SHORT COMMUNICATION

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Familial diagnostic experiences in paediatric oncology

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Background: Diagnostic delays may not have significant prognostic implications in paediatric oncology, but psychological impacts remain understudied.

Methods: Interviews exploring diagnostic experiences were conducted with childhood cancer survivors ($n = 19$), parents ($n = 78$) and siblings ($n = 15$).

Results: Median diagnostic time was 3 weeks. Participants described a mixture of rapid diagnoses (28.9%), plus delayed appraisal intervals (that is, parent- or patient-associated diagnostic delays; 40.0%) and diagnostic intervals (that is, healthcare-associated delays; 46.7%). Families experiencing delays described guilt and anger and deleterious impacts on the family–clinician relationship. Some believed delays impacted on treatment and prognosis.

Conclusions: The effect of the diagnostic experience can be considerable.

Non-specific presenting symptoms and the rarity of childhood cancer can result in diagnostic delays (Dang-Tan and Franco, 2007). As described by Weller *et al* (2012), these can result from prolongation of the patient/parent appraisal interval (time from symptom onset to first medical presentation, that is, 'parent- or patient-associated delays') or the diagnostic interval (time from first presentation to the time of histopathological diagnosis or first diagnosis-specific healthcare, that is, 'healthcare-associated delays'). Numerous risk factors for delays have been investigated for both paediatric and adult cancers, with cancer type and stage, parental education, age, nature of presenting symptoms and understanding of symptom significance implicated (Dang-Tan and Franco, 2007; Quaife *et al*, 2014).

Poorer outcomes are often attributed to long diagnostic times and many paediatric medical malpractice suits revolve around alleged delays (Wilne *et al*, 2013). However, although rapid diagnoses improve survival in many adult cancers (Allgar and Neal, 2005) and in retinoblastoma (Derkinderen *et al*, 1989; Rodrigues *et al*, 2004), the prognostic implications of diagnostic delays on other paediatric cancers remain unclear (Brasme *et al*, 2012; Loh *et al*, 2012; Wilne *et al*, 2013).

The implications of delay on aspects other than survival have not been widely studied. Few articles address the psychological challenges associated with childhood cancer diagnosis (Dixon-Woods *et al*, 2001; Clarke and Fletcher, 2005; Gibson *et al*, 2013). These studies suggest that significant delays may have profound emotional impacts; however, they do not explore interviewees' perceived implications of delays. This study therefore documented the diagnostic experience of families of children treated at Sydney Children's Hospital, an Australian paediatric tertiary referral hospital. It aimed to highlight the psychological implications of different diagnostic experiences and document whether families perceive delays as having important implications for treatment and diagnosis.

MATERIALS AND METHODS

Participants. Medical records were utilised to identify families of children completing cancer treatment between January 2007 and January 2010. Parents were invited to participate if their child was

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under 18 years at diagnosis, and had completed the treatment with a curative intent within the past 5 years. Survivors and siblings over 12 years were also invited. After receiving ethics approval, eligible families received an invitation letter and were provided with a return-paid 'opt-in' card. Parental consent was obtained for participants under 18 years.

Data collection. Semi-structured interviews were conducted by two investigators, with interview guides developed by a multidisciplinary expert team. Data was mostly obtained via telephone ($n=108$), although some interviews were conducted face-to-face at the participant's request ($n=4$). Each interview began with a request to describe the family's cancer journey, including a subjective description of their diagnostic experience and time to diagnosis. In accordance with the gold standard methods described by Miles and Huberman (1994) and guidelines for rigorous qualitative interviews (Mack *et al*, 2005), the interview guide was adapted iteratively as the study progressed. One key adaptation was to ask 13 families to state, in weeks, their diagnostic intervals. To further refine the data collected, 16 follow-up interviews with participants detailed the perceived impact of delays on coping, relationship with healthcare professionals and medical outcomes (see Supplementary Table 1 for semi-structured interview prompts).

Demographic data was obtained at each interview's conclusion. Medical records of all families were manually searched to determine the documented parent/patient appraisal intervals and diagnostic intervals.

Data analysis. Interviews were recorded and transcribed verbatim. Using an inductive analytical approach, transcripts were coded line by line by one researcher under the close guidance of a senior researcher. Emergent themes were identified and cross-tabulated to allow comparative analyses across the participant groups, assisted by qualitative analysis software QSR NVivo9.2 (QSR International Pty Ltd, Doncaster, VIC, Australia). Correlational analyses to explore the relationship between parent-reported and recorded diagnostic intervals were completed using IBM SPSS V20 (International Business Machines Corp., Armonk, NY, USA).

RESULTS

Participant characteristics. In total, 112 interviews were conducted with cancer survivors ($n=19$; median age at time of interview: 16.1), mothers ($n=44$), fathers ($n=34$) and siblings ($n=15$) from 45 families. Table 1 outlines their demographic information.

Time to diagnosis. Medical records showed that median diagnostic time was 3 weeks (range: 0–28 weeks) (Table 2). Families commonly presented to their general practitioner (46.6%), often with non-specific symptoms (pain: 42.2%; fever: 33.3%; lethargy: 28.9%). Interview data revealed that 18 families (40.0%) subjectively believed they experienced a 'significantly delayed' appraisal interval, because either parents ($n=14$; 31.1%) or patients ($n=4$; 8.9%) failed to report or recognise the importance of symptoms. A total of 21 families (46.7%) subjectively described a prolonged diagnostic interval, when clinicians did not recognise or rapidly refer the patients for definitive diagnostic procedures.

Spearman's rho testing demonstrated a positive correlation between parent-reported diagnostic time and that documented in records ($r_s=0.66$; $P=0.015$).

Themes

Psychological impact. Participants reported significant psychological impacts when symptoms were not recognised or 'taken seriously' by clinicians. Delays in the diagnostic interval were associated with families feeling guilt and regret, either for failing to correctly identify symptoms, rapidly seek assistance or demand appropriate investigations. Family members described anger, frustration and desperation when they were dismissed by doctors.

> *'I'll never get over the fact that my son was riddled with cancer and I was none the wiser, to the extent that it was only by chance that he was [diagnosed]'* (Mother of child, 14 years, rhabdomyosarcoma).
>
> *'I kept going back to the doctors and getting blood tests and blood tests, like every week…they were not doing anything about it'* (Child, 15 years, Hodgkin's Lymphoma).

When a diagnosis was confirmed, several parents were relieved to have a clear management strategy.

> *'The diagnosis [was] obviously horrifying…but also a bit of relief'* (Mother of child, 0 years, neuroblastoma).

Conversely, those who did not experience delays reported feelings of shock, which contributed to an inability to process information or adequately respond to the demands of the diagnosis:

> *'It was crazy…I couldn't believe it…everyone [was] running…[as if with] their heads cut off, not knowing what was going on.'* (Sibling of child, 13 years, leukaemia).

Perceived impact of delays. Some families believed lag-time contributed to poorer medical outcomes, more complicated treatment or increased side-effects.

> *'What's worried me is if we'd left it another day or two would he have survived? I doubt it.'* (Mother of child, 10 years, non-Hodgkin's lymphoma).

Six families described distrust in clinicians following diagnostic delays.

> *'I had no confidence whatsoever…I didn't trust anyone.'* (Mother of child, 12 years, Hodgkin's lymphoma).

Table 1. Demographic information and other characteristics of interview participants ($n=112$)

	Survivors ($n=19$)	Mothers ($n=44$)	Fathers ($n=34$)	Siblings ($n=15$)
Mean age at interview in years (s.d.) Range	16.1 (2.2) 12–20	42.5 (6.7) 29–64	45.9 (8.5) 31–65	21.2 (7.4) 13–38
Mean age at diagnosis of survivor (s.d.) Range	13.3 (2.6) 7–17	38.5 (7.2) 25–60	41.8 (8.6) 28–61	17.4 (7.3) 8–34
Mean interview length in minutes (s.d.) Range	40.2 (14.5) 20–73	70.8 (20.7) 33–126	55.2 (14.5) 24–117	28.6 (6.6) 18–39
Mean time since treatment completion of survivor (s.d.) Range	12.46 (8.3) 1.9–27.7	11.6 (8.7) 1.9–41.0	10.5 (9.0) 0.2–41.0	11.8 (7.5) 4.0–27.7

Table 2. Survivor demographic and diagnostic information ($n = 45$)

Total diagnostic time (weeks)	
Median (range)	3.0 (0.0–28.0)
⩽3.9 (%)	23 (51.1)
4.0–7.9 (%)	11 (24.4)
8.0–23.9 (%)	5 (11.1)
⩾24.0 (%)	5 (11.1)
Data not available (%)	1 (2.2)
Parent/patient appraisal interval (weeks)	
Median (range)	1.0 (0.0–24.0)
⩽3.9 (%)	23 (51.1)
4.0–7.9 (%)	5 (11.1)
8.0–23.9 (%)	4 (8.9)
⩾24.0 (%)	2 (4.4)
Data not available (%)	11 (24.4)
Diagnostic interval (weeks)	
Median (range)	0.7 (0.0–24.0)
⩽3.9 (%)	28 (62.2)
4.0–7.9 (%)	4 (8.9)
8.0–23.9 (%)	1 (2.2)
⩾24.0 (%)	1 (2.2)
Data not available (%)	11 (24.4)
Diagnosis (%)[a]	
ALL	13 (28.9)
AML	4 (8.9)
Neuroblastoma	4 (8.9)
Non-Hodgkin's lymphoma	4 (8.9)
Wilms tumour	4 (8.9)
Ewing sarcoma	3 (6.7)
Germ cell tumour	3 (6.7)
Hodgkin's lymphoma	3 (6.7)
Rhabdomyosarcoma	3 (6.7)
CNS tumour	2 (4.4)
Osteosarcoma	2 (4.4)
Other	2 (4.4)

Abbreviations: ALL = acute lymphoblastic leukaemia; AML = acute myeloid leukaemia; CNS = central nervous system.
[a]Of the 45 patients, 2 had 2 diagnoses; both reported.

'We have stopped seeing [the GP]… this doctor couldn't see [the child's symptoms]…[child was] left suffering and there was a long delay of two or three months…[we ended up] losing confidence' (Father of child, 15 years, osteosarcoma).

DISCUSSION

In paediatric oncology, recent evidence suggests that diagnostic delays may not have significant medical and prognostic implications for all cancer types (Brasme *et al*, 2012, 2014). This study, however, suggests that there remains an important need to minimise diagnostic delay and devise interventions to assist families that are at a risk of delay-associated distress. Failing to address the psychological ramifications of delays may impact families' capacity to adjust to the diagnosis and may have lasting implications for treatment and/or screening adherence (Oeffinger *et al*, 2006).

Education about childhood cancer symptoms is currently being investigated in programs such as the 'HeadSmart' campaign in the United Kingdom (Wilne *et al*, 2013). However, a major challenge is devising interventions to raise public awareness without causing undue distress in the healthy population (Quaife *et al*, 2014). A higher index of suspicion among primary care clinicians may minimise delays and reduce distrust toward the healthcare system, but may also result in over-testing and parental anxiety. It is important to target not only patients and families with balanced education about cancer symptom recognition, but also clinicians, who may benefit from education about the less common cancer warning signs, particularly among less prevalent cancers, such as childhood cancers.

The degree of concurrence between diagnostic times in medical records and interviews suggests that the health care workers in tertiary referral hospitals are aware of and are recording delays. This provides an opportunity to use the information as a 'teachable moment' in their interactions with referring doctors, if appropriate. It also suggests that parents are reasonably accurate in recalling their diagnostic experiences, although analysis with a larger sample is clearly needed to confirm these findings.

Although not unusual for interview studies in paediatric oncology (Shepherd and Woodgate, 2011; Schweitzer *et al*, 2012), the response rate was low at 34%. The sample may have been biased towards participants concerned about delays, affecting the potential external validity of results. It would also be prudent to conduct studies with larger samples, especially of survivors and siblings. Very few studies have addressed the impacts of delays on family members other than mothers. Interviewing fathers, patients and siblings was therefore a strength of this study, although parents were the largest responders and most vocal about delays, which may be a reflection of the smaller sample sizes in the patient and sibling groups. Longitudinal follow-up of families experiencing delays would also be beneficial in analysing any longer-term impacts on the clinician–family relationship.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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