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'Shouting from the roof tops': a qualitative study of how children with leukaemia are diagnosed in primary care

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ABSTRACT

Objectives: To investigate the prehospital presentation of paediatric leukaemia and identify the disease and non-disease related factors which facilitate or impede diagnosis.

Design: Thematic analysis of qualitative semistructured interviews.

Setting: One tertiary referral centre in Southern England.

Participants: 21 parents and 9 general practitioners (GPs) of 18 children (<18-year-old) with a new diagnosis of acute leukaemia.

Results: The majority of children were first seen by GPs before the characteristic signs and symptoms of leukaemia had developed. In their absence, behavioural cues such as the child becoming apathetic or 'not themselves' often triggered parents to seek medical help. Most GPs were unclear about the nature and severity of the child's presentation: then, safety netting, thorough history-taking and examination, and reliance on contextual information about the parents or from prior hospital paediatrics experience were used to manage diagnostic uncertainty. The nature of the doctor–parent relationship helped and hindered the diagnostic pathway. GPs' prior perceptions of parents as being 'sensible' or 'worriers' influenced how gravely they treated parental concerns, with 'worriers' being taken less seriously. Some parents believed GPs failed to listen to their anxieties and discounted their expert knowledge of their child. Specific delay factors included lack of continuity of GP; some GPs' reluctance to take blood from children; and some parents feeling unable to voice effectively their concerns.

Conclusions: The presentation of paediatric leukaemia in primary care differs from that described in many hospital studies, with greater diversity and intermittency of symptoms, and the frequent absence of 'red flags' of serious illness. A wide range of non-disease related factors potentially delay the diagnosis of paediatric leukaemia, including tensions in the doctor–patient relationship and the doctors' cognitive biases. The identification and attempted modification of these factors may minimise diagnostic delay more successfully than raising awareness of 'red flags' of disease.

Strengths and limitations of this study

- This is the first study, to our knowledge, to explore factors in the prehospital diagnosis of a serious childhood illness (paediatric leukaemia) from the perspectives not only of parents but also of general practitioners (GPs).
- It provides an original perspective on the challenges inherent in diagnosing rare illnesses, identifying a wide range of non-disease related factors potentially delaying the diagnosis of paediatric leukaemia, including tensions in the doctor–patient relationship, doctors' cognitive errors and systems factors such as discontinuity of care.
- It also reveals that the presentation of paediatric leukaemia in primary care differs from that described in many hospital studies, with greater diversity and intermittency of symptoms, greater prominence of behavioural changes and the frequent absence of 'red flags' of serious illness.
- The study is potentially limited by the recruitment rate for GPs, with approximately half choosing not to take part, and the fact that data were gathered from a single tertiary centre whose catchment area may not be representative of practice nationally.
- The inclusion of control children without leukaemia could also have strengthened our analysis.

INTRODUCTION

Accurate discrimination between presentations of rare, life-threatening disease and those of common, self-limiting illness is one of the defining challenges of general medicine.[1] Paediatric cancer is perhaps the archetypal disease 'not to miss'. In developed countries, cancer now causes more childhood deaths than any other serious illness, including meningitis, yet a general practitioner (GP) will encounter a child with cancer only once every 20 years, and a quarter of children wait more than 3 months to obtain a diagnosis.[2 3] Failure to spot a childhood malignancy can lead to adverse outcomes including avoidable deaths, while overcautiousness may generate unnecessary investigations and referrals.

Improving the early diagnosis of childhood cancer is a key priority for many health services. The UK's National Health Service Cancer Plan, for example, stipulates that all patients, including children, with 'red flag' features of possible cancer, should be seen by a specialist within 2 weeks of referral by their GP.[4] However, the vast majority of children with cancer are not diagnosed by this pathway, but are identified by other routes, such as direct presentations to emergency departments.[5] Relying on identification of red flag features to prompt suspicion of a serious illness may be flawed if the red flags occur only late in the evolution of an illness, lack sufficiently discriminatory value or are not applied in practice. In addition, for low-prevalence diseases (such as childhood cancer in primary care), even clinical features with high positive likelihood ratios only increase post-test probability by a small amount. Furthermore, the red flag approach may be too simplistic, focusing only on the disease-related determinants of diagnostic delay.[6] The reality of the diagnostic process is a series of interactions over time between clinicians, patients and in paediatrics, their parents/carers. Therefore, a wide range of non-disease related factors such as the nature of the doctor–patient relationship, continuity of care or a doctor's cognitive biases may equally impede the diagnostic process.

We aimed to identify disease and non-disease related factors which facilitate or impede the diagnosis of paediatric cancer, by exploring the period leading to the diagnosis of leukaemia through narrative interviews with parents and GPs of newly diagnosed children. We chose to investigate a diagnosis of leukaemia for a number of reasons. It is the most common malignancy of childhood, with an annual incidence of nearly 4000 cases in the USA and 450 cases in the UK, and is responsible for a third of child cancer deaths.[7] Leukaemia represents a major diagnostic challenge for primary care clinicians as the disease symptomatology is diverse, affects all body systems and mimics common childhood illnesses. Moreover, the time from symptom onset to diagnosis ranges from 1 day to several months, but is typically several weeks, suggesting considerable scope for improvement in speed of diagnosis.[8]

METHODS

Design

We used a qualitative study design so that participants could describe the whole diagnostic process in their own terms, and potentially raise issues not anticipated by the researchers.

Sample

Parents of all children (<18 years) admitted to one tertiary referral centre in Southern England between July 2009 and July 2012 for the treatment of newly-diagnosed leukaemia were eligible for inclusion. GPs who had seen the child during the period from symptom onset to diagnosis became eligible once the parent had agreed to participate. Parents were initially approached in person by the child's consultant or nurse specialist (the clinical team used their discretion to not invite parents for whom they felt the study was inappropriate). If parents agreed to consider participation, a researcher (RTC, a female doctor) met them in person on the paediatric oncology ward to discuss the project and supply a patient information sheet. RTC emphasised that she was not involved in the child's treatment and that the care the child received would not be affected in any way by the decision to participate or not; she also emphasised that the interview would be confidential. During a subsequent telephone call or face-to-face meeting with RTC, a minimum of 48 h later, verbal consent for participation was obtained from parents and a date for the interview was arranged. Written consent was obtained immediately before each interview was recorded. GPs were contacted by letter and a follow-up telephone call to be informed of the study and invited to participate. Those who agreed gave verbal consent by phone and written consent immediately before each interview was recorded. Recruitment was stopped once saturation was reached (ie, new interviews no longer elicited new themes).

Data collection

All interviews were conducted by one researcher RTC. Parent interviews took place in a private room at the hospital or in parents' homes, according to their preference, and GPs were interviewed in their consulting rooms. Interviews, which took place within 3 months of diagnosis, lasted around an hour, ranging in length from 15 min to 2 h. GPs were offered reimbursement for their time, though 7 of the 9 GPs interviewed declined this.

Interviews were semistructured, using a topic guide to which additional questions were added as new themes emerged. The topic guides and prompt lists were informed by issues identified in the literature pertaining to diagnostic delay in general, with an emphasis on childhood cancer. Parent and GP topic guides were non-identical, though the topics overlapped to some degree. Parents were asked to tell the whole story of their child's illness, from when they first noticed something was not right, through to diagnosis. GPs were asked to describe their consultation(s) with the child, their clinical reasoning and actions, and their views of any factors which may have influenced the diagnostic pathway. New topics were added iteratively as they emerged during interviews, for example, parental use of the internet to diagnose leukaemia. Interviews were digitally recorded and transcribed verbatim. The accuracy of interview data was cross-referenced against the presentation as described in the child's electronic primary care record and hospital paper records.

Data analysis

Data were analysed thematically according to the grounded theory approach. Analysis and interviewing

proceeded concurrently, with two researchers (RTC and CHDJ, a non-clinical researcher) independently reviewing all transcripts and identifying categories and themes. Together they developed a systematic coding frame, and RTC used this to assign segments of transcripts to coding categories. The coding frame was edited as analysis and data collection proceeded. Parent and GP interviews were analysed simultaneously using similar coding frames, with the analysis of each informing the other. All authors discussed the most salient themes regularly, starting after the first few interviews, to refine and develop the coding frame. All research team members discussed any differences in interpretation, until consensus was reached.

RESULTS

We interviewed 18 mothers, 3 fathers and 9 GPs of 18 children with leukaemia. Children's characteristics are summarised in table 1. Given the low prevalence of paediatric leukaemia, and hence the increased risk of patients being inadvertently identifiable, we have presented only a limited selection of characteristics to maintain anonymity. Furthermore, in order to preserve patient, parent and doctor confidentiality, we have assigned the patients' individual identification codes (numbered 1–18) which are entirely unrelated to the GP identification codes (letters A–I). All parents approached agreed to participate, and 9 of the 18 eligible GPs agreed. Three parents were not approached because their clinical team did not feel it was appropriate. When parental and GP verbal accounts were cross-referenced against hospital and GP records, no major discrepancies were found (but parent accounts were much more detailed). We present the results under categories reflecting the main steps in the diagnostic pathway, namely presentation of leukaemia, parents' interpretation of symptoms, doctors' appraisal of children and parent–doctor interactions. The main subthemes to arise during analysis are presented within these categories. Figure 1 summarises all those subthemes to emerge as factors potentially impeding the diagnosis of paediatric leukaemia.

Presentation of leukaemia

The presenting features of leukaemia described by parents were diverse, intermittent and non-specific, covering a broad range of behavioural and physical changes (table 2). Typically, the symptomatology evolved over weeks to months, with non-specific early features such as fever, pallor and fatigue mimicking those of common, self-limiting, minor illnesses. More specific features, such as a non-blanching rash, usually appeared later in the illness trajectory, though in a small minority of children they heralded its onset. Altered behaviour was often parents' first cue that something was seriously wrong:

> On Tuesday last week he kept crying for a scooter and I said okay, let's go to Toys R Us, I'll buy a scooter for you… We came home, I cobbled it together, he said he wasn't playing and I [asked], 'what's wrong?' He said, 'I don't want to play.' I touched his body. I noticed it was

Table 1 Characteristics of participating children with leukaemia

Age range (years)	Child identification number	Sex	Leukaemia type*	Symptom interval (period from symptom onset to diagnosis)	Number of GP appointments	Number of other medical contacts prior to diagnosis
0–2	1	Female	ALL	5 months	7	
	15	Female	AML	6 months	2	
	17	Female	ALL	1 week	1	1 (Minor injuries unit self-referral)
3–6	2	Male	ALL	2 months	2	1 (A&E self-referral)
	3	Female	ALL	5 days	1	1 (Practice nurse)
	4	Male	ALL	3 months	7	1 (A&E self-referral)
	5	Female	ALL	1 week	2	
	8	Female	ALL	3 months	4	2 (A&E self-referrals)
	10	Male	ALL	2 months	3	3 (A&E self-referrals)
	12	Male	ALL	5 days	1	
	13	Female	ALL	3 months	4	
	16	Male	ALL	1 week	1	1 (Practice nurse)
7–10	6	Male	ALL	2.5 months	2	
	9	Male	ALL	3.5 months	1	
11–17	7	Female	ALL	3 weeks	1	
	11	Male	AML	3 months	1	
	14	Male	AML	3 weeks	1	
	18	Male	AML	1 months	2	

*ALL, acute lymphoblastic leukaemia; AML, acute myeloid leukaemia GP, general practitioner.

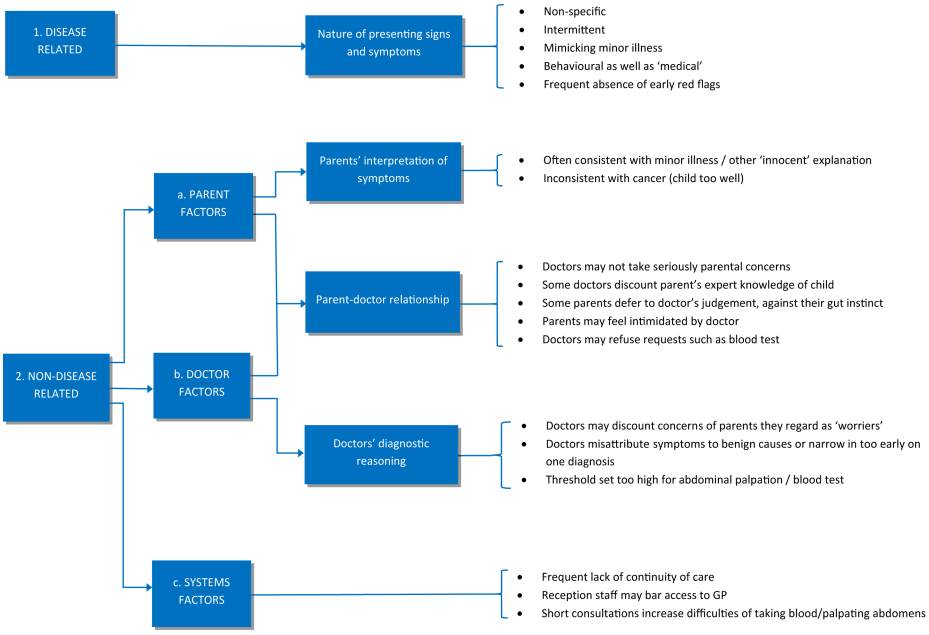

Figure 1 Classification of factors potentially delaying the diagnosis of paediatric leukaemia.

> a little hot and I said, 'now this is very unusual… No matter how sick he was, [he] would get up and play with a new toy. (mother of child 2)

The emergence of unusual and alarming signs such as a 'meningitis-type rash' (mother of child 5) was infrequent.

Parents' interpretations of symptoms

Most parents initially attributed their child's symptoms to minor, viral illnesses or to other 'innocent' explanations such as ascribing joint pains to sporting activities, refusal to walk to attention-seeking and bleeding gums to poor dental hygiene:

> He's a 12 year old boy who plays a lot of sport, and he's probably going to get growing pains. And, OK, the bruises, well, he's playing rugby, what do you expect? (mother of child 11)

Some parents believed initially that since their child was behaving normally, their physical symptoms must not be serious. Most parents had minimal or no prior knowledge of leukaemia, but imagined it to be incompatible with normal behaviour. Altered behaviour (as above) often triggered parents' first serious concerns (and consultations), as did the persistence or worsening of initially innocuous physical symptoms, such as fevers or bruises.

Doctors' assessment of children

GPs, like parents, commonly considered initial presentations to be consistent with mild, self-limiting illnesses such as viral infections, a pulled muscle or growing pains. A smaller number of doctors were certain that the child before them was severely unwell, given their physical appearance and behaviour, and the gut instincts those provoked:

> This was one of those ones where I opened the door and thought, 'I ought to telephone, to get the hospital on the line, yeah.'… I just remember that lurch of your stomach, you know, when you think 'oh'… She wasn't bouncy, she was very quiet, she seemed to be in pain actually, she was moaning, and she looked slightly swollen, her face was rounder than it should have been. (GP I)

The absence of a specific diagnosis in these cases did not deter GPs from making an urgent hospital referral. Indeed, some explicitly identified their most important role as being that of discriminating between seriously unwell and essentially healthy children, rather than making a specific diagnosis of leukaemia or any other illness:

> I didn't have differential… No, my differential diagnosis was at that point not important... Leave that to the hospital. He needed to go in there and then, whatever the diagnosis turned out to be. (GP D)

A few GPs suspected leukaemia, but only one was certain of this diagnosis, based on the patients' red flags of chronic fevers, pallor and unexplained bruising.

When GPs were unsure about the nature and severity of the child's illness, they deployed a range of strategies to manage their uncertainty. Some gave explicit 'safety netting' advice, about when to return for further consultation, for instance, should the child's symptoms not resolve. Others sought to eliminate the most serious,

Table 2 Prehospital signs and symptoms in children presenting with leukaemia, described by parents

Physical	Behavioural
Infective	Abnormally quiet
Chattering teeth	Bad moods
Clamminess	Below par
Cold	Clingy
Conjunctivitis	Disinterested in normal activities
Cough	Excessive sleeping
Ear infection	Falling asleep at school/public places
Fever	Floppy
Recurrent infections	Grumpy
Sore throat	Impatient
Sweatiness	Irritable
Uncontrollable shivering	Just not right
	Listless
Cutaneous/mucosal	No enthusiasm for toys
Bleeding gums	Not wanting to play
Bruising	Reluctant to do sport
Dark shadows under eyes	Tired all the time
Pallor	Too well-behaved
Rash	Unresponsive
Spongy gums	Whingy
Yellow appearance	
Musculoskeletal	
Abnormally stiff gait	
Arm/leg pain	
Back pain	
Foot pain	
Joint pains	
Limp	
Refusal to walk	
Swollen joints	
Gastrointestinal	
Abdominal pain	
Diarrhoea	
Reduced appetite	
Vomiting	
Weight loss	
Miscellaneous	
Cold hands	
Collapse	
Delirium	
Dizziness	
Fatigue	
Heavy periods	
Palpitations	
Shortness of breath	
Wheeze	

albeit uncommon, potential causes of the child's presentation:

> He did look pale and tired and unwell, but as you know there are many reasons for that, but his mum was very keen on getting a blood test… I do remember thinking, yes, leukaemia is the reason I want to do a blood test, because that's the one thing I wanted to exclude. (GP F)

Some GPs drew on contextual information such as their prior perceptions of parents as 'sensible' or 'worriers', though several GPs acknowledged this strategy could backfire if the legitimate concerns of 'worriers' were not taken seriously. Attempts to refine the differential on the basis of children's social and cultural context could also have a negative impact, as in the case of a girl of ethnic minority origin, who presented with recurrent bone pain, fevers and weight loss. When a blood test showed a microcytic anaemia, her GP prescribed supplements for iron and vitamin D deficiency, presuming that, like other girls of her ethnicity, she had a restricted diet and limited exposure to sunlight. He later reflected that the cultural context: 'may have produced a fog in our brains…. I'd gone off on this vitamin D deficiency and malnutrition concept in my head, and wasn't breaking out of that, but the picture of the presentation just wasn't fitting' (GP E).

When one child's presentation and recent consultation record were difficult to interpret clearly, the GP managed uncertainty by asking the parent to re-tell the whole story from the beginning, leading to the suspicion of leukaemia:

> If somebody tells me something and it just doesn't match what's on the computer, then I would prefer to make my own judgement and so I prefer to hear the patient's story from themselves. Sometimes… if they go back to the beginning they'll remember something that suddenly is a bit more meaningful that they didn't think of two weeks ago to tell you. (GP A)

GPs commonly cited the importance of training and prior experience in shaping their differential. Some considered duration of clinical experience per se to be key, while others regarded specific experience of hospital paediatrics, and particularly managing acutely unwell inpatients, as being more important, particularly in developing a gut feeling or intuition for spotting unwell children.

Some parents expressed surprise that the GP did not physically examine their child, as in child 10, who presented with severe abdominal pain the morning after a vomiting bout:

> The cold went away but the pulled muscle of the tummy ache didn't go away. So we went to the GP who didn't really examine him, I have to say, just sort of looked at him and said, 'he's got a pulled muscle, it will go away. If it doesn't go away, come and see us in a week. (mother of child 10)

Some GPs also acknowledged reluctance to palpate young children's abdomens, for a number of reasons including the difficulties of so doing within a 10 min consultation.

Parent–doctor interaction

Most doctors explicitly assessed the parent and their child during consultations, with the degree of parental concern influencing some GPs' own level of anxiety. Several GPs, particularly those with additional training in paediatrics, underlined the importance of a parent stating that their child was 'not right':

> As paediatricians you know from the word go, it's drummed into you, that if a parent says, 'she's not right', if you're thinking of sending a child home and she's still saying 'they're not right', you admit them. You always listen because the parent's intuition is right and you've got to find out why they're feeling that way. (GP A)

Changes in parents' typical consultation behaviours, for example presenting with greater frequency, were also perceived as concerning. One GP arranged an unscheduled, emergency appointment after the child's mother uncharacteristically approached her in a public setting: 'my alarm bells were completely ringing…she'd never normally do that, she'd normally leave a message or something, so that was unusual and made me think that she must be very worried' (GP I).

GPs placed greater importance on parental concerns in parents they perceived to be 'sensible' compared with those they judged as 'neurotic' or 'worriers' (see above). For example, one mother explicitly told her GP that she believed her child had leukaemia after searching for information about this disease herself, and consequently asked for a blood test. The GP advised against doing so, in part since the child seemed well on examination, but also because the mother had been excessively anxious in the past about minor symptoms:

> Maybe my brake on doing a blood test was that I felt she was worried unnecessarily, because I'd seen her quite a lot, lots of phone calls about little things like a rash and a dot and a scratchy area and lots of things that, you know, always seemingly caused a lot of alarm, that weren't a problem… It is a case of… the seemingly sensible Mum versus the Mum who is, maybe you should or you shouldn't stereotype, just being particularly anxious, and yes, it does make a difference as to what they're telling you and the emphasis on it (GP G).

Just as parental concern influenced GP concern, so too were some parents guided by their GP's degree of anxiety:

> I'm so naïve. If a GP is saying to me 'she's well in herself, I really don't think it's serious,' then… in the future the first thing that I think to myself is, 'the GP was fairly confident that it was nothing to worry about, so I'm not going to worry about it. (mother of child 15)

Indeed, several parents described using their GP's innocent interpretation of their child's symptoms to allay their deeper, unvoiced instincts that something more serious was wrong:

> You don't want your child to be sick, terribly ill, you just want someone to say 'it's fine, it's alright' so we kind of clung onto that, I suppose (mother of child 10).

Some parents believed their GP dismissed, rather than listened to their concerns, yet felt unable to voice this. Over a 4-month period, one young girl had multiple GP contacts, for recurrent infections, fever, weight loss and pallor, while her mother became increasingly distressed: "I always feel very intimidated by male doctors in particular because I always feel that, you know, I haven't got half a brain that they've got… [I was] made to feel as though, yes, you might have given birth to this thing, but you don't know anything about this thing… I almost wanted to scream at him, 'look there is something wrong with my child!. (mother of child 1)

Several GPs acknowledged that they may have failed to heed parents' concerns sufficiently:

> She did feel like she was shouting from the roof tops that there was something not right and no one was listening… Seemingly she was shouting but maybe not saying the words that we could hear (GP G).

On feeling that their GP failed to take their concerns seriously, some parents circumvented the GP's traditional gatekeeper role to hospital services, for instance by calling the emergency services or taking their child to an emergency department. Other parents played a direct role in obtaining their child's diagnosis by researching their symptoms on the internet and diagnosing leukaemia themselves. Several consequently asked their GP directly for a blood test, as described above, with some GPs agreeing and others disagreeing to do so. Time constraints during the 10 min primary care consultations were highlighted as a barrier to taking blood from a young child, as were fear of traumatising children, lack of experience and a general ethos in general practice of not taking blood from children:

> There is very much that statement, "oh, we don't want to put him through it", and I've adopted that statement, and I never used to say it because of having done paediatrics. I've taken so much blood from so many little ones, and you can do it in a nice way and… with a bit of skill make it pretty much a non-event, and so I didn't actually have that spin on it too much, I've adopted this from my general practice training (GP G).

Lack of continuity of GP between consultations was cited by parents and GPs alike as contributing to diagnostic delay. Several parents also identified the problem of reception staff denying access to doctors, such as one father who was forced to describe the crippling nature of his daughter's pain 'to a receptionist, because you can't speak to the GP unless you have an appointment with the GP. I said again, 'look this is the problem, talk to the GP' and they wouldn't even transfer me to the GP' (father of child 8).

DISCUSSION

Main findings

A wide range of interacting disease and non-disease related factors affected the speed and accuracy of diagnosis of leukaemia in children prior to hospital admission. The main themes to emerge from this study are first that the majority of children were seen initially by GPs before the characteristic clinical features of leukaemia had developed. In their absence, behavioural cues, such as the child becoming apathetic or 'clingy,' featured prominently in triggering parents to seek help. When a child did present as acutely unwell, GPs recognised this swiftly, drawing primarily on their physical appearance and behaviour, and the gut feelings these provoked. A second major theme, applicable both to parents and GPs, was the misattribution of presenting signs and symptoms to common, self-limiting illnesses or other 'innocent' explanations. In part, this reflects the non-specific and diverse nature of the early presenting features. The third main theme relates to what GPs do when unsure what is wrong with the child. Strategies to manage diagnostic uncertainty included 'going right back to the beginning' when the clinical course is vague; having a good safety netting system in place; seeking to eliminate the most serious potential causes of the presentation; and drawing on their prior contextual knowledge of the parent and their prior experience and training in paediatrics. Fourth, the doctor–patient relationship had a significant impact on helping and hindering the process of obtaining a diagnosis. For example, sometimes GPs' concerns, and hence actions, were shaped by how anxious they judged the parent to be, yet some parents also found the doctor's level of anxiety assuaged or exacerbated their own. A GP's prior view of a parent as being 'sensible' or a 'worrier' could influence how gravely they treated their concerns, with 'worriers' being taken less seriously. While some GPs stressed the importance of listening to parents, many parents believed GPs failed to take their anxieties seriously, with these concerns not always being voiced. A fifth major theme was the influence of system factors on the diagnostic pathway. For example, both parents and GPs identified lack of continuity of care between GP visits as being detrimental. Some parents also raised the problem of non-medically trained reception staff determining access to urgent appointments. Finally, the constraints of short appointments were raised by some GPs as potentially discouraging them from two diagnostically useful activities: taking blood and palpating abdomens.

We have classified some of the key findings above in terms of whether they are disease or non-disease related factors impeding diagnosis (figure 1). We believe this is a valuable conceptual framework for thinking about rare illnesses since it highlights the full range of factors potentially influencing speed to diagnosis, rather than the presenting features of the illness alone.

Strengths and weaknesses

This is the first study to explore factors in the prehospital diagnosis of paediatric leukaemia from the perspectives not only of parents but also of GPs. In-depth narrative interviews are ideally suited to investigating a behavioural process such as diagnosis, since they allow participants to articulate fully their own account of events, enabling a rich understanding of why individuals behaved as they did at each stage. Being drawn from a tertiary referral centre covering a large geographical area, our sample was diverse and included fathers as well as mothers, parents from various socioeconomic and ethnic backgrounds, and GPs with various levels of experience.

The study had several limitations. First, we recruited a limited number of GPs, with approximately half choosing not to take part. We were not able to ascertain specific reasons for this, but some GPs may have been dissuaded from participating by fears of judgement or medical litigation mentioned in the consent forms they had to sign, or due to time constraints. Second, there was an unavoidable risk of recall bias, but this was minimised by interviewing participants as soon as possible after diagnosis. Finally, the inclusion of control children without leukaemia could have strengthened our analysis.

Comparison with existing literature

Most descriptions of how childhood leukaemia presents are based on small cohorts of hospital patients, describing a relatively short and defined list of signs such as pallor, fever and unusual bruising.[9–12] In contrast, our study highlights the importance of wide-ranging behavioural changes and clinical features in heralding illness onset and triggering consultations in primary care. Previous qualitative studies of childhood meningococcal disease have analysed doctors' diagnostic reasoning, and several qualitative studies have explored parents' experiences of a cancer diagnosis in their child.[13 14] No studies, to our knowledge, have sought to combine simultaneously the analysis of parental and clinician perspectives. The small but growing literature on diagnostic error tends to view 'doctor delay' exclusively from the doctor's perspective, with the implicit assumption that it is predominantly what the doctor does or does not do that shapes this component of the symptom interval.[15 16] However, our data demonstrate the proactive role many parents play in obtaining their child's diagnosis, for example, through refusing to accept their doctor's decisions, bypassing the GP's role as a gatekeeper to secondary care, or asking for a blood test. Our findings demonstrate clearly how doctors' cognitive errors can potentially delay diagnosis in primary care. For example, some GPs decided early in the illness process that they knew the underlying cause of the child's symptoms (a minor illness) and therefore did not actively seek alternative diagnoses—so-called 'premature closure'.[16]

Implications for practice and research

'Red flag' based algorithms for aiding diagnosis of paediatric cancers such as leukaemia may be of limited value, given its frequently non-specific and fluctuating early features. Our results concur with a recent case control study of primary care records in the UK, which found that the alert signs identified in National Institute of Health and Care Excellence guidance for childhood cancer were recorded more commonly in cases than controls, yet that only a quarter of the cases had any alert symptoms recorded in the 3 months prior to diagnosis.[4 6] Clinicians should also be aware that non-disease related factors are potentially drivers of diagnostic delay. Specific practical implications might include GPs having a lower threshold for taking blood in children, examining young children's abdomens more readily and introducing a system whereby any children presenting more frequently than usual, trigger more detailed scrutiny by clinicians. More generally, current UK GP training, in which only 40% of trainees experience a hospital paediatrics placement, should be reformed so that all GP trainees have core paediatric training.[17] On-going professional development should build skills and confidence in examining and taking blood from young children.

While the importance of good doctor–patient communication is nothing new, our findings underscore its unique significance for diagnosing rare childhood illnesses in general practice. 'Listening to the parent' is something of a mantra in paediatrics; our findings highlight in addition the importance of what is unsaid between parent and clinician in shaping the diagnostic pathway. First, some doctors and parents silently take their cue from each other as to how anxious they should be themselves, with the concomitant risk of false reassurance. Second, some parents feel their concerns are neither listened to, nor acted upon by GPs, yet do not voice this overtly. Conversely, some GPs believe they are good listeners, yet do not explicitly check this with the parent. If parents can be 'shouting from the rooftops', yet using words which GPs do not hear, then perhaps the most pressing concern is using GP training, revalidation and continuing professional development to enhance the three way communication skills between doctor, parent and child.

Future research in this area should include the accounts of older children themselves, in addition to those of parents and doctors. More work is also needed to clarify the diagnostic processes of GPs who correctly suspected leukaemia at an early stage, to see if these may be disseminated and incorporated into training. Finally, our study highlights the scant evidence for how most serious illnesses present in primary care.

Conclusion

Leukaemia is the most common malignancy in children, but a rare occurrence in primary care. Its diagnosis requires information to be communicated to a clinician by a patient and/or parent in a way that enables the clinician to interpret the information correctly, recognise that the child may have cancer and consider the appropriate examination and investigations. This study identifies a wide range of non-disease related factors potentially impeding this diagnostic process. Their attempted modification may minimise diagnostic delay more successfully than raising awareness of red flags of leukaemia, and this approach could be extrapolated to the other rare diseases of childhood.

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