	117.00	8.30	10.0	0.20
HPNT (Raw score/100)	92.33	6.71	96.33	4.55	10.0	0.20

Note: * p significant at < 0.05
CELF-3/Preschool: Clinical Evaluation of Language Fundamentals - Third Edition / Preschool
PPVT-III: Peabody Picture Vocabulary Test - Third Edition
HPNT: Hundred Pictures Naming Test

requirements. Consequently, no group statistics could be calculated at a subtest level. Despite this, however, the data yielded by both the CELF-3 and CELF-Preschool could be collapsed, as the CELF-Preschool is a downward extension of the CELF-3 version. Therefore, comparisons of overall receptive, expressive, and total scores were used in group analysis.
Statistical analyses revealed no significant differences (p > 0.05) between the groups in the Receptive Language component of both versions of the CELF assessments, the Peabody Picture Vocabulary Test (Third Edition) and the Hundred Pictures Naming Test. Significant differences (p< 0.05), however, were observed between the group of children managed for supratentorial tumour and the individually matched control group on both the Expressive and Total Language components of the CELF-3/Preschool, with the children managed for supratentorial tumour exhibiting comparatively reduced scores.

Individual Analysis
The size of the group necessitated the examination of participants managed for supratentorial tumour on an individual basis, in order to analyse variability and potential language difficulties amongst cases that may have been masked at group comparison level. It has also been previously reported that differing treatment approaches and individual presentations of tumour types and treatments undergone may also contribute to the occurrence of individual variations, with additional comprehensive information gained at an individual level (Murdoch & Hudson-Tennent 1994). Consequently, it is important that each participant is examined individually in the context of the many potential individual factors often present in this population of children. In this level of analysis a comparison of the individual standard scores of each participant managed for supratentorial tumour to the normative data provided for each assessment was carried out. Overall, four cases were found to have normal performance across all general language measures, while two cases (Cases 1 and 4) were noted to exhibit reduced performance in areas of general language.

Case 1

From the age of twenty months, Case 1 exhibited subtle neurological changes, with delays in gross motor and speech skills reported. At that time, Case 1 also began experiencing left-sided headaches and her mother noted a tilting of her head to the right. Problems with co-ordination and walking, reduction in fine and gross motor skills, and visual problems were also reported. Case 1 was diagnosed with a very large left parietal tumour at the age of three years one month. A cranial MRI revealed a 7.2 x 7 x 6.5 cm solid tumour in the left basal gangliomic region with a small amount of oedema, gross deformity of the left lateral ventricle, high-grade interforaminal obstruction and moderate right lateral ventriculomegaly (see Figure 1). At this point, Case 1 had experienced a 12-month history of headaches and vomiting.

A biopsy and partial resection was conducted, and histology confirmed a juvenile pilocytic astrocytoma. While extensive removal was carried out, a postoperative
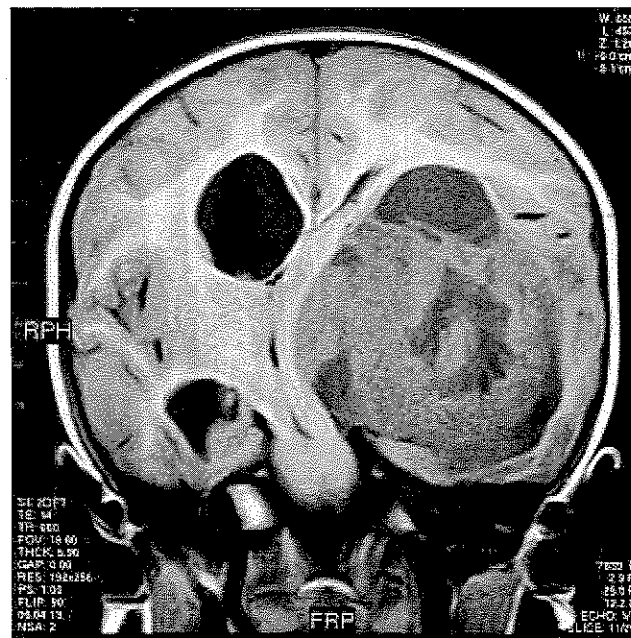

Fig. 1. Case 1 at diagnosis: T1 weighted axial MRI scan indicating a 7.2 x 7 x 6.5 cm largely solid tumour in the left basal ganglionic region with some associated oedema. Gross deformity of left lateral ventricle, relatively high grade interforaminal obstruction and moderate right lateral ventriculomegaly

CT suggested a small amount of residual tumour medially against the thalamus. Residual features included a right visual field defect and right-sided hemiparesis with the arm more involved than the leg. Facial weakness, which was evident for a number of years, also persisted. Three years following surgery, a dense right-sided homonymous hemianopia was reported as a residual symptom. No radiotherapy or chemotherapy treatment was conducted.

Case 1 underwent speech therapy subsequent to treatment, with language abilities assessed on the CELF-Preschool at both 12 months and 24 months post-surgery. At 12 months post, at a chronological age of four years one months, general language abilities were considered within the normal range (Receptive Language Standard Score = 114, Expressive Language Standard Score = 90, Total Language Standard Score = 102). However, at a subtest level, performance on the Formulating Labels subtest, which evaluates word knowledge and naming abilities, was considered slightly below the normal range (standard score = 6). The speech pathologist noted at the time that a mild degree of difficulty was evident with the ability to name pictures representing nouns and verbs. Therapy focussing on this area of difficulty was then undertaken. At 24 months post treatment, Case 1 was re-assessed and performed

within normal limits on the Receptive Language component of the CELF-Preschool (standard score = 114), and above the normal range on both the Expressive Language component (Standard Score = 122) and Total Language Score (standard score = 118). At this time, performance specifically
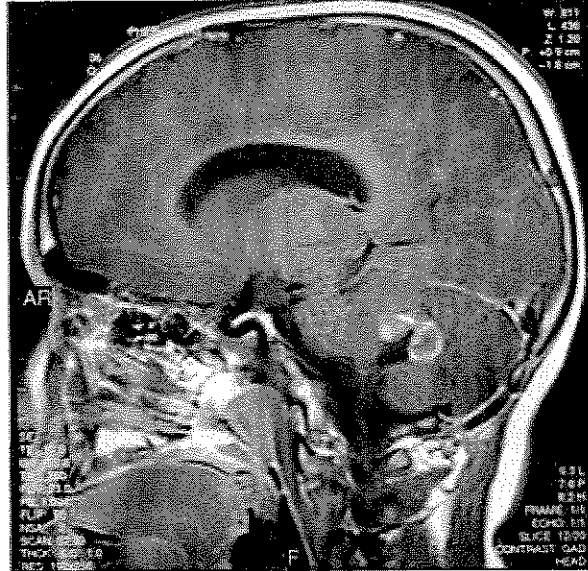
Fig. 2. Case 1 at language testing (4 years 2 months post treatment): T2- weighted sagittal MRI scan demonstrating previous left temporoparietal craniotomy and partial resection of left temporal pilocytic astrocytoma. Post-surgical encephalomalacia with residual tumour noted. Overall appearances stable with no significant progression or evidence of new disease

on the Formulating Labels subtest (standard score = 15) indicated above average performance. At the age of 7 years 4 months (4 years 2 months following surgery), Case 1 completed a general language assessment battery for the present study. A MRI of the brain taken 3 months prior to this assessment indicated post surgical residual tumour, with stable overall appearance compared to the previous examination three months earlier (with no significant progression or new disease) (see Figure 2). The current assessment revealed that while the composite Receptive Language score on the CELF-3 was considered to fall within the normal range, the Expressive Language component was determined to be more than one standard deviation below the mean (see Table 3). At the subtest level, the receptive subtest, Sentence Structure, which evaluates the comprehension of structural rules at the sentence level, and the expressive subtest, Formulated Sentences, which assesses the formulation of simple, compound, and complex sentences, were both considered well below the normal range. For a child in Grade 2, Case 1 also exhibited reduced naming abilities that were considered to be one standard deviation below the mean on the HPNT.

Table 3. Individual general language assessment results (represented in standard scores) of Cases 1-5 managed for supratentorial tumour on the CELF-3/Preschool, PPVT-III, and HPNT

Tests	Case 1	Case 2	Case 3	Case 4	Case 5
CELF-3					
Receptive Language	96	120	118	92	110
Concepts & Directions	13	13	11	12	12
Word Classes	11	12	15	5*	11
Semantic Relationships^	-	-	13	9	12
Sentence Structure#	4*	15	-	-	-
Expressive Language	84*	102	120	88	104
Formulated Sentences	5*	9	15	10	12
Recalling Sentences	9	11	12	7	9
Sentence Assembly^	-	-	13	7	11
Word Structure#	8	11	-	-	-
Total Language	89	111	120	89	107
PPVT-III	88	111	112	83*	122
HPNT (Raw score / 100)	84*	95	99	94	98

Note: # = Level 1 subtest variation; ^ = Level 2 subtest variation; * = below normal range (Overall receptive, expressive, and total standard scores (in *italics*) are represented by a normal range of 85 - 115; Subtest standard score normal range = 7-13).
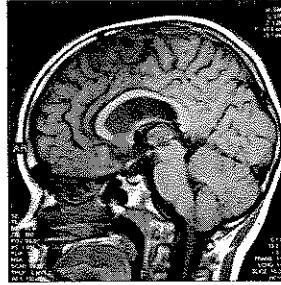
Case 2

Case 2 was diagnosed with a germ cell tumour in the pineal region at the age of 6 years 2 months (see Figure 3). Presenting symptoms included precocious puberty and obstructive hydrocephalus, for which a shunt was inserted. Surgical resection was not undertaken. Three weeks following shunt insertion, Case 2 began a course of radical radiotherapy to the pineal gland which involved a dose of 16 Gy in 10 fractions over sixteen days. As the tumour did not respond, the full extent of tumour was treated to an additional dose of 56 Gy in 32 fractions using a lateral and superior field over approximately three months. Chemotherapy commenced four months later. Six cycles of chemotherapy were employed over a period of six months, with the chemotherapeutic agents cisplatin, etopophos, cyclophosphamide, bleomycin, and lenograstin administered in cycles 1 and 2, and the agents cisplatin, amifostine, etopophos, cyclophosphamide, bleomycin, and lenograstin in cycles 3 to 6.

Six months following completion of chemotherapy treatment, Case 2 underwent general language testing for the present study. Brain MRI 3 weeks prior indicated a stable tumour compared to the previous scan 5 months before, with a minor reduction in ventricular size noted (see Figure 4). Across all language assessments, Case 2 was considered to have general language abilities that were within or above the normal range (see Table 3).
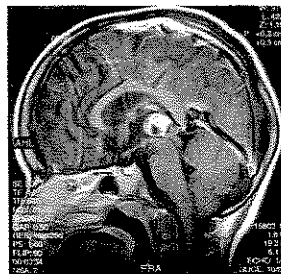
Fig. 3. Case 2 at diagnosis: T1 weighted sagittal MRI scan indicating a pineal region tumour with associated obstructive hydrocephalus. Dilatation of the third and both lateral ventricles. Mass arises from floor of posterior aspect of third ventricle on anterior aspect of the tectum

Fig. 4. Case 2 at language testing (six months post treatment): T1 weighted sagittal MRI scan demonstrating stable pineal region tumour following radiotherapy treatment

Case 3

At the age of 10 years 2 months, Case 3 presented with significant hydrocephalus due to a 1.2 cm focal intraparenchymal low-grade glioma in the tectal plate of the upper midbrain obstructing the aqueduct, as well as a right optic nerve glioma secondary to neurofibromatosis. Case 3 had a history of neurofibromatosis that was suspected at birth, in addition to persistent headaches and reported clumsiness prior to diagnosis. Hydrocephalus was treated by an endoscopic third ventriculostomy. No further treatment for either identified tumour was undertaken, with Case 3 being monitored on a yearly basis. No progression of either of the lesions was noted up until the time of testing. Residual features recorded one month prior to testing included occasional headaches, a large head and persistent enlargement of the ventricular system, some stumbling, and poor co-ordination. Small lesions in the basal ganglia regions bilaterally were also noted at this time, reflecting known neurofibromatosis-associated hamartomas.

Eleven months following surgery to relieve hydrocephalus, language testing was carried out at the age of 11 years 1 month. All general language abilities, as measured by the CELF-3, PPVT-III, and the HPNT were considered to be well within or above the normal range (see Table 3).

Case 4

Case 4 was diagnosed with a suprasellar / left optic nerve glioma at the age of 5 years 8 months, following a history of deteriorating vision and global developmental delay. On presentation, Case 4 demonstrated nystagmus and visual abnormalities. A biopsy was carried out and histology confirmed a pilocytic astrocytoma. A subtotal resection of the tumour was carried out 9 months later via a left frontal craniotomy. Residual vision remained following surgery. Neither radiotherapy nor chemotherapy was employed.

At 5 years 11 months post treatment, Case 4's general language abilities were assessed for the present study at the age of 11 years 7 months. While overall general receptive and expressive language abilities as measured by the CELF-3 were considered within the normal range, the receptive subtest Word Classes, which assesses the ability to perceive relationships between words that are categorised by part-whole and semantic class features and synonyms and antonyms, was considered to be below the normal range (see Table 3). Receptive vocabulary abilities as assessed by the PPVT-III were also considered below the normal range at more than one standard deviation below the mean.

The highest age range available on the HPNT with respect to normative data was 7 years 1 month to 9 years 2 months (with a mean of 7 years 7 months). The mean accuracy score for this group of children (n = 260) was 84.11, with a standard deviation of 9.85. Therefore, while Case 4's raw accuracy score of 94 would be considered within the normal range for these lower age limits, it was impossible to ascertain whether this level of performance represented intact or reduced naming skills for this

participant.

Case 5

Case 5 was noted to have experienced a long history of headaches since the age of 4 years, with an

increase in frequency over the 6 months prior to diagnosis (featuring 2 particularly extreme headaches lasting a day long each during the 2 weeks directly prior to diagnosis). Headaches were reportedly constant and in the left retroorbital area, and had been associated with vomiting. Case 5's mother reported a change in personality and an increase in aggression over the few months prior to diagnosis, despite a reported gentle disposition previously. Case 5 was also reported to be experiencing learning difficulties following a period of no academic problems for the first 3 years of schooling. He had repeated Grade 4, and was in Grade 5 at the time of diagnosis. Although no seizures were experienced, occasional dizzy spells were reported. Case 5 underwent surgical removal of a left frontal ganglioglioma, at the age of 10 years 7 months. No radiotherapy or chemotherapy treatment was administered. Case 5 was seen for general language testing at the age of 14 years 1 month, 3 years 6 months following surgical treatment. Performance across all assessments indicated an absence of deficits, with all scores falling within or above the normal range (see Table 3).

Case 6

Case 6 was diagnosed with a hyperthalamic hamartoma with secondary precocious puberty at the age of 2 years 8 months. Treatment was designed to primarily address the symptoms of precocious puberty. Therefore, neither surgery, radiotherapy, nor chemotherapy was involved in the management of this case. Case 6 received monthly injections of hormone therapy (7.5 mg Depo-Lucrin) for 1 year prior to involvement in the study, with 3- to 6-month check-up appointments. Language testing was conducted at the age of 3 years 10 months, 1 year 2 months post diagnosis. General language abilities as measured by the CELF-Preschool, PPVT-III, and HPNT were noted to be within or above the normal range (see Table 4).

Table 4. Individual general language assessment results (represented in standard scores) of Case 6 managed for supratentorial tumour on the CELF-3/Preschool, PPVT-III, and HPNT

Tests	Case 6
CELF-3	
Receptive Language	*131*
Concepts & Directions	15
Word Classes	13
Sentence Structure	16
Expressive Language	*108*
Formulated Sentences	10
Recalling Sentences	13
Word Structure	11
Total Language	*117*
PPVT-III	*114*
HPNT (Raw score / 100)	84

Note: Overall receptive, expressive, and total standard scores (in *italics*) are represented by a normal range of 85 - 115; Subtest standard score normal range = 7-13.

DISCUSSION

Findings from a group level analysis revealed significantly reduced performance by a group of children managed for supratentorial tumour when compared to a group of individually matched peers, on both the Expressive and Total Language components of an assessment of general language abilities (CELF-

3/Preschool). Disturbances in the language function of children with brain tumour have been previously documented in a group of studies of acquired aphasia examining children with varied aetiologies, including head injury, cerebrovascular accidents, and epilepsy. Cooper and Flowers (1987) represented such a study, in which the language and academic abilities of a group of 15 children with brain lesions inclusive of head injury, stroke, brain infection, and posterior fossa tumour (in 1 child), were examined. As a group, these children were reported to perform significantly below their normally developing matched peers on measures of language, particularly in word, sentence, and paragraph comprehension, naming, oral production of complex syntactic constructions and word fluency. Within the group, however, Cooper and Flowers (1987) reported that language difficulties ranged from an absence or mild level of deficit to a significant disorder, as was observed across individual analysis in the present study. Specifically, the findings of expressive language deficits across the current group of children with brain tumour are consistent with the findings of other authors. An early study by Alajouanine and Lhermitte (1965) examined the language abilities of 32 children with varying hemispheric lesions (including 2 children who had received surgical treatment for an astrocytoma). These authors reported that a reduction in expressive language abilities in both the oral and written modalities characterised the most significant language deficit of the subject group, with expressive deficits noted to be present in all 32 children studied (Alajouanine & Lhermitte 1965). A pattern of reduced oral expression is also commonly reported in studies of acquired language disorders in children with mixed aetiologies (Carrow-Woolfolk & Lynch 1982, Satz & Bullard-Bates 1981, VanDongen et al. 1985). It is possible that the group result in the present study is related to the impact of a supratentorial tumour on expressive language areas in the left hemisphere, as authors such as Satz and Bullard-Bates (1981) and VanDongen et al. (1985) attributed language findings in populations of children with brain injury of various aetiology to a left hemisphere location.

The specific expressive language difficulty observed in the present group however is slightly contrasted by an earlier study which reported difficulty in both verbal processing and expressive tasks by a child with thalamic tumour (van Lieshout et al. 1990). In the case study reported by these authors, difficulties were particularly evident in auditory short-term memory and auditory word comprehension, with expressive disturbances including non-word repetition and syntactic abilities as well as mild impairment of word-finding and naming. van Lieshout et al. (1990) attributed findings of reduced verbal processing to the localised thalamic region, indicating that the findings in this case may have been specific to this particular supratentorial location. Sands et al. (1998), however, who documented the language abilities of 10 children following chemotherapy treatment for various tumour types in both the supratentorial region and posterior cranial fossa, reported a greater degree of impairment in the expressive area of language compared to receptive abilities, particularly in expressive picture vocabulary and naming. Although some significant findings were yielded upon group comparison analysis in the present study, it is important to acknowledge that at the single case level, examination of performance across all assessments of general language indicated specific areas of impairment for just two of the six children (Cases 1 and 4) managed for supratentorial tumour. Although this equates to 33% of the target group, it is suggested that in light of the small numbers, group results should be interpreted with caution. Variability amongst the group of children managed for supratentorial tumour in the present study was highlighted by language performance in the individual analysis, and in particular the patterns of language ability demonstrated between Cases 1 and 4. Not only did these two cases yield variable performance by being the only participants in the present study to demonstrate any general language deficit, but they were also noted to each present patterns of general language deficit that were somewhat distinct from each other. Variability of language performance was also noted by Murdoch and Hudson-Tennent (1994) and Murdoch and Hudson (1999b), in examining children treated for posterior fossa tumour, and suggested that the establishment of typical language patterns in this population may be difficult.

The first case, Case 1, exhibited reduced performance on the overall Expressive Language component of the CELF-3 with a standard score below the normal range. Findings of reduced expressive language abilities is representative of the group level findings as well as previous reports cited above (Alajouanine & Lhermitte 1965, Satz & Bullard-Bates 1981, VanDongen et al. 1985). At a subtest level, skills assessed by the expressive language subtest Formulated Sentences were considered below the normal range. As

the Formulated Sentences subtest examines the planning and formulating of descriptive sentences, difficulties in this area may indicate an impairment in the generative language aspects relating to planning and producing sentences for conversation, classroom discourse, academic interactions, and written language (Semel et al. 1995). Case 1 was observed to have particular difficulty with all items involving adjectives, conjunctions (co-ordinating, subordinating, and correlative conjunctions), conjunctive adverbs, or phrases.

Case 1 was also noted to perform below normal limits on the receptive subtest of Sentence Structure. The Sentence Structure subtest examines a child's ability to comprehend the changes in meaning that arise from different sentence structures (syntax) and to analyse the syntactic features of error responses (Semel et al. 1995). Such a skill is considered essential in pre-reading, early reading and early writing activities (Semel et al. 1995). Problems in this area may indicate difficulties in expression and comprehension of spoken and written language, with difficulties integrating both the surface structure and deep structure of sentences (Semel et al. 1995). Case 1 was noted to experience most difficulty in items involving structures such as negation, prepositional phrases, indirect objects, infinitives, relative clauses, subordinate clauses, interrogatives, and compounds. It is also noted that both subtests that were difficult for Case 1 involved syntax. This finding supports previous reports in which simplified syntax was documented to occur in populations of children with left hemispheric lesions of mixed aetiology inclusive of brain tumour (Alajouanine & Lhermitte 1965, Carrow-Woolfolk & Lynch 1982, Satz & Bullard-Bates 1981, VanDongen et al. 1985), as was the tumour location in the case of Case 1.

Difficulties in naming were also noted in the general language profile of Case 1. This was represented by reduced performance on the Hundred Pictures Naming Test, which is designed to assess lexical extent. Two particular error response categories were noted, including providing the incorrect name and supplying a "don't know" response. Most error responses in which an incorrect name was given were observed to be in one of two semantic categories, coordinate (e.g. feather instead of leaf, or toaster instead of iron) or function (king's hat instead of crown). Two out of sixteen errors were noted to be visual-perceptual in origin (e.g. plate/wheel instead of button). However, almost as many "don't know" responses were yielded as incorrect names, reflecting that for these items the child either did not recognise the item, did not know the name, or did not want to guess. This type of response may indicate a limited lexicon, difficulty in learning and retrieval of names, or a more general difficulty in categorising and retrieving information (Fisher & Glenister 1992). A finding of naming difficulties in this case supports several previous studies reporting reduced naming and word-finding difficulties in the language performance of children diagnosed with acquired aphasia resulting from brain lesions of varying aetiologies (Carrow-Woolfolk & Lynch 1982, Cooper & Flowers 1987, Hécaen 1976, Satz & Bullard-Bates 1981, VanDongen et al. 1985).

Case 1's performance on assessments of general language ability that were administered 12 and 24 months post-treatment as part of speech pathology intervention revealed that the present assessment results represented a significant decline in expressive language ability. A standard score of 122 on the Expressive Language component of the CELF-Preschool administered at 24 months (following a 12 month period of speech pathology intervention) represented intact to above average abilities, compared to reduced performance reflected by a standard score of 84 (at 4 years 2 months post treatment) in the present study. While still considered within normal limits, it is also noted that the Receptive Language abilities, found to be almost one standard deviation above the mean at both 12 and 24 months post treatment (both standard scores were 114), were reduced to the current standard score of 96 in the 2 years 2 months since the previous assessment. Similarly, the Total Language Score that had improved at 24 months post treatment from 102 (at 12 months post) to 118 (24 months post) has also comparatively reduced to the current level of 89, despite still being considered within the normal range. Such comparisons with previous language results may indeed reflect a slowing of development in general language abilities, particularly in the expressive language of this participant. Bates et al. (1992) documented findings from a longitudinal study investigating the language development of children who had been diagnosed with hemispheric lesions prior to the age of six months. Delays in babbling, preverbal communication, vocabulary and syntax were initially observed, with delays in both comprehension and production of speech regardless of site, size, or side of lesion in the hemisphere

occurring at the age of 12 to 18 months. At age 18-36 months, expression was noted to be increasingly impaired in children with left-sided lesions, particularly associated with the more posterior hemispheric region (Bates et al. 1992).

Additionally, in examining the cognitive function of children with brain tumour, Carlson-Green et al. (1995) documented that the greater the time since diagnosis at the time of assessment, the greater the tendency to exhibit lower intelligence scores than children who had been diagnosed more recently prior to assessment. Such findings are therefore also likely to have implications for associated reductions in language function. At present, however, there is a paucity of longitudinal data relating to language function post treatment for supratentorial tumours. Consequently, there is a need for more research in this area before any definitive conclusions can be made.

In addition to the specific impairments observed in Case 1, individual case analysis also revealed specific areas of difficulty in the general language abilities of a second case (Case 4). At a subtest level, particular difficulty was noted in the Word Classes subtest in the Receptive Language component of the CELF-3. The Word Classes subtest of the CELF-3 assesses the ability to perceive relationships between words that are categorised by part-whole and semantic class features and synonyms and antonyms, which is required for classroom listening and reading comprehension (Semel et al. 1995). A memory component is also present in this subtest, as the child is required to select two related responses from three or four words presented at one time. According to Semel et al. (1995), difficulties in this assessment may indicate that a child does not associate related words automatically or efficiently.

Reduced receptive vocabulary skills were also noted in the general language profile of Case 4, indicated by performance that was noted to be more than one standard deviation below the normal range on the PPVT-III. Difficulties in receptive vocabulary were also reported to be evident in a 10-year-old child described by Carrow-Woolfolk and Lynch (1982), who had undergone surgery for a supratentorial tumour. It is interesting to note that the above components of general language relate to difficulties in areas of receptive language involving semantics and word meanings, and may represent a general area of weakness for Case 4 in particular.

Several factors may have contributed to the presence of the reported deficits in both Case 1 and 4, including site, type, and size of tumour, long-term existence of the tumour, age at diagnosis, and the effects of the treatments employed. Several studies have reported cognitive and language findings associated with a left tumour location. Carrow-Woolfolk and Lynch (1982) reported a case study of a ten-year-old child who had undergone surgery for a left parietal tumour which had been present for an extended period of time (up to two years prior to treatment). Assessments carried out two years following surgery indicated comprehension on the Peabody Picture Vocabulary Test that corresponded to an IQ of 70. Additionally, this patient was noted to be unable to respond to questions about a paragraph that had been read to her, with results from a vocabulary test reportedly placing her in the "dull-normal range" (Carrow-Woolfolk & Lynch 1982). Speech was also observed to be limited to single words or short phrases, and visual perceptual skills were considered limited (Carrow-Woolfolk & Lynch 1982). It was also noted in a study by McFie (1961), where patients were grouped according to site of cerebral lesion, that the mean IQ for both the left parietal and the right frontal groups was noted to be the most reduced (mean IQ = 73.3 and 64.0 respectively) compared to the left frontal (mean IQ = 89.3), right and left temporal (mean IQ = 88.8 and 91.3 respectively), and right parietal region (mean IQ = 88.6) groups. Such research suggests that cognitive function may be most affected following parietal region surgery, which has potential implications for reductions in language function.

Post-operative language disturbances associated with a left parietal region location were also reported in a 10-year-old boy with an arterio-venous malformation (Tanabe et al. 1989). Observed deficits were characterised by decreased spontaneous and conversational speech, literal paraphasias, mild word-finding difficulty, and normal comprehension. These findings of disturbances to expression in the presence of comparatively intact comprehension are similar to the performance noted for Case 1, and are therefore likely to be related to the direct impact on structures seen in this case. The above evidence suggests that the present findings may reflect site of tumour in the left supratentorial region. It is also particularly likely in the presentation of Case 1 that the large size of the tumour noted in the left hemisphere, with obvious distortion of the cerebral structures noted on the diagnostic MRI, would have clearly had a direct impact on the language centres of the left cerebral hemisphere. The

considerable size of the tumour in this case also indicated the length of presence and therefore is related to impact on language development.

In support of findings noted for Case 4, Danoff et al. (1982) reported the presence of intellectual impairment in three patients with optic nerve gliomas. As these authors reported that the amount of radiation administered in these cases did not affect intellectual functioning, since all these patients received local-field treatment only, it was suggested that location was a primary factor in the reported deficits (Danoff et al. 1982). Cognitive deficits noted in this study cannot be excluded from consideration of language findings in the present study, however, as the two functions are often not exclusive in patients.

Specific symptoms associated with the presence of a supratentorial astrocytoma have been widely reported to be relative to the location and local effects of the tumour (e.g. Becker & Yates 1986, Heideman et al. 1993, Yachnis 1997). Thus the left-sided location of the astrocytoma in Case 1 and the left of midline location in Case 4 may have influenced the presence of the particular general language impairments observed. As language functions are currently acknowledged to be lateralised from birth, damage to supratentorial language areas during the process of language acquisition is considered to result in some form of impairment (Martins 1997). It is therefore suggested that a tumour in the left frontal or parietal region in the two cases in the present study may have either resulted in direct impact on language areas of the brain due to the mass effect of the tumour itself or oedema, or via the effects of a consequent increased intracranial pressure in the local area in both cases.

The long-term existence of symptoms such as subtle neurological changes, headaches, head tilting, difficulties in co-ordination, delayed fine and gross motor skills, delayed speech skills, visual problems, and facial weakness noted in the history of Case 1, together with a reported lengthy history of deteriorating vision and global developmental delay for Case 4, indicated that the tumours in both cases may have been present for an extended period of time prior to diagnosis. A slow-growing tumour, juvenile pilocytic astrocytoma, was diagnosed to be present in both cases and may have contributed to the long-term presence of increased intracranial pressure. It is the presence of increased intracranial pressure that has been considered responsible for many of the neurological, neuropsychological, and intellectual impairments evident in patients surviving childhood brain tumours (McWhirter & Masel 1987, Silverman & Thomas 1990). In cases such as our Cases 1 and 4, when a slow-growing tumour may be present for several years in addition to the gradual appearance of clinical symptoms, the effects of an associated extended period of long-term increased intracranial pressure may produce prolonged effects (McWhirter & Masel 1987). However, it has also been documented that even a short period of increased intracranial pressure may produce damage (McWhirter & Masel 1987). The specific general language deficits observed in Cases 1 and 4 could, therefore, be attributed to the long-term existence of an increase in intracranial pressure as demonstrated by the prevailing presence of symptoms prior to diagnosis.

The effect of young age at diagnosis/treatment on intelligence is also well documented (Carlson-Green et al. 1995, Heideman et al. 1993, Mostow et al. 1991, Mulhern et al. 1992), and may have also contributed to language findings in Cases 1 and 4. When increased intracranial pressure occurs in the first few years of life, developmental delays have been reported to be a frequent early sign followed by intellectual declines (Heideman et al. 1993). Mulhern et al. (1992) indicated that cognitive performance is directly and negatively impacted by young age at treatment for brain tumour. This finding was supported by Carlson-Green et al. (1995), who found in their examination of cognitive ability following brain tumour that children who were older at the time of diagnosis performed better on measures of intelligence and academic achievement than younger children. Although an earlier report by Vargha-Khadem, O'Gorman, and Watters (1985) documented more profound impairments in children acquiring left hemisphere lesions after the age of five years, these authors acknowledged that subtle language deficits persisted in children with early left hemispheric injuries. Considering that diagnosis and treatment occurred at a young age in both cases, it is conceivable that age may be another factor which predisposed the two cases in the present study to subsequent language deficits post treatment.

Despite a significant difference at group level between six children managed for supratentorial tumour and their age and gender matched controls on both the Expressive and Total Language components of the CELF-3, it is notable that 4 of the 6 cases (Cases 2, 3, 5 and 6) performed within the normal range

across assessments of general language abilities. This is despite the use of a combination of both a high total dosage of radiotherapy as well as chemotherapy in treating Case 2, a left-frontal tumour location in Case 5, and known hydrocephalus in both Cases 2 and 5. Consequently, determining exactly which factors directly influenced the deficits observed in the two cases with difficulties is not clear. Considering the low incidence of deficits identified, the small population of children who present with supratentorial tumours and the wide variety of different treatment regimes and tumour sites, it may always be difficult to accurately determine factors which influence the occurrence of language deficits in this population. It is suggested, however, that several factors other than absence of neural damage may be responsible for the lack of observable general language effects in the four children in the present study. These include the absence/ extent of treatment, length of time post treatment, age at diagnosis/treatment, and test sensitivity. As mentioned previously, the greater the duration of time post diagnosis, the greater the tendency to exhibit lower intelligence (Carlson-Green et al. 1995). Therefore, the length of time post treatment in the remaining cases treated may not have yet been great enough for each individual to account for possible long-term effects. In fact, the language abilities of Case 2, the only participant in the present analysis to have received both radiotherapy and chemotherapy, were tested at just six months post treatment. Given the dosage of radiotherapy that this case received and the fact that it was combined with a subsequent program of chemotherapy in treatment of the pineal germ cell tumour, it is highly possible that later adverse effects on language function may appear. Long-term effects subsequent to radiotherapy have been reported to occur up to 14 years later (Lee & Suh 1977) with a peak at 3 years following treatment (Heideman et al. 1993, Silverman & Thomas 1990). Additionally, reports of neuropathological changes to the brain have been associated with dosages greater than 50 Gy administered over five or more weeks (Barrett & Donaldson 1992; Heideman et al. 1993). As Case 2 received a total dosage of 72 Gy, evidence of changes such as calcification may indeed manifest at greater time post treatment, with unfortunate effects on language function.

An additional factor that may have contributed to a lack of findings in the remaining four cases may have been due to the limitations of the general language assessments administered, in their capacity to explore the subtle language abilities that may be represented by high-level language skills. Therefore, it is likely that these cases would also benefit from further exploration into high-level ability, since such assessments may reveal an impairment of high-level skills that have not yet been accessed by the current general language testing. Additionally, further monitoring of general language abilities would be of great benefit to ensure that the development of any possible or additional later effects in general language function may be accounted for, particularly in children who receive aggressive treatment combinations.

CONCLUSION

The present analysis established that a reduction in general expressive language skills, as well as a comparatively reduced total overall language score, occurred at a group level when comparing a group of six children managed for supratentorial tumour with a group of individually matched peers. However, at an individual level, only two participants revealed evidence of general language deficits when compared to the normative data. While reduced performance in the area of expressive language and specific difficulties in syntax were evident in a child who had undergone surgical treatment for a left parietal astrocytoma, deficits in receptive semantic abilities were noted in another child who had been treated for an optic nerve glioma. The remaining four cases with largely similar profiles in treatment and various tumour locations, which included similar sites to those with deficits, demonstrated intact general language abilities.

Factors such as site and type of tumour, long-term presence of tumour prior to diagnosis, young age at diagnosis and treatment, and the varied duration post treatment were considered as potential contributing factors to the language findings in these cases and an absence of findings in the four children with intact general language skills. It was also proposed that perhaps the failure to detect individual language subtleties in the remaining four cases may have been related to the assessment parameters used, which were limited to assessment of general language abilities. Future research with

larger group sizes is necessary to further analyse the patterns of contributing factors in this population, and to build on gaining a greater understanding of language deficits that may occur following supratentorial tumour in children. Additionally, the pattern of declining function observed in Case 1 indicates that longitudinal studies documenting the resolution patterns in this population would also have great clinical value for ongoing treatment and management. Long-term monitoring of children with and without deficits is still advised, given the potential for difficulties to occur at a greater time post

diagnosis/treatment as observed in the single case in the present study.

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