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The Malta Journal of Health Sciences is a peer-reviewed, open access publication that promotes the sharing and exchange of knowledge in Health Sciences. It provides a platform for novice and established researchers to share their findings, insights and views within an inter-professional context. The Journal originates within the Faculty of Health Sciences, University of Malta.

The Malta Journal of Health Sciences disseminates research on a broad range of allied health disciplines. It publishes original research papers, review articles, short communications, commentaries, letters to the editor and book reviews. The readership of the journal consists of academics, practitioners and trainee health professionals across the disciplines of Applied Biomedical Science, Audiology, Communication Therapy, Community Nursing, Environmental Health, Food Science, Health Services Management, Medical Physics, Mental Health, Midwifery, Nursing, Occupational Therapy, Physiotherapy, Podiatry and Radiography.

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(v) study has serious flaws in the Methods section;
(vi) authors did not obtain ethics approval from a properly constituted ethics committee, where appropriate.

Manuscripts that qualify for review are evaluated by at least two experts as appropriate. A double-blind review process is adopted.

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Authors are expected to meet all three criteria. Contributors who do not qualify under the authorship criteria should be included in the Acknowledgments section.

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If the article was not supported by funding, authors should include the statement below:

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Ethical approval

Ethical approval from a research ethics committee and/or institutional review board is usually required and details regarding the board providing approval should be listed in the Methods section of the manuscript.

Publishing policy

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Guest editorial

THE SIR ANTHONY MAMO ONCOLOGY CENTRE

Stefan Laspina
Consultant, Transfusion Medicine and Clinical Chairperson, Department of Haematology and Oncology,
Mater Dei Hospital / Sir Anthony Mamo Oncology Centre, B'Kara, Malta

In 2009, the development of a medical brief, effectively detailing the specifications for a purpose-built oncology hospital and including the medical equipment and human resources required, was commenced. Robust engagement by the relevant stakeholders, many of which hailed from the extant Sir Paul Boffa Hospital, ensured a very relevant proposal. The project (ERDF 196), led by the Foundation for Medical Sciences, was subsequently approved for partial funding through European Regional Development Funds. The new Sir Anthony Mamo Oncology Centre, as it came to be named, first opened its doors for service in December 2014 when the Out-patients Department received the first oncology patients. In April 2015, this was extended to include haematology and paediatric oncology patients. Full migration of services, including in-patient care, took place in September 2015.

The distribution of services within the new Centre includes five clinical areas for in-patients made up of two oncology wards, one radioisotope unit, one haematology ward and one palliative care ward, with a total of 88 beds, an out-patient unit with 12 clinic rooms, a day area for day-treatment with a total of 21 couches and eight beds, a clinical support services unit and a radiotherapy department.

The staff complement, a significant number of which are Health Sciences graduates, is very diverse. Specific training related to the care of cancer patients is currently ongoing and opportunities for continued professional development for all staff are being prioritised.

Special mention must be made of the dual-qualified radiographers who graduated through a course run jointly between the University of Malta and the University of Cardiff who were deployed within the Centre's radiotherapy unit, and the medical physicists in radiotherapy, whose training, partially funded through ESF 4.175, occurred under the joint auspices of the University of Malta and Leeds Teaching Hospitals NHS Trust.

The very scope behind this large project is to ensure that the level of cancer patient management in Malta continues to improve, if possible on a par with that of countries with similar health systems. Recently published research has shown Malta to be somewhat middle-ranked on overall survivorship, demonstrating an age and case-mix standardised five-year relative survival of 51.3% for all cancers, only just below the average quoted for Europe (52.5%) (Baili et al., 2015).

To this effect, a number of developments are planned within the radiotherapy unit that will result in a decrease in patient treatment complications, and will allow for the treatment of individuals who hitherto were sent abroad, thus providing for a better patient experience. Within the next three years, plans are in place to develop Intensity-Modulated Radiation Therapy (IMRT) and Volumetric Modulated Arc Therapy (VMAT), optimising treatment of relevant cancer sites through the provision of high dose conformance to the tumour, thereby reducing risk to critical structures. A reduction in treatment time, which potentially decreases the likelihood of patient movement during the therapy, will increase accuracy which, together with on-board on-line imaging, will significantly enhance treatment precision.

Though in the past, a few Maltese patients were enrolled in clinical trials, plans for a significant drive to put this process on a more formal and universal footing through the set-up of a clinical trials unit within the Centre, are being addressed. The aim is to significantly improve access for Maltese patients to drugs-in-development and to allow for more significant participation in leading edge clinical trials. In conjunction with this, a well-resourced framework needs to be set up to facilitate the timely adoption of new pharmaceuticals, including the use of immunological therapies, especially in the context of personalised cancer management.

The local performance of autologous bone marrow transplants in the long term is also being given due consideration. However, this depends heavily on further staff training, recruitment of additional expertise, and on other infrastructural projects, mainly the Innovation Centre for Excellence – Blood, Tissues and Cells (ICE-BTC), approval of which is currently pending a specific ERDF call for applications.

In order to truly ascertain that the patient is at the very core of this enterprise, the Sir Anthony Mamo Oncology Centre should attempt to achieve accreditation. Similar hospitals in Europe have undergone this laborious yet very constructive quality measure and have been certified by institutions such as Joint Commission International. This will undoubtedly have a profound impact on the operational performance of the hospital, ensuring that all processes within the hospital are optimally geared towards the patient.

Of course, the Sir Anthony Mamo Oncology Centre will not be operating in a vacuum. Significant co-operative ventures are ongoing, whereby expertise is being transferred and co-operation is being sought. A bilateral arrangement with Leeds Teaching Hospitals NHS Trust has contributed significantly to the developments in radiotherapy, both in terms of equipment validation and of planned evolution in treatment methodology. Malta is also actively participating in the European Reference Networks project, an EU Commission initiative whereby highly specialised healthcare providers are designated as centres of expertise or reference. This in turn enables the concentration of expertise and patient numbers in one place to optimally manage rare or complex diseases including many cancers. On a more local level, the existing synergies with Mater Dei Hospital should be strengthened and new ones built with the University of Malta and possibly the Life Sciences Centre, exploiting the adjacency of these institutions. There should also be additional focus on strengthening the existing relationships with non-governmental organisations since these all bring a particular ethos of their own that touches patients in different ways.

All this should be complemented by a continued transfer of care from an in-patient/hospital environment to a community-based one that will allow patients to enjoy the comforts of their personal surroundings and families for a larger part of their treatment. Stronger co-operation with community-based care providers is extremely important, as is the investment in robust information systems that would effectively underlie all the highlighted developments.

The Sir Anthony Mamo Oncology Centre will therefore, in the coming years, effectively serve as the backdrop for significant advancements which will hopefully alter for the better the outcome of the battle against cancer.

Reference

Baili, P., Di Salvo, F., Marcos-Gragera, Sieslin, S., Mallone, S., Santaquilani, M., Micheli, A., Lillini, R., Francisci, S. & the EUROCARE-5 Working Group (2015) *European Journal of Cancer*, 51(15), pp. 2120 – 2129.

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Research paper

THE DEVELOPMENT OF EARLY EXPRESSIVE VOCABULARY IN CHILDREN WITH DOWN SYNDROME

Christina Coppini, Daniela Gatt
Department of Communication Therapy, Faculty of Health Sciences, University of Malta, Msida, Malta

Abstract. A delay in expressive language in children with Down Syndrome (DS) is common, and often a major challenge of the condition. This study aimed to investigate the early expressive vocabulary skills of Maltese children with DS, whose first languages were either Maltese or English, while taking into account chronological age. Language preference was further explored in the context of a bilingual environment. A multi-method design was implemented across seven participants whose language abilities ranged from the expression of single words in isolation to simple word combinations. The expressive vocabularies of four boys and three girls between 2;10 and 11;9 years were assessed through caregiver report, picture naming and language sampling. Performance of the children was analysed in relation to local findings on lexical production in typically-developing children. The study revealed that productive vocabularies of Maltese bilingual children with DS escalated with increasing age, notwithstanding inevitable individual variation.

Keywords: lexical development, Down Syndrome, bilingual, expressive vocabulary

1 Introduction

Down Syndrome (DS) is a chromosomal disorder caused by a third copy of chromosome 21 (Grant et al., 2010). The disorder is typically associated with physical and cognitive deficits, which may affect speech, language and communication at large (Kumin, 2003). Characteristics that impact communication include macroglossia, oro-motor hyper- or hyposensitivity, and intellectual disability (Kumin, 2003). Hearing loss as a result of recurring ear infections is common, which may delay the processing of complex auditory stimuli (Rondal, 2009). Moreover, impaired auditory-vocal short-term memory is known to account for limitations in lexical learning among children with DS (Jarrold, Nadel & Vicari, 2007).

Lexical, or vocabulary, acquisition makes up an integral part of language learning, and is ultimately a prerequisite for the development of other language domains (Gatt, Grech & Dodd, 2013). Rondal (2009) identified a delayed onset of babbling by two to three months in infants with DS, with the production of single words tending to emerge between two and three years. Hence, a significant delay is evident when considering the onset of expressive language in typical development (Galeote et al., 2008). Children with DS present with limited pre-linguistic skills, namely eye contact, joint attention and functional playing skills (Kumin, 2003), which may contribute to their delayed lexical acquisition. Moreover, Feltmate and Kay-Raining Bird (2008) found receptive language skills to be a strength in language development, while expressive language was delayed.

Structural differences in the input languages received are expected to affect vocabulary development in typically-developing (TD) children exposed to varying language contexts. For example, in their study on TD Italian- and English-speaking children aged 0;8 to 1;4 years, Caselli et al. (1995) found no discrepancy in the onset and development of major grammatical categories, including nouns and verbs, but identified a slower rate of overall vocabulary growth in Italian children.

Differences in language development related to linguistic input received are expected across children with DS. In the context of bilingual input, Rondal and Buckley (2003) hold that the language pairs to which children with DS are exposed may also impel variation in lexical development. Likewise, differences in rate of lexical acquisition in TD children have been attributed to the language pair being learnt (Thordardottir et al., 2006). Importantly, Rondal (2009) claims that children with DS are capable of exhibiting features of bilingual competence. Feltmate and Kay-Raining Bird (2008) found first language (L1) proficiency to be similar in monolingual and bilingual children with DS, since both groups presented with equivalent receptive skills and expressive language delays. This indicates that bilingual exposure should not have a detrimental effect on language development in children with DS.

Children with DS growing up in Malta are exposed to societal bilingualism. The functional use of two linguistic codes occurs at a societal level, since both Maltese and English are official languages (Gatt, Letts & Klee, 2008).

The Down Syndrome Association Malta (2009) reported an average of 12 births with DS per year in Malta. Norms for typical lexical development have not yet been established for Maltese children, although developmental trends for expressive lexical acquisition have been investigated (Gatt et al., 2013), providing reference measures that allow more objective analysis of expressive lexical skills identified in Maltese children with DS.

This study is driven by the following research questions:

- How do Maltese children with DS perform on measures of expressive vocabulary?
- How does chronological age affect expressive vocabulary size in Maltese children with DS?
- What proportions of Maltese and English words are employed in these children's expressive vocabularies?
- To what extent do grammatical categories (content and function words) feature in their expressive vocabularies?

Correspondence to Christina Coppini
(christinacoppini@gmail.com)
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2 Methods

2.1 Participants

Seven Maltese children with DS participated in the study. Two boys and one girl were English-dominant, while two boys and two girls were Maltese-dominant. The selection criteria were a diagnosis of DS, the linguistic level of each child[1] and primarily Maltese or English exposure. Table 1 lists the salient characteristics of the participants, who were identified and approached via their speech-language pathologists (SLPs). Permission to collect data was obtained from the Primary Health Care, Speech-Language and Education Departments in Malta prior to assessment. Ethical approval was obtained from the University of Malta Research Ethics Committee (reference number 029/2013).

Table 1. Participant characteristics depicted by gender, age, first language (L1) and venue of testing for each child (C)

C	Gender	Age (years; months)	L1	Venue
1	Male	2;10	English	District clinic
2	Male	4;4	Maltese	District clinic
3	Female	4;5	Maltese	District clinic
4	Male	4;5	English	District clinic
5	Male	5;4	Maltese	State school
6	Female	8;5	Maltese	District clinic
7	Female	11;9	English	District clinic

2.2 Research design and procedure

A methodological design comprising three methods for measuring lexical expression, namely parental report, picture naming and language sampling, was employed. A triangulation of methods was preferred to validate vocabulary data and avoid methodological bias (Bogdan & Biklen, 2006). Structured interviews targeting parents or guardians were also used to obtain background information on each child's general and language development. The duration of each session was approximately 15 to 20 minutes.

2.2.1 Parental report

Hoff (2012) found parent-based instruments to be widely used in the assessment of children's emerging vocabulary. Gatt et al. (2013) also claimed that parent-based information facilitates the process of identification of early vocabulary delays. A parent-based measure of the participants' expressive vocabulary skills was obtained through the use of a vocabulary checklist (VC), which is described in detail in the next section.

2.2.1.1 Vocabulary checklist

A detailed overview of each child's expressive vocabulary was obtained by using an adaptation of the VC of the first edition of the MacArthur Communicative Development Inventory: Words and Sentences (CDI: WS) (Fenson et al., 1993) for Maltese children, as formulated by Gatt (2010).

The adaptation included both Maltese and English lexical items across 24 semantic categories, as well as words that are not considered Maltese- or English-specific, such as onomatopoeic sounds and across-language homophones (e.g. 'blue' and '*blu*'). These words are referred to as Generic words in this study. The VC was given to the primary caregiver of each participant. Caregivers were expected to recognise and mark the lexical items produced spontaneously by the participants, while words not provided in the checklist were to be added in the recall section, as specified in the VC, following each semantic category.

The VC score, representing the total number of reported words, was broken down into smaller component scores. The first component score consisted of a differentiated sum of recognised and recalled words spontaneously produced by the child. The second was based on language classification of words as Maltese, English and Generic. A percentage of language classification scores across participants was calculated. Content words[2] and function words[3] were then identified with reference to a classification system formulated by Gatt (2010).

2.2.2 Structured assessment

Gatt, Grech & Dodd (2014) hold that informal structured assessment tools are ideal alternatives to standardised tests in contexts where norms of early language development are not available. A structured, informal picture naming task (PNT) formulated by Gatt (2010) provided supplementary information on the children's vocabulary skills via direct assessment.

The PNT consisted of a booklet containing 18 coloured graphical representations of everyday objects, namely a ball, car, cat, baby, pair of shoes, dog, doll, aeroplane, telephone, glass, bicycle, egg, guitar, bird, spoon, hat, flower and comb. The picture items were revealed to the children by their caregiver or SLP, to avoid risks of performance anxiety due to unfamiliarity with the researcher. Their responses were recorded orthographically on a score sheet and also phonetically if deemed necessary by the researcher, namely when responses lacked intelligibility. An audio recording was obtained to support manual transcription and to ensure accuracy. Following analysis, a raw score of the number of items labelled appropriately and independently was computed. Percentages of Maltese, English and Generic words were then calculated across participants.

2.2.3 Language sampling

Language sampling allows deeper analysis of language use in unrestricted contexts (Shipley & McAfee, 2004). Moreover, naturalistic sampling is known to provide a measure of the child's "expressive potential" (Gatt et al., 2014). In the current study, language samples (LS) were obtained during a 10-minute play situation using a standard set of toys comprising a set of farm animals, namely a horse, pig, cow, sheep, donkey and goat, two bales of hay and a gate.

The LS was audio-recorded and transcribed orthographically, post-session, to determine the spontaneous production of lexical items in relation to the toys provided. Words produced on imitation were not considered. The utterances of each child were split into single words and tabulated in alphabetical order. In this way, the researcher was able to calculate the total number of words spoken on different occasions within each sample, to determine the token count. Words expressed more than once by the participant were grouped to calculate the number of different words used (types) by the child. Based on type counts, the proportions of Maltese, English and Generic word types were then calculated.

1 Each participant was able to produce at least single words, with simple word combinations being the upper limit considered.

2 Words that refer to particular objects, attributes or actions such as nouns, adjectives or verbs.

3 Words that represent grammatical relationships between words and contribute to sentence structure, such as pronouns, prepositions and conjunctions.

2.2.4 Structured interviews

Besides the methods and instruments described above, intended to provide measures of the participants' expressive vocabularies, structured interviews based on a background questionnaire for bilingual children (BQ) were intended to provide information on the participants' general and language development. Frattali (1998) acknowledges the significance of using adult informants in measuring child development and disability. A structured face-to-face interview using the BQ was therefore administered to the primary caregiver of each child to gain insight on participants' developmental milestones, hearing and feeding abilities, education and language exposure patterns, with the latter section adapted from the Language Background Questionnaire formulated by Gatt (2010). Specific focus was placed on the languages with which participants were addressed at home and school, as well as exposure through the media. This was intended to provide an outlook on the language environment of each child, which also allowed the analysis of vocabulary measures in context. During each interview, the child was left to interact with his/her SLP.

2.2.5 Data coding and measures

The total scores obtained in the VC, PNT and LS were analysed to determine the total vocabulary (TV) per participant. The TV was expressed in terms of overlapping and non-overlapping scores. The overlapping score consisted of the sum of all the words counted in each assessment measure (including the total number of words (tokens) produced in the LS), irrelevant of multiple occurrences across datasets, to provide insight on the talkativeness of each child. The non-overlapping count consisted of a composite score, which was made up of the number of different words available in the child's vocabulary. Words reported more than once were computed in terms of a matching score, which evidenced the number of repeated words.

Similar performance across methods further confirmed validity and objectivity of findings. An auxiliary observer was employed to inter-transcribe a LS chosen at random, to verify the consistency and accuracy of transcripts, as suggested by Lammie Glenn et al. (2010). Agreement was 87% for tokens and 96% for types in the sample.

2.2.6 Data analysis

Analysis of data combined a quantitative approach using descriptive statistics to explore common trends in the participant group, and a qualitative account of individual performance. A tentative comparison with lexical development trends identified in TD Maltese children (Gatt, 2010) was also attempted.

3 Results

3.1 Individual analyses

C1, a boy who was primarily English-speaking, was 2;10 years at the time of testing. His expressive language profile consisted of a combination of key-word signing (based on Maltese sign language) and single words, many of which were not yet fully intelligible. According to the BQ, C1 attended an independent kindergarten school twice weekly. Exposure to television and stories was conveyed in English. C1 achieved a composite non-overlapping TV score of 19 words across the three assessment measures, which consisted mainly of English (55%) and Generic words (45%). A total of 10 content words and two function words were reported in the VC. Familiarity with colour terms was evident. Unintelligibility in the production of words was reported in the VC and also observed in the PNT. C1 often pointed at picture items and produced the social word 'there', instead of labels (see Table 2). Only one word was reported twice across measures, resulting in a matching score of 1. No evidence of expressive lexical items emerged in the LS.

Table 2. Performance on the picture naming task (PNT) per participant

Picture item	C1	C2	C3	C4	C5	C6	C7
Ballun/ball	✓	✓	✓	✓	✓	✓	✓
Karozza/car	there	✓	✓	✓	✓	✓	✓
Qattus/cat	NR	✓	miaw	NR	✓	✓	✓
Tarbija/ baby	there	✓	✓	✓	✓	✓	✓
Żarbun/ shoes	NR	✓	✓	boots	✓	✓	✓
Kelb/dog	there	✓	✓	✓	NI	✓	✓
Pupa/doll	there	✓	*pupi* (dolls)	✓	✓	✓	✓
Ajruplan/ aeroplane	✓	✓	✓	✓	NI	✓	✓
Telephone	✓	NI	✓	hello	✓	✓	✓
Tazza/glass	there	✓	KWS	drink	P	jar	NR
Rota/ bicycle	there	✓	✓	NI	✓	✓	✓
Bajda/egg	there	✓	P	✓	✓	✓	✓
Kitarra/ guitar	there	NR	NR	✓	✓	✓	✓
Għasfur/ *pappagall*/ bird/ parrot	✓	✓	✓	chicken	✓	✓	✓
Kuċċarina/ *mgħarfa*/ (tea/table) spoon	P	NR	✓	✓	NI	✓	✓
Kappell/ hat	NR	✓	✓	✓	✓	✓	✓
Fjura/ flower	there	NI	✓	✓	✓	✓	✓
Petne/comb	NR	brush	*xagħri* (my hair)	✓	P	✓	brush

Key: NR = no response; NI = not intelligible; KWS = key-word signing; P = prompted; ✓ = correct response

C2's language profile at 4;4 years consisted of simple word combinations, which reportedly emerged at approximately three years of age. He had one older sister who was TD. He attended a state school where both Maltese and English were used interchangeably. The child's L1 was Maltese, although the incorporation of some English words in his repertoire was reported. Television programmes and story-telling were mostly provided in English. C2 achieved the highest composite score of 341 words among children in his age group (C3 and C4), of which 61% were Maltese, 16% English and 23% Generic. Maltese words were observed more frequently in the VC and PNT data than in the LS. The knowledge of both Maltese and English word forms to represent particular items, such as 'ball' and '*ballun*', was reported in the VC. The child was observed to spontaneously label the picture of a ball in Maltese in the PNT. A total of 301 content words were reported in the

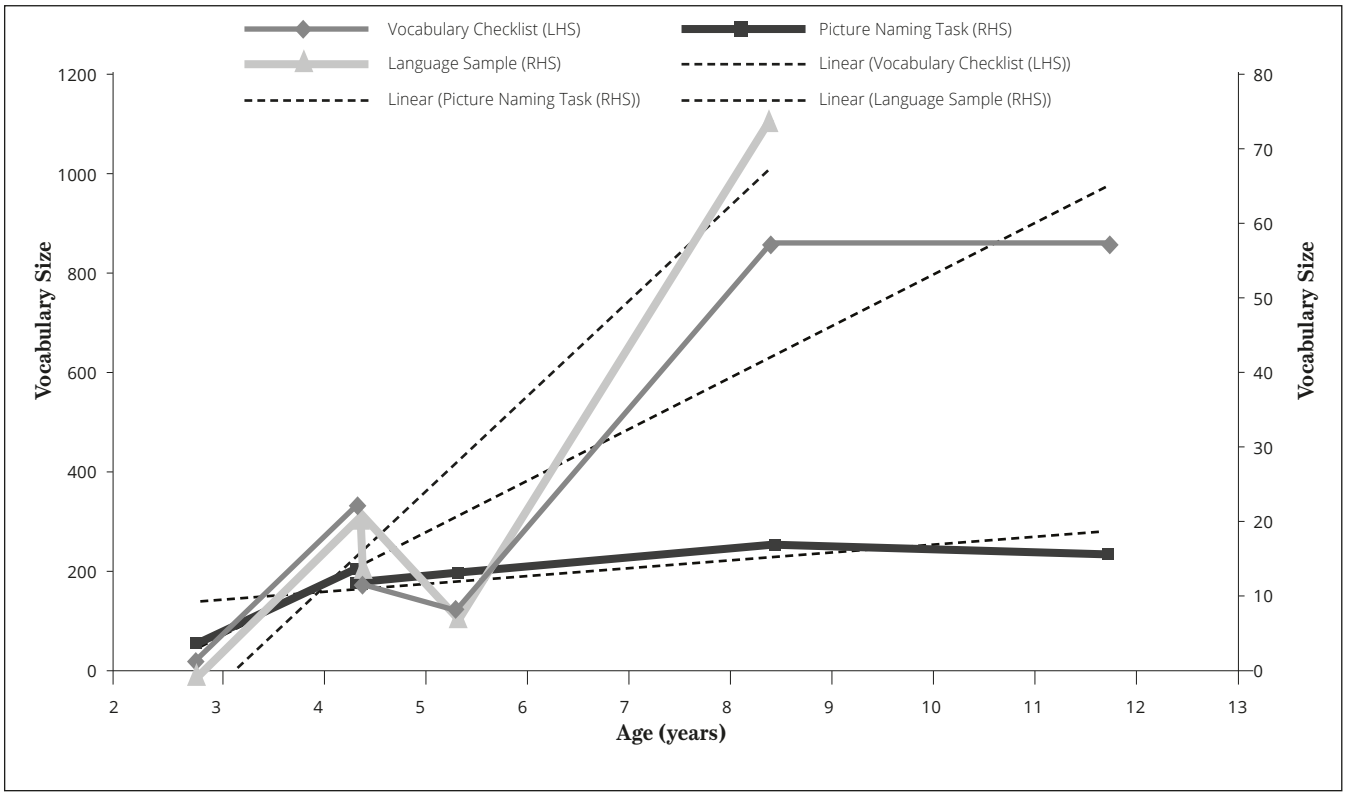

Figure 1. Vocabulary measures obtained through the vocabulary checklist (VC) plotted against the left hand side (LHS), and the picture naming task (PNT) and language sample (LS) plotted against the right hand side (RHS), including linear trend lines for the progression of vocabulary size across ages

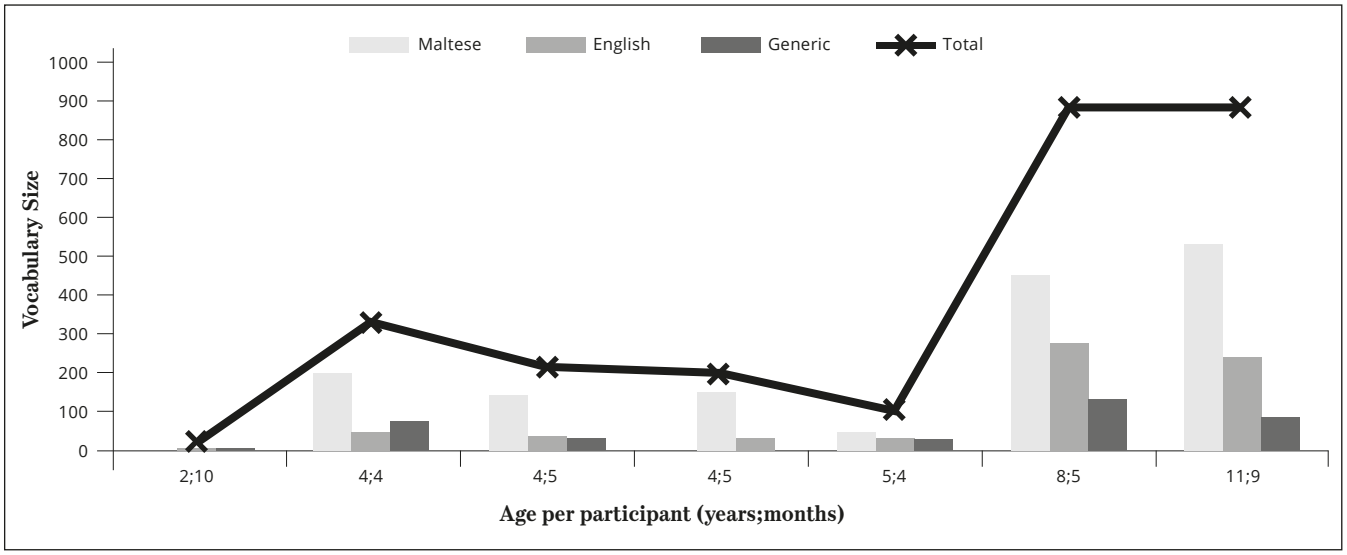

Figure 2. Total number of recognised and recalled words in the vocabulary checklist (VC), including language classification according to Maltese, English and Generic words, for each participant

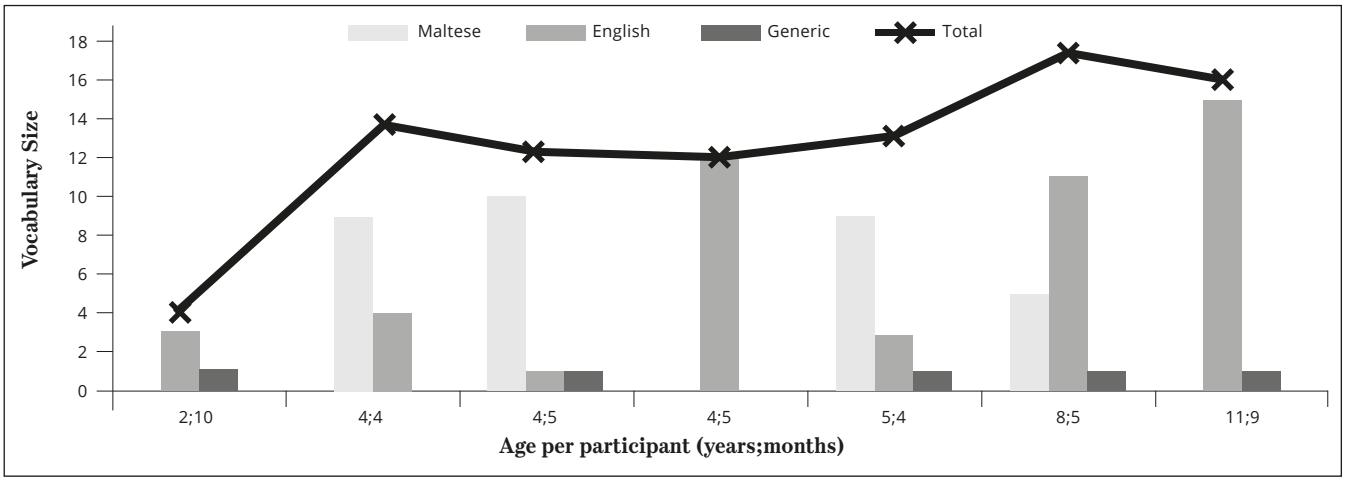

Figure 3. Total number of target words scored in the picture naming task (PNT), including language classification according to Maltese, English and Generic words for each participant

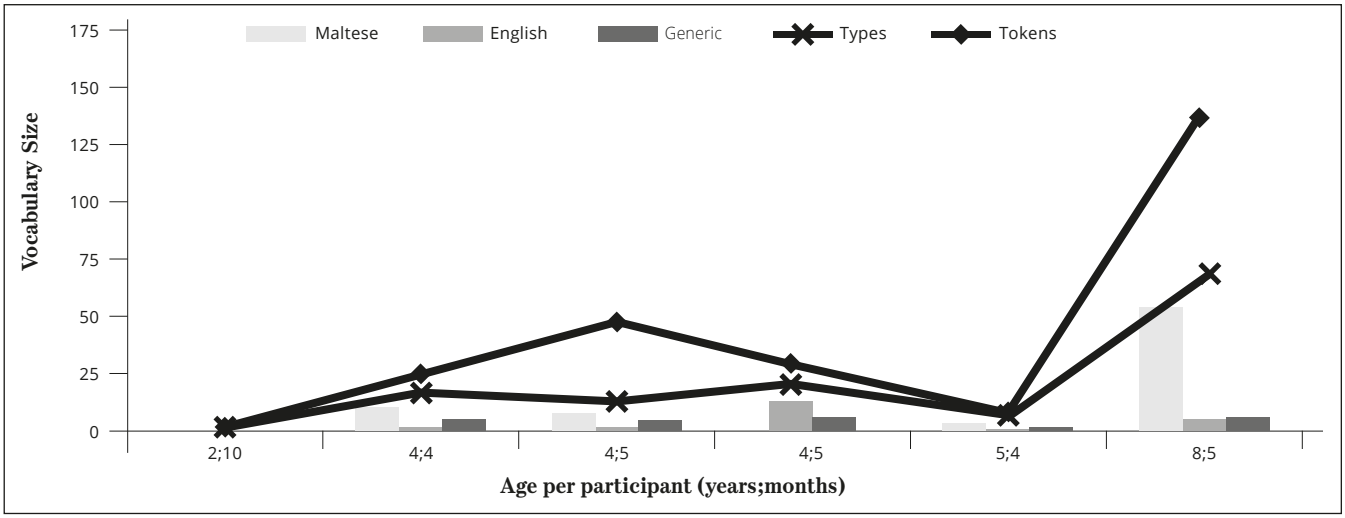

Figure 4. Number of types and tokens recorded in the language sample (LS), including language classification according to Maltese, English and Generic words for each participant

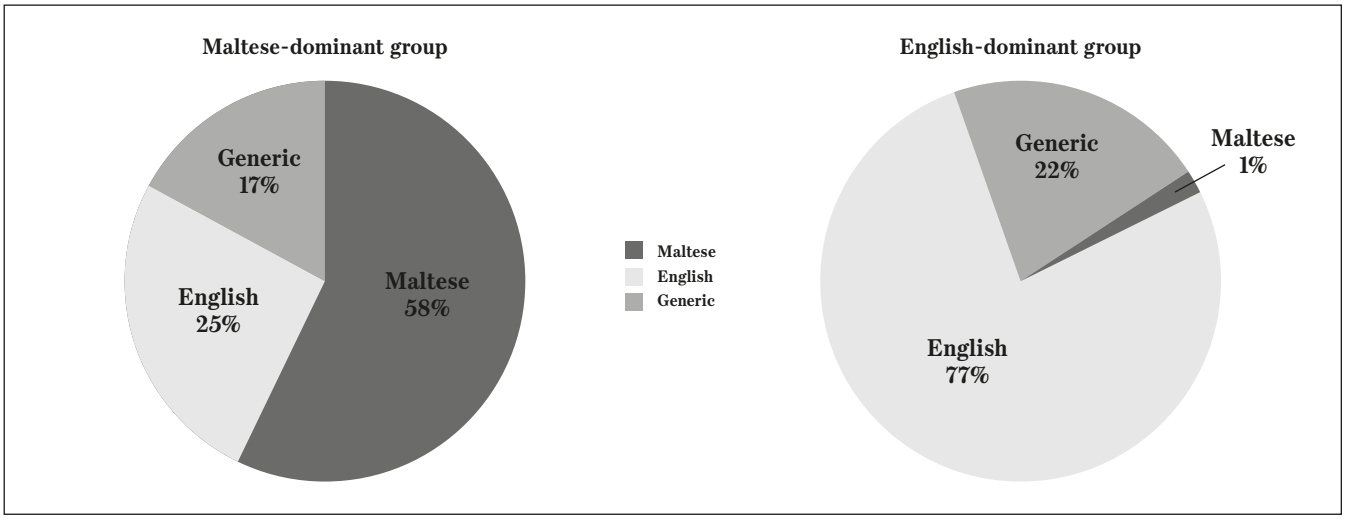

Figure 5. Percentage of words spoken according to different language classes for Maltese-dominant (N = 4) and English-dominant (N = 3) groups

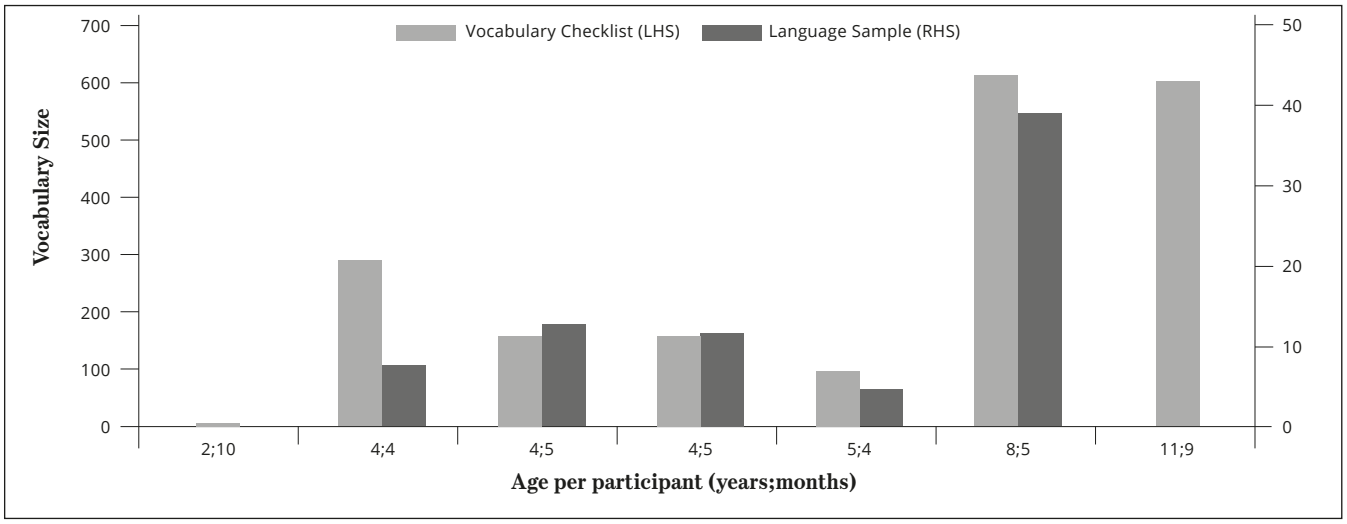

Figure 6. Content words recorded for each participant, with the vocabulary checklist (VC) scores plotted against the left hand side (LHS) and language sample (LS) scores plotted against the right hand side (RHS)

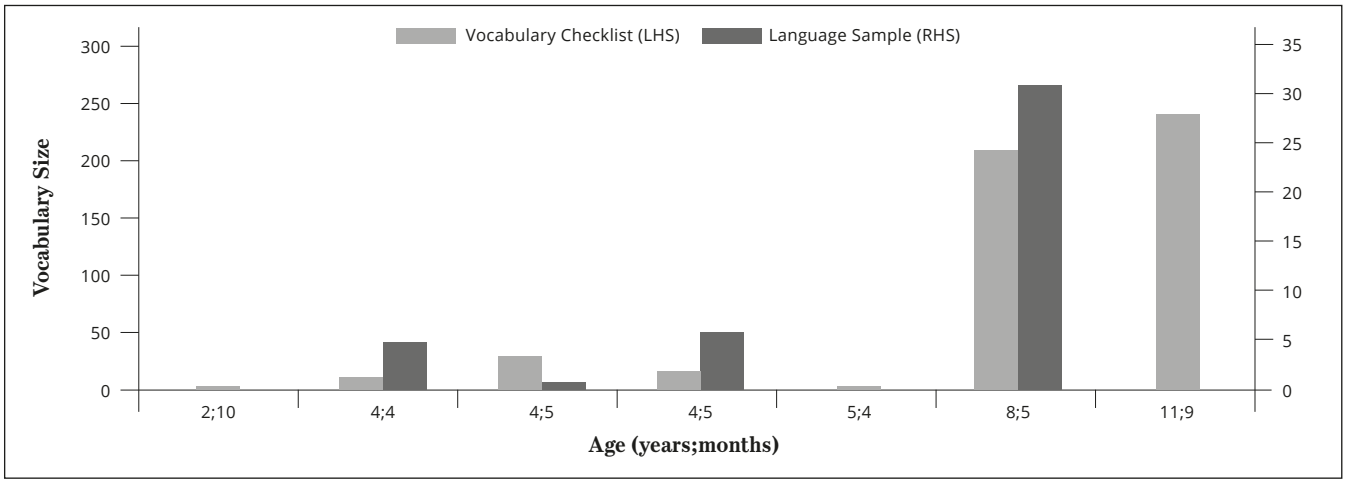

Figure 7. Function words recorded for each participant, with the vocabulary checklist (VC) scores plotted against the left hand side (LHS) and language sample (LS) scores plotted against the right hand side (RHS)

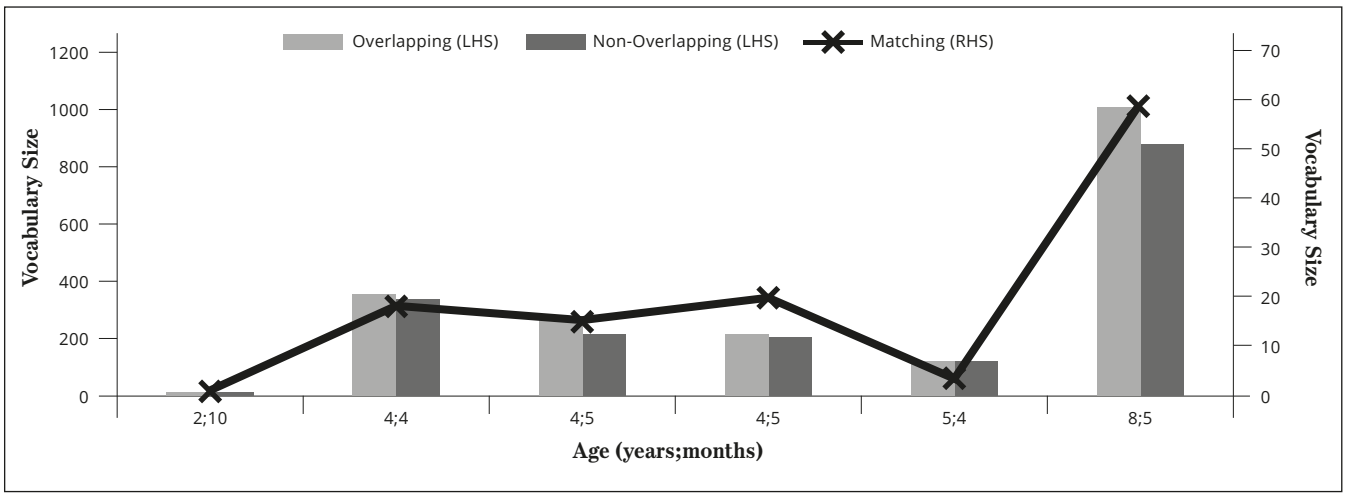

Figure 8. Total vocabulary (TV) including overlapping and non-overlapping scores plotted against the left hand side (LHS), and matching scores for each participant plotted against the right hand side (RHS)

VC, and eight in the LS. Thirteen function words including pronouns were identified in the VC, while an example (‘*bħalu*’) was expressed in the LS. A matching score of 18 was calculated across all three vocabulary measures.

C3, a 4;5-year-old girl, was primarily Maltese-speaking. The use of some English words was reported to be inevitable within the home setting. There was also some exposure to Italian television. C3 attended a state school where Maltese was the L1. Story exposure often varied between Maltese and English. C3 showed a TV of 225 words, with a percentage of 69% Maltese, 16% English and 15% Generic components. Despite minimal exposure to Italian, no spontaneous expression in this language was recorded. The VC data comprised recognised words only, with no words added in the recall section. A preference for Maltese was observed for verbal labelling during direct assessment, with only one word on the PNT and two words in the LS produced in English. The use of social (onomatopoeic) words was common in the LS e.g., the production of a clicking sound to represent ‘horse’. Many of the sound effects sampled were correspondingly marked in the VC. Despite the production of various sounds to represent words, 169 content words were still recorded in the VC and 13 in the LS, while 31 function words were marked in the VC and only one was produced in the LS. The production of function words reported in the VC was not observed in the LS. Repetition of words was common in the LS, with 16 types and 48 tokens calculated. A matching score of 18 across assessment measures was identified.

C4, a 4;5-year-old boy, had one older TD brother. His first word was spoken at approximately 2;0 years and word combinations were emerging. Feeding problems, including aspiration and chewing difficulties, had been a hurdle in C4’s development. These were under control at the time of data collection. The child’s L1 was English. However, both Maltese and English were used at the state school he attended. Media exposure consisted of English. A TV of 216 words was recorded across measures, of which 79% were English, 20% Generic and only 1% Maltese. The lowest percentage translated into the use of two Maltese words recognised by the caregiver in the VC, namely ‘*nanna*’ and ‘*nannu*’, which the child employed to refer to his grandparents. Content words totalled 164 in the VC and 12 in the LS, while the 17 function words reported in the VC were not observed among the six words counted in the LS. A matching score of 19 was calculated across the VC, PNT and LS.

C5, a boy aged 5;4 years, had two older TD sisters aged approximately seven and 10 years. C5’s family was primarily Maltese-speaking, although mixing of Maltese and English within the home setting was common. Mixing was reported to be more evident in the first two years of C5’s life, prior to enrolment in a state school. Adequate comprehension of both Maltese and English was reported, while verbal expression consisted mainly of Maltese with the inclusion of some English words. Television and stories comprised visual exposure through non-verbal programmes and picture books. Across measures, 130 spoken words were calculated for C5, of which 48% were Maltese. English and Generic components both amounted to 26%. The VC revealed the Maltese production of ‘*ballun*’ (ball). The word ‘ball’ was not recognised by the caregiver in the VC, yet was expressed in English on the PNT. In the VC, 105 content words and four function words were reported. The LS revealed five content words and no use of function words. The VC showed the child’s tendency to use sounds to represent animals, which was also noted in the LS. Evidence of C5’s knowledge of words to represent animals also in Maltese was observed. A total of three matching words were counted.

C6, a girl of 8;5 years, was approximately 2;6 years old when she spoke her first word. The child attended a Church school where English was the primary language of exposure with limited inclusion of Maltese. The latter, however, was the child’s L1. C6’s parents agreed that she was able to use both languages adequately to communicate her needs. Television and DVDs were provided in English, while exposure to stories took place in both languages. A TV of 953 spoken words was estimated across measures, comprising 55% Maltese, 31% English and 14% Generic words. The words calculated in the VC and LS consisted mainly of Maltese items, while the PNT revealed a majority of English words. The VC showed the use of 628 content words and 210 function words. In the LS, 40 content words and 31 function words emerged. A matching score of 61 was calculated.

C7 (11;9 years) spoke her first word at around two years of age and began to form single word combinations at approximately four years. Maltese was reportedly used more than English among family members within the first few years of her life, yet English was considered her L1. C7 attended an independent school, which was also primarily English-speaking. Language exposure thus consisted mainly of English with the use of some Maltese words during communicative exchanges as well as television and story-telling exposure. A total of 878 words were calculated, based on the VC and PNT (61% Maltese, 29% English and 10% Generic words). A majority of Maltese words was recorded in the VC. No words were expressed in Maltese during the PNT. The VC revealed the use of 622 content words and 240 function words. No scores were available for the LS due to technological failure of the recording equipment.

3.2 Group analysis

Descriptive statistics showed that the number of spoken words gradually increased with participant age, particularly for participants beyond the ages of four to five years (Figure 1). A breakdown of language classification in terms of the total number of words recognised and recalled on the VC shows that 54% of the words reported were Maltese, 31% English and 15% Generic (Figure 2). Participant vocabulary grew by an average of 103.5 words per yearly increase in age, with the sharpest improvement at 8;5 years. Figure 3 depicts 38% of the words expressed in the PNT as Maltese, while 56% were English and 6% Generic. An average increase of one picture recognised per year was calculated. Based on a total possible raw score of 18 on the PNT, the highest score (94%) was achieved at 8;5 years of age, while an equal score of 67% was obtained by the two 4;5-year-olds. Participants aged 4;4 and 5;4 years achieved an equal score of 72%. Based on the computed type counts, the words produced in the LS were mostly Maltese (62%), followed by Generic (20%) and English (18%) words respectively (Figure 4). An average increase of 12.8 different words per year for the participant group was identified. A decline in spoken words was observed at 5;4 years, while a sharp increase was evident from this age up to 8;5 years. Generally, an increase in the number of different words produced by a participant was coupled with a comparable increase in tokens. The Maltese-dominant group (C2, C3, C5, C6) appeared to use a higher percentage of English words than the English-dominant group (C1, C4, C7) used Maltese words, with a difference of 24% (Figure 5). Comparable percentages of Generic words were spoken in both groups. A larger number of content words than function words was calculated in the VC than in the LS (Figure 6). A marked increase in content words, identified at 4;4 years, was interrupted by a gradual decrease until 5;4 years and once again exploded up to 8;5 years. More function words in the VC than in the LS were evident (Figure 7). A considerable difference in overlapping and non-overlapping vocabulary scores was evident in the maximal calculated TV, with a matching score of 61 represented at 8;5 years (Figure 8). The smallest composite vocabulary (TV) was identified in the youngest participant, followed by participant C5 aged 5;4 years.

4 Discussion

This study aimed to investigate early lexical production skills of bilingual Maltese children with DS. Findings showed that vocabulary grew with participant age, corresponding with findings for TD Maltese children aged 1;0 to 2;6 years (Gatt, 2010). More specifically, a considerable growth in vocabulary development beyond four and five years was identified.

Parent-reported information showed first words to appear at an average age of 2;3 years. This corresponded with findings from

Berglund, Eriksson and Johansson's (2001) study, where the onset of lexical acquisition varied between the ages of one and two years in children with DS. While the VC indicated a TV of 19 different words in the youngest participant (2;10 years), Oliver and Buckley (1994) estimated a comparable number of approximately 24.4 words spoken at 2;6 years, also according to parent-reported information. In the current study, the largest improvement across vocabulary measures was prominent at 8;5 years and no advancement at a higher age point was evident. The participant with the widest vocabulary appeared to be the most talkative, which corresponded with Gatt's (2010) findings and hence confirmed the phenomenon of wider expressive vocabularies among more talkative children.

One must keep in mind that classification of words according to their grammatical features is difficult in early lexical development (Caselli et al., 1995). The grammatical categories probed in this study highlighted word forms based on content words and function words generated by checklist data and sampling measures. Content words were used more frequently than function words on both the VC and LS measures. While children with the smallest vocabularies produced little to no function words, the latter were more evident with increasing age and consequently larger vocabularies, further complementing Caselli et al.'s (1995) findings. The results suggested a trend in vocabulary development not only for TD children across languages, but also among children with DS, as far as demonstrated by the limited dataset in the current study.

In terms of language use (i.e., either Maltese or English), the highest percentage of words spoken per participant matched the child's reported L1. The occurrence of over-extensions[4] among the majority of participants, irrelevant of chronological age or vocabulary size, was observed. These were mainly expressed through the use of sound effects (e.g., to represent animals) and semantic associations (such as 'brush' instead of 'comb').

In the PNT, it is possible that word meaning conveyed by the participants did not necessarily coincide with the conventional form. Nevertheless, the misinterpretation of picture items may have led to erroneous responses. Picture naming resulted in the preference of English labels (56%) over Maltese ones (38%). A likely reason for this is the formal structure of the PNT, which may have imposed a certain pressure on participants, thus leading to conventional responses with the intention to meet expectations. Caselli et al. (1995) proposed that word usage is most likely subject to preference, not ability. However, it is also likely that academic and therapeutic routines may have influenced performance. Checklist data confirmed a higher predominance of Maltese words in relation to recognised and recalled items. This sheds light on methodological bias that may be associated with situational impact. For example, the VC was based on parent-reported lexical expression across a range of daily settings, whereas the LS provided an informal opportunity for word use during free play.

The Maltese-dominant group generally appeared to use more English words (25%) than the English-dominant group used Maltese (1%), while Generic words were relatively on a par in both language groups. A likely reason for the use of English lexemes is the absence of Maltese equivalents, as proposed by Gatt et al. (2008). Moreover, Feltmate & Kay-Raining Bird (2008) acknowledged code mixing in adult input as an obvious factor influencing children's vocabularies. The latter authors claimed that the vocabulary of bilingual children with DS may in fact surpass that of their monolingual counterparts. With this in mind, it may be accepted that bilingualism should not affect lexical acquisition in children with DS.

Some limitations in the present study were identified. A condensed sample size was not the original intention of the research design. However, the inclusion criteria allowed a constrained group of eligible participants. Methodological biases, namely parental inclination in the VC and response constraints in the PNT, may have impinged on the data. Missing LS scores for participant C7 must also be considered. Still, consistency emerging across the triad of assessment measures employed signifies validity in results.

4 The use of a word for a broader range of referents than in the adult language.

5 Conclusion

The current study revealed that productive vocabularies of Maltese bilingual children with DS escalated with increasing age, notwithstanding inevitable individual variation. Findings further extend existing research by demonstrating that, based on the sample group, Maltese bilingual children with DS were indeed able to develop expressive vocabulary skills in the context of their exposure to both Maltese and English languages. Moreover, they, too, had the potential to use the two languages functionally.

Further research may benefit from a multiple baseline approach across ages, to investigate sequential development of bilingual expressive vocabulary. Investigation of the effect of primarily monolingual versus balanced bilingual input for children with DS on language development may assist clinical decisions taken by professionals for optimal language exposure in the local context of bilingualism.

6 Funding

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7 Conflicts of interest

The authors report no conflicts of interest.

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Research Paper

MALTESE CHILDREN WITH A HEARING IMPAIRMENT: ANALYSIS OF THE CURRENT SITUATION AND ITS IMPACT ON THE QUALITY OF LIFE OF PARENTS

Stefan Sciberras, Ritienne Grima
Department of Communication Therapy, Faculty of Health Sciences, University of Malta, Msida, Malta

Abstract. Understanding the effect that a diagnosis of a childhood hearing loss has on parents would help professionals adopt an approach which diminishes parents' possible negative feelings and concerns. A sample of parents of children with hearing impairment was interviewed to document demographic data related to hearing loss in Maltese children. The present study also attempted to analyse the effect of hearing loss on the parents' wellbeing. Parents of 23 children with a hearing loss and parents of eight children without a hearing loss participated in the study. Structured face-to-face interviews were carried out with both groups of parents. A self-devised questionnaire was used with parents of children with hearing impairment to collect information about several factors related to the hearing loss. This included documentation of the different types, degrees and causes of hearing loss as well as the ages of suspicion, diagnosis and amplification of children with a hearing loss. The World Health Organisation Quality of Life-BREF (WHOQOL-BREF) questionnaire (World Health Organisation [WHO], 1998) was then used with both groups of parents to obtain a quality of life profile in four domains: physical health, psychological health, social relationships and environment. Slight quality of life differences, which were not statistically significant, were noticed between parents of children with a hearing loss and parents of children without a hearing loss. Quality of life scores were marginally higher for parents whose gap between the day of diagnosis and the interview date was more than 24 months, when compared to parents whose gap was 24 months or less. These findings extend the limited data on the effect of hearing loss on parents' quality of life in the Maltese context. More intensive support may be indicated for parents of children with hearing impairment, particularly in the initial stages following a diagnosis of a hearing loss. Support would help parents better understand and accept their child's hearing impairment.

Keywords: hearing impairment, hearing loss, quality of life, parents, children, Malta

1 Introduction

Hearing loss is a partial or total inability to hear. It is the most common sensory impairment, which considerably increases with age (World Health Organistion [WHO] & The World Bank, 2011). It is estimated that 360 million people worldwide have a disabling hearing loss, of which 32 million (9%) are children (WHO, 2013). Hearing loss is believed to be more common in boys than in girls. In fact, the Gallaudet Research Institute (2011) reports a male to female ratio of 1.2:1. From their literature review, Stevens et al. (2013) found that the global prevalence of hearing loss (with an average hearing level of 35 dB or more in the better ear) was 1.4% for children aged five to 14 years, 9.8% for females older than 15 years and 12.2% for males older than 15 years. Moreover, Hille, van Straaten and Verkerk (2007) report a prevalence of 3.2% in neonatal intensive care units. The latest census on the Maltese population revealed that 5,673 (0.01%) of the census respondents felt that they were not able to hear clearly, of whom 46 were younger than 10 years of age and 108 were aged from 10 to 19 years (National Statistics Office, 2014).

Childhood hearing loss can result in an impaired ability to communicate, inadequate language acquisition leading to inability to interpret speech sounds, economic and educational disadvantages and social isolation (Stevens et al., 2013). Furthermore, individuals with a hearing loss may be at a social disadvantage in both developing and developed countries (Olusanya, Ruben & Parving, 2006). Untreated hearing loss may also have an emotional impact on the individual with a hearing impairment (Garstecki & Erler, 2009).

Several types of hearing loss have been identified. In 1999, Grech collected data from 76 (81%) of the 94 hearing impaired individuals who received a service from the Special Education Department in Malta and Gozo. The sample population included data from 46 boys and 30 girls aged between 1;11 and 17;10 years. From her parental interviews, the author found that 70 (92%) of the subjects had a bilateral loss, while six (8%) suffered from a unilateral loss. Furthermore, 50 subjects (66%) had a congenital loss while 15 subjects (20%) had an acquired loss. Parents of 11 (14%) subjects were uncertain whether the loss was congenital or acquired.

Diefendorf (2009) states that 50% of the cases of congenital sensorineural hearing loss have a genetic cause, with 70% of these being non-syndromic and the remaining 30% being associated with particular syndromes. Diefendorf adds that the other 50% of cases of congenital sensorineural hearing loss have an environmental cause, with the following being the most common: bacterial infections, viral infections, ototoxic antibiotics, environmental toxins, physical trauma and acoustic trauma. A considerable number of hearing loss cases are of unknown aetiology. In fact, the Gallaudet Research Institute (2011) found that the aetiology of the hearing loss was not known in 57.8% of their sample.

When describing a hearing loss, it is also important to describe the extent of the severity of the loss. The degree of hearing loss affects speech production and spoken language outcomes (Sininger, Grimes & Christensen, 2010). Grech (1999) explained that although information about the degree of hearing loss collected from her local sample may have been subjective, in most cases parents acknowledged a substantial degree of loss.

Correspondence to Stefan Sciberras
(stefan.sciberras.10@um.edu.mt)
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Ozcebe, Sevinc and Belgin (2005) carried out research in Turkey on children with severe to profound hearing loss whose hearing was not screened at birth and found that the mean ages of suspicion, diagnosis and amplification were 12.5, 19.4 and 26.5 months respectively. Similarly, Jafari, Malayeri & Ashayeri's (2007) study on children with profound hearing impairment revealed that the mean ages of suspicion, diagnosis and amplification were 12.6, 15.2 and 20.5 months respectively. A study on the Maltese population showed that only 6.1% of the individuals reported to have a congenital hearing loss were diagnosed by six months of age. Moreover, 75.5% of the subjects were identified after one and a half years of age, with more than half of the sample being identified beyond three years of age (Grech, 1999).

There is currently a lack of data on the demographics of Maltese children with a hearing loss. The current study takes a parental perspective to document the types, degrees and causes of hearing loss as well as the ages of suspicion, diagnosis and amplification in Maltese children. This study also aims to understand the effect that a hearing loss may have on the parents' quality of life (QOL).

1.1 The effect of the child's hearing loss on the family

A diagnosis of hearing loss does not only affect the individual with hearing impairment but would possibly affect the QOL of the entire family. In their study, Mitchell and Karchmer (2004) found that 92% of children with hearing impairment are born to hearing parents. This means that the majority of parents have little or no experience of hearing loss. In fact, the presence of a child with hearing impairment in a hearing family may be a cause of family stress (Moores, Jatho & Dunn, 2001) and it may have a drastic impact on all the areas of family life, with several factors influencing the degree and type of impact (Jackson & Turnbull, 2004). Childhood hearing loss may influence multiple dimensions of family life including the child, other family members, and their participation in the community (Jackson, Traub & Turnbull, 2008). A diagnosis of a hearing loss may also affect the QOL of the extended family. Grandparents, for example, may experience disappointment, grief and loss as a result of a diagnosis of hearing loss in their grandchildren (Morton, 2000).

1.1.1 Age of diagnosis of hearing loss

Late diagnosis of a hearing loss may have an even more negative effect on the family. In fact, Young and Tattersall (2007) found that an overwhelming majority of parents whose children were diagnosed early were positive about the fact that the hearing loss was identified early, regardless of the degree of the loss. Frustrations and negative family experiences associated with a later diagnosis may be attenuated or prevented with early identification and early intervention (Jackson, Wegner & Turnbull, 2010). Since newborn hearing screening reduces the age at which infants with hearing impairment are diagnosed and treated, it would, in turn, improve the quality of parents' and infants' life (Canale et al., 2006). However, even though parents clearly support knowing early, early knowledge may bring emotions of grief and distress (Young & Tattersall, 2007). Knowing early may put pressure on parents to perform within a timetable in order for their child not to lose any of the advantages of early intervention. Parents of children with hearing impairment face important decisions, including the type of assistive technology which the child may benefit from and the communication modality that may be chosen for the child. Support for parents of children with hearing impairment is crucial in order for parents to be able to make informed decisions about their child's future.

1.1.2 The way in which diagnosis is reported

Breaking bad news is a difficult task for professionals since people may react differently to a diagnosis of hearing loss. Indeed, parents perceive the time immediately after the diagnosis as detrimental to their QOL (Burger et al., 2005). Jackson, Traub and Turnbull (2008) reveal that feelings reported by parents whose children were diagnosed with a hearing loss included shock, fear and uncertainty about the future, denial and indifference. Planning and implementation of effective strategies for breaking bad news should be an integral part of universal newborn screening programmes (Gilbey, 2010). Grech's (1999) research on the parents of 76 Maltese children with hearing impairment showed that 18.4% of the parents felt that the diagnosis was reported too quickly, while 10.52% felt that they were given no support immediately after the diagnosis. Other parents felt that the way in which the diagnosis was reported was cold and lengthy, with no explanation given.

1.1.3 Change in stress and quality of life (QOL) with time

The wellbeing of families of children with hearing impairment may change with time. Lederberg and Golbach (2002) found that when children with hearing impairment were 22 months old, there was a significant difference between stress levels of their mothers and those of mothers of same-age children with typical hearing, whilst there was no significant difference when the children were three and four years old. Burger et al. (2005) revealed that, with time, there was an improvement in the QOL of both parents of children who received cochlear implants and parents of children who were fitted with hearing aids. The median age of the children who used hearing aids in this study was 28.8 months whilst the median age of children who used cochlear implants was 29.1 months. Burger et al. believe that the process of adjustment to the child's hearing loss and the improvement in language development are important influential factors of the QOL of parents.

Meinzen-Derr et al. (2008) found that, following a diagnosis of hearing loss, carers' stress related to emotional wellbeing and health care decreases with time, whilst stress related to educational aspects increases with time. Family stress following a cochlear implantation may not decrease with time, as a result of the high expectations parents have at the beginning of the implantation and rehabilitation processes, and since parents' attitudes may become less positive with time (Weisel, Most & Michael, 2007).

1.1.4 Support for families of children with hearing impairment

Appropriate support for families of children with hearing impairment may reduce the negative effects of permanent hearing loss (Fitzpatrick et al., 2008). Parents who are informed bluntly of an existing hearing loss and who are not given support may feel helpless and frustrated (Gilbey, 2010). One of the recurring themes reported by parents of children with hearing impairment was the importance of parent support groups and the need for social networks with other parents (Jackson, Wegner & Turnbull, 2010). Professionals need to be attuned to the needs of the extended-family members. Morton (2000) believes that grandparents of deaf children may benefit from support groups which would help them express their negative feelings. Further examination of the impact of deafness on family members may assist clinicians in providing family-centred support following identification of a hearing loss. Providing information and support to the parents would in turn enhance the children's language acquisition and educational achievement following diagnosis of a hearing loss (Kushalnagar et al., 2010).

Grech's (1999) study showed that counselling was recommended for 34% of the families which, in most cases, was provided by teachers of the hearing impaired. Grech reported that family support groups are helpful in supporting parents of children with hearing impairment. Spiteri et al. (2004) insist that there needs to be more support for parents and professionals working with deaf children in Malta. There are no official support groups for parents of children with hearing impairment in this country. However the Malta Cochlear Implant Association offers such support (D. Camilleri, personal communication, June 3, 2015). The Deaf People Association (Malta) also gives support to parents of

children with a hearing loss (A. Vere, personal communication, January 22, 2014).

The following research questions were addressed in the current study:

- What are the different types, degrees and causes of hearing loss among children in Malta?
- What are the ages of suspicion, diagnosis and amplification of children with hearing impairment in Malta?
- What feelings did the diagnosis of a hearing loss evoke in parents and would parents have benefitted from more counselling and support?
- Is there a significant difference between the QOL of parents whose children use hearings aids, the QOL of parents whose children use cochlear implants and the QOL of parents of children without a hearing loss?
- How does time after diagnosis of a hearing loss affect the QOL of parents?

2 Methods

2.1 Research design

In the attempt to answer the research questions, a mixed research approach was used in this study. A convergent parallel mixed method design allowed the merging of quantitative and qualitative data to provide a comprehensive demographic overview of hearing loss in Maltese children, as well as parental QOL as reported by the parents themselves (Creswell, 2014). Quantitative data was obtained through close-ended questions whilst qualitative data was gathered through open-ended questions.

Various methods for data collection were considered, including postal questionnaires, internet questionnaires, self-administered questionnaires and face-to-face interviews. The face-to-face interview approach was chosen because this enables the interviewer to clarify questions and to encourage participation and involvement of the respondents (Robson, 2011). This approach is the best for making use of open-ended questions, as it enables the interviewer to build a better rapport with the interviewee and to have more control over the response situation (Czaja & Blair, 2005). Interviews were audio recorded in order for the researcher to be able to analyse the parents' exact responses.

2.2 Participants

Two different samples were required for this study. Sample A included 23 mothers and 16 fathers of 23 children with a hearing loss aged between 0 and 6;11 years, with a mean age of 4;10 years (SD = 20.25). Thirteen children (57%) were males and 10 (43%) were females. Participants were recruited through the Audiology Department of the state general hospital of Malta. Parents of 27 children with hearing impairment who use the state general hospital services were first approached by the audiologist of the hospital. Parents of 23 children (85%) accepted to participate in the study. Two questionnaires, described in Sections 2.3.1 and 2.3.2, were used with these parents.

Sample B served as a small control group and included eight mothers and seven fathers of eight children without a hearing loss, aged between 0 and 6;11 years, with a mean age of 4;10 years (SD = 17.18). Gender of the children was equally distributed in the sample. Participants of Sample B were randomly recruited from community parent and child groups. These parents were approached by the president of these groups. The parents were then contacted by the researcher for an appointment to be set up. The Milestones of Development Checklist (Childsupport, 2007) was used with the parents who accepted to participate in the study. Parents of all eight children stated that, to their knowledge, their children were typically-developing. All parents reported that their children achieved more than 90% of the milestones expected according to their chronological age. Hence, all eight children were considered as being within the range of typical development (Dosman, Andrews & Goulden, 2012). Subsequently, the hearing of each child was tested under the guidance and supervision of a qualified audiologist. The audiologist confirmed that all eight children had a hearing level within the normal range. The researcher then interviewed the parents using Questionnaire 2, which is described below.

2.3 Research tools

Two different questionnaires and a checklist were used in this study. Below is a description of these research tools.

2.3.1 Questionnaire 1: Evaluation of factors related to children with hearing impairment and their parents

This questionnaire, which consists of 26 questions, was formulated following an extensive literature review (Gilbey, 2010; Grech, 1999; Jafari, Malayeri & Ashayeri, 2007; Lederberg & Golbach, 2002; Meinzen-Derr et al., 2008), and highlights various factors related to hearing loss (Table 1). Questionnaire 1 was devised in English and later translated to the Maltese language. This questionnaire included a variety of close-ended and open-ended questions in order to obtain more comprehensive responses from the parents of children with hearing impairment.

Table 1. Themes analysed in Questionnaire 1

Questions	Themes analysed
1-4	Type of hearing loss Degree of hearing loss Cause of hearing loss Presence of any additional impairments
5-23	Factors related to suspicion of hearing loss Factors related to identification of hearing loss Factors related to the amplification device used
24-26	Availability of resources and counselling Modes of communication used with the hearing impaired child

2.3.2 Questionnaire 2: The World Health Organisation Quality of Life-BREF (WHOQOL-BREF)

A number of QOL questionnaires were considered including the Adult Carer Quality of Life Questionnaire (Elwick et al., 2010), the Second European Quality of Life Survey Overview (European Foundation for the Improvement of Living and Working Conditions, 2009) and the World Health Organisation Quality of Life-BREF (WHOQOL-BREF) (WHO, 1998). The WHOQOL-BREF was considered to be the best tool to obtain QOL scores from the parents of both groups. This questionnaire assesses persons' perceptions of their position in life in the context of the culture and value system where they live, in relation to their goals, expectations, standards and concerns (WHO, 1998). This questionnaire consists of 26 items which include two questions on the overall perceived QOL and satisfaction with health, followed by 24 items which are based on four different domains: physical health, psychological health, social relationships and environment (Table 2). The WHOQOL-BREF has good to excellent psychometric properties of reliability and performs well in preliminary tests of validity (Skevington, Lotfy & O'Connell, 2004). All items were rated on a 5-point Likert-form scale with a higher score indicating a higher QOL. Scores for each domain were transformed to a common 4-20 scale in order to facilitate the interpretation of results (WHO, 1996). The WHOQOL-BREF was interview-administered in order to avoid problems concerning the understanding of questions. Questionnaires with more than 20% of the

data left unanswered (or one item in the three-item social domain) were discarded (as suggested by WHO, 1996). Permission to use the English and Maltese versions of the WHOQOL-BREF was sought and granted by the WHO.

Table 2. Domains of the WHOQOL-BREF

Domains	Facets incorporated within domains
Physical Health	Activities of daily living
	Dependence on medicinal substances and medical aids
	Energy and fatigue
	Mobility
	Pain and discomfort
	Sleep and rest
	Work capacity
Psychological	Bodily image and appearance
	Negative feelings
	Positive feelings
	Self-esteem
	Spirituality / Religion / Personal beliefs
	Thinking, learning, memory and concentration
Social relationships	Personal relationships
	Social support
	Sexual activity
Environment	Financial resources
	Freedom, physical safety and security
	Health and social care: accessibility and quality
	Home environment
	Opportunities for acquiring new information and skills
	Participation in and opportunities for recreation / leisure activities
	Physical environment (pollution / noise / traffic / climate)
	Transport

2.3.3 Milestones of Development Checklist

The Milestones of Development Checklist, proposed by Childsupport (2007), was used in this study to document the children's stage of development in different aspects, including cognitive, motor, socio-emotional, and speech and language, from their parents' perspective. This checklist was only used with the control group, in order to reduce the possibility of including parents of children who were not following typical developmental stages. Children who achieved more than 90% of the milestones expected according to their chronological age were considered as being within the range of typical development. High-quality evidence suggest that the 90th percentile criterion can quickly identify typical versus atypical development (Dosman, Andrews & Goulden, 2012).

2.4 Ethical considerations

The study was approved by the University of Malta's Research Ethics Committee (proposal number 035/2013). Confidentiality was assured to all participants. Before meeting with parents of samples A and B, the respective parents were provided with a recruitment letter detailing all the necessary information about the study and about participation. Consent forms were signed by each parent who was willing to participate in the study. Participants had the right to withdraw their consent from the study at any time without penalty, even after the interview was finished.

3 Results and Discussion

3.1 Type, aetiology and degree of hearing loss

The results presented in Table 3 reveal the types of hearing loss reported by the parents. Twenty subjects (87%) were reported as having a sensorineural hearing loss[1] which was stable, while three (13%) were reported as having a mixed hearing loss[2] which was fluctuating. Parents of 19 children (83%) stated that the hearing loss was bilateral, while parents of four children (17%) reported a unilateral loss. Fifteen subjects (65%) were reported as having a congenital loss. In eight subjects (35%), parents were uncertain whether the loss was congenital or acquired. In 15 subjects (65%), a sudden loss was reported while parents of eight children (35%) were uncertain whether the loss was sudden or progressive. Table 4 displays the degree of hearing loss in the left and right ear of the children with hearing loss as reported by parents. The vast majority of subjects in this study were reported as having a hearing loss which ranged from moderately severe to profound. This may imply that a number of children with milder losses may not have been identified or may have used audiological services from the private sector. The lack of a neonatal hearing screening programme may be one of the reasons why children with mild losses are missed (Grech, 1999).

Table 3. Type of hearing loss

Type of hearing loss	*N*	%
Stable	20	87
Fluctuating	3	13
Bilateral	19	83
Unilateral	4	17
Congenital	15	65
Uncertain whether congenital or acquired	8	35
Sudden	15	65
Uncertain whether sudden or progressive	8	35

Note. N = frequency; % = percentage

Table 4. Degree of hearing loss

Degree of hearing loss	*N*	%
Normal	0	0
Slight	0	0
Mild	2	9
Moderate	2	9
Moderately severe	4	17
Severe	2	9
Profound	13	57

Note. N = frequency; % = percentage; guidelines proposed by the British Society of Audiology (2011) were used to calculate the average hearing level

Figure 1 summarises the reported causes of hearing loss. None of the parents reported rubella as being the cause of their child's hearing loss. This contrasts with data for congenital hearing loss in Malta, published in 1999, where 21% of the subjects reported that contracting rubella was the cause of their child's hearing loss (Grech, 1999). Primary prevention strategies such as increased awareness and immunisation against this disease may be the reason for the decline in the number of hearing losses caused by rubella.

1 A sensorineural hearing loss can be either cochlear, or more rarely, retrocochlear (Busacco, 2010)

2 A mixed hearing loss is a loss that has both sensorineural and conductive elements (Busacco, 2010)

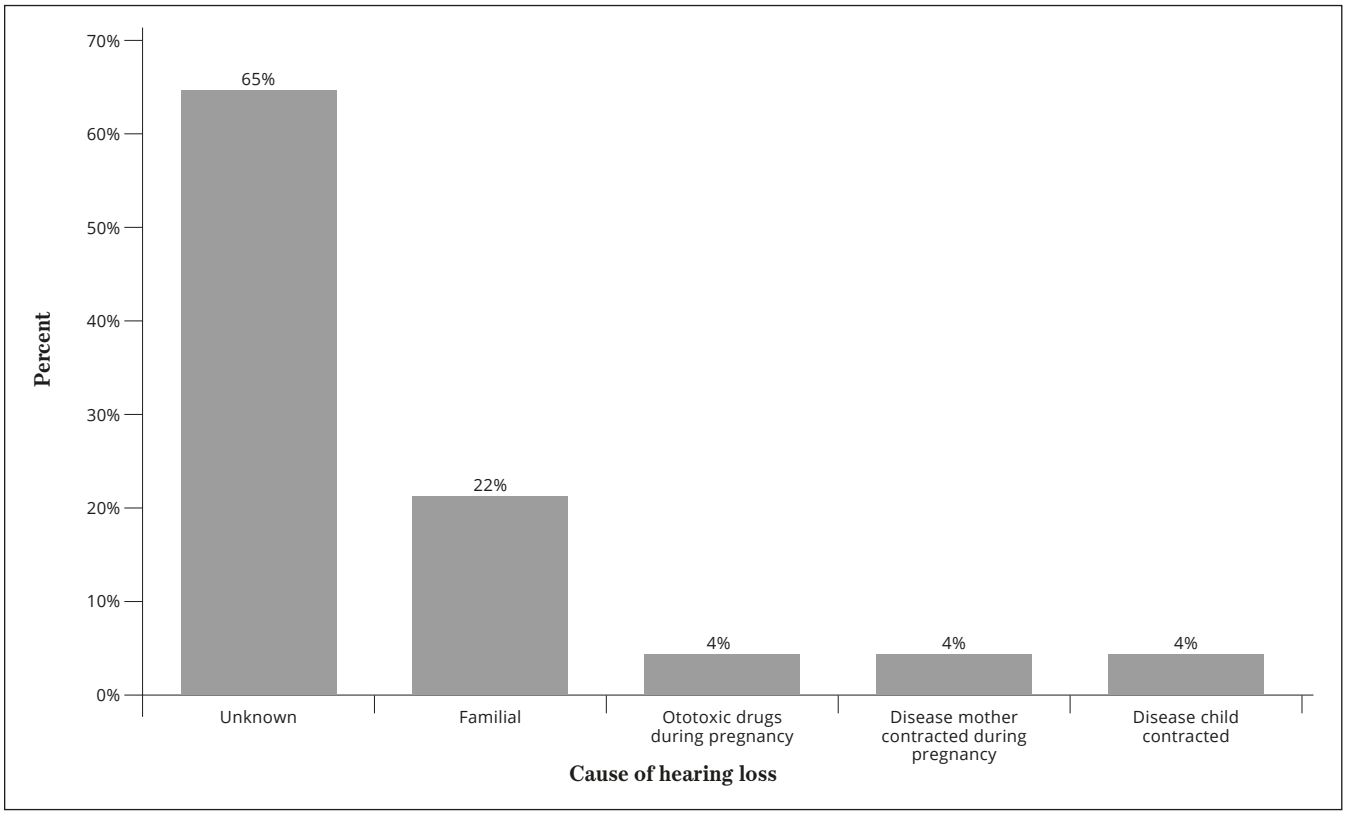

Figure 1. Cause of hearing loss

3.2 Suspicion, diagnosis and amplification of children with hearing impairment

Figure 2 shows an error bar graph of the mean ages of suspicion, diagnosis and amplification, as reported by parents. The mean gap between suspicion and diagnosis of a hearing loss was 6.00 months (SD = 5.43) while the mean gap between diagnosis and amplification was 3.22 months (SD = 3.55).

Hearing loss was suspected in the first year of life in nine subjects (39%). Similar to other studies (e.g. Grech, 1999; Harrison & Roush, 1996; Jafari, Malayeri & Ashayeri, 2007), the current study reveals that in the majority of the cases (70%), it was the parents or other family members who suspected the hearing loss. Parental reports from this study reveal that five subjects (22%) were diagnosed within the first year of life, while only three subjects (13%) received amplification devices by the time they reached one year. Furthermore, 10 subjects (43%) were diagnosed and received their first amplification after three years of age. From her study on the Maltese population, Grech (1999) found that 53% of children with hearing loss were diagnosed beyond the third year of age. Sharma, Dorman and Spahr (2002) argue that the brain has the highest plasticity in the first three and a half years of age, making this a critical window for language learning. Language learning has a critical period since infants and young children have a greater ability to learn language when compared to adults (Bruer, 2008). Humphries et al. (2012) emphasise that if children are not exposed to a natural language during early childhood, they might never be completely fluent in any language. It is evident that a considerable number of children with hearing impairment did not receive their amplification during the period in which language experiences can best contribute to optimal language development. There is currently no national newborn hearing screening programme in Malta and Gozo. However, the fact that children who receive intensive care have their hearing screened is a

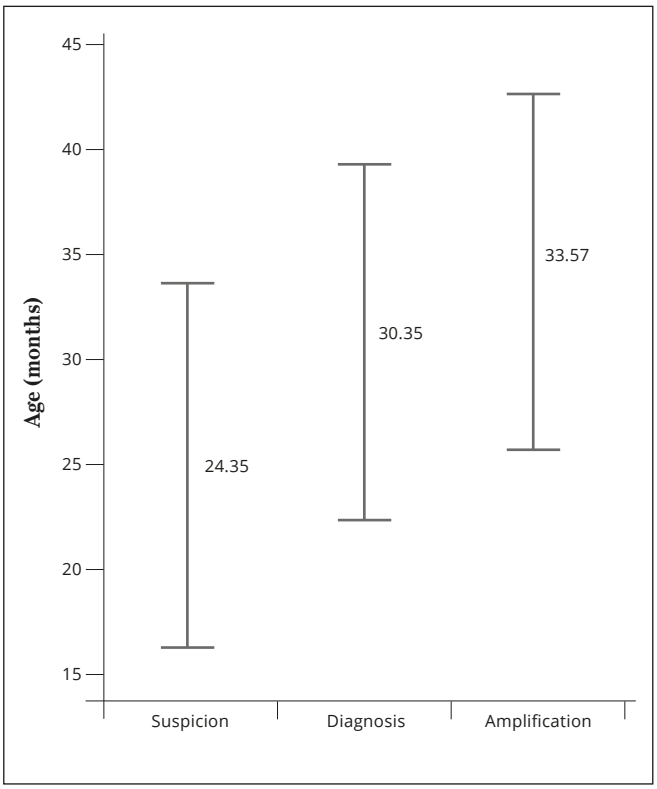

Figure 2. Mean ages of suspicion, diagnosis and amplification

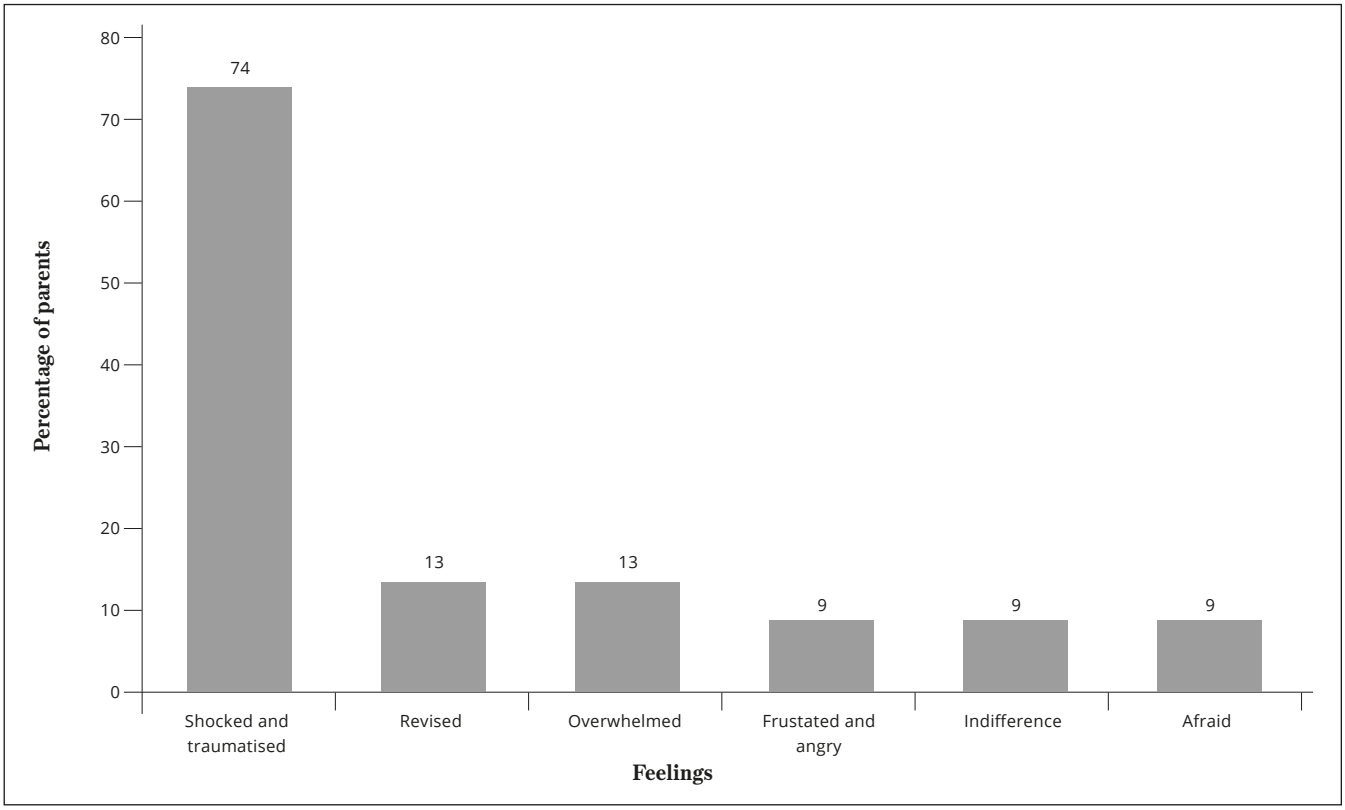

Figure 3. Feelings when the diagnosis was reported

good starting point, since there is a higher prevalence of hearing loss in such children (Hille et al., 2007). Patel and Feldman (2011) believe that the absence of a newborn hearing screening programme significantly delays identification of children with hearing impairment. In fact, the introduction of a newborn hearing screening programme in Slovakia resulted in a lower average age of diagnosis of children with a hearing loss (Jakubíková et al., 2009).

3.3 The impact of hearing loss on the parents' quality of life (QOL)

The feelings of the parents after receiving the diagnosis of a hearing loss are illustrated in Figure 3. The majority of parents in this study reported that the diagnosis of a hearing loss evoked negative emotions, with shock and trauma being the most commonly mentioned feelings. Congruently, Jackson et al. (2008) reveal that feelings reported by parents whose children were diagnosed with a hearing loss included shock, fear and uncertainty about the future, denial and indifference. When questioned about the support they received after diagnosis, parents of 13 subjects (57%) felt that they would have benefitted from more support, while parents of nine subjects (39%) believed that more support would not have been more beneficial for them. Parents of one subject (4%) were uncertain whether they would have benefitted from more support or not.

Thirteen mothers (57%) and 15 fathers (94%) from Sample A described their overall QOL as good or very good, while all mothers and fathers (100%) from Sample B described their QOL as good or very good. Moreover, 18 mothers (78%) and 13 fathers (81%) from Sample A were satisfied or very satisfied with their own health while seven mothers (88%) and all fathers (100%) from Sample B were satisfied or very satisfied with their own health.

A Kolmogorov-Smirnov test confirmed that the dependent variables, thus, the transformed QOL scores, had a normal distribution in all four QOL domains provided by mothers and fathers. The one-way Analysis of Variance (ANOVA) test was used to compare the mean transformed QOL scores provided by mothers and fathers for three independent groups, which included parents of children who use hearing aids[3], parents of children who use cochlear implants[4] and parents of children with normal hearing (Table 5). Figure 4 illustrates the mean QOL scores of mothers and fathers of the aforementioned groups. The discrepancy between the QOL of the three groups of parents was not statistically significant in any of the four domains. This may be attributed to the fact that, in the majority of the cases, more than one year had passed since the diagnosis and, thus, the parents may have habituated to the situation. As time goes by following a diagnosis of a hearing loss, parents acquire resources which enable them to adjust to the child's hearing loss (Burger et al., 2005). Moreover, the fact that all children received amplification devices may have affected the parents' QOL. In fact, Burger et al. (2005) argue that in many families, a return to normality may be noticed once amplification devices are fitted. Despite the fact that the difference between the three groups was not statistically significant, a discrepancy in the parents' satisfaction with the amplification devices used was observed. Whilst all parents of children who use cochlear implants were very satisfied with this device, half of the parents whose children used or were currently using digital behind the ear hearing aids reported that they were dissatisfied or very dissatisfied with this device. Parents of children who were currently using hearing aids rated their QOL slightly lower in all four domains when compared to parents of children who were using cochlear

3 This group included parents of children who use one or two hearing aids, depending on whether their loss was unilateral or bilateral.

4 This group included parents of children who use two cochlear implants and parents of children who use a cochlear implant and a hearing aid.

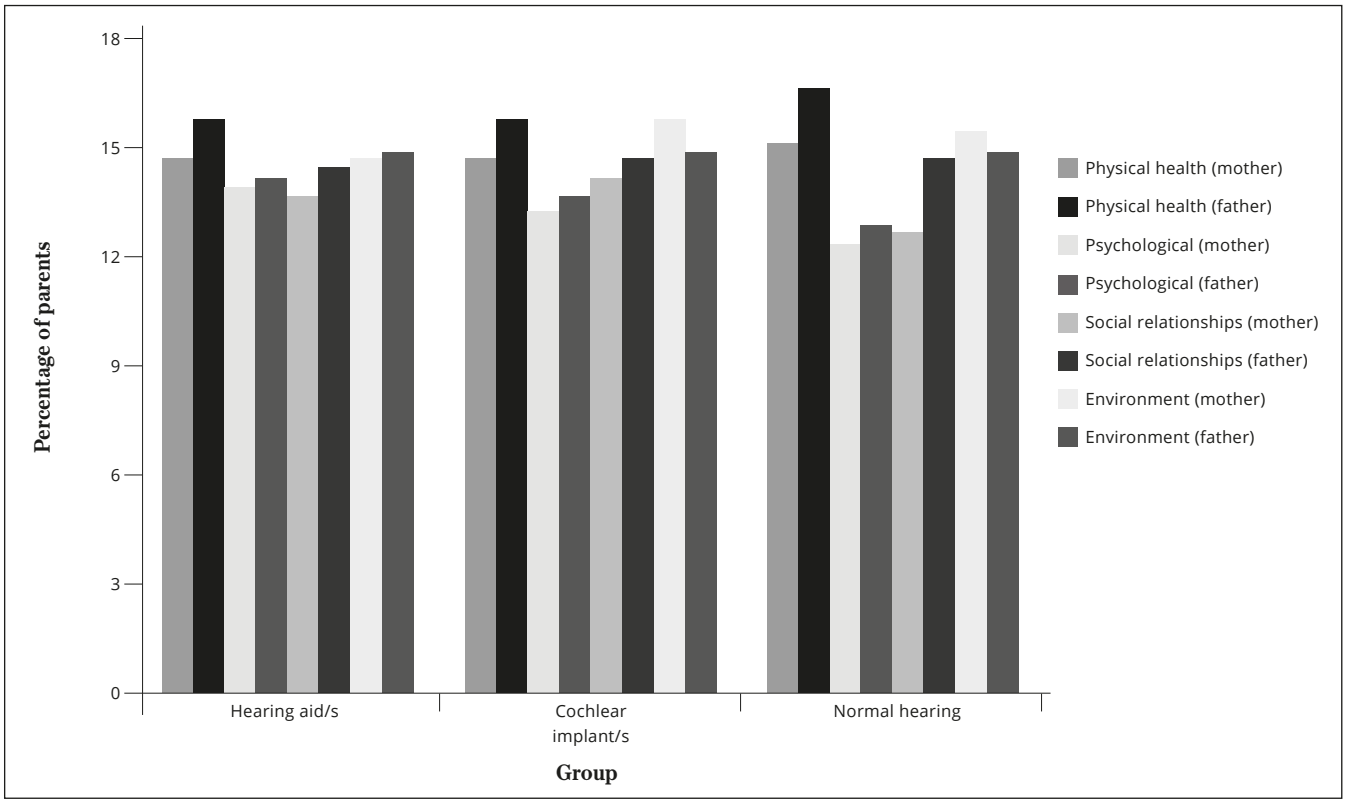

Figure 4. Analysis of quality of life

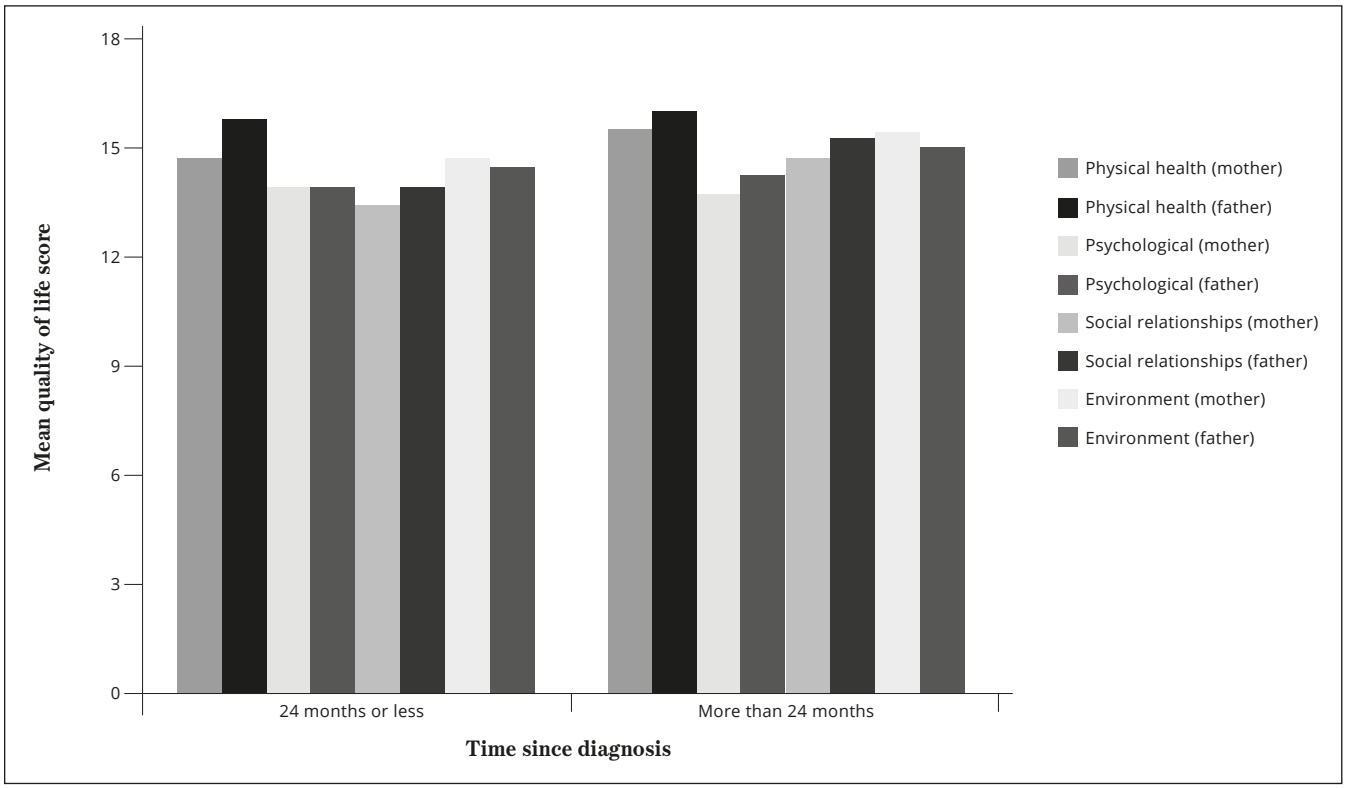

Figure 5. The effect of time after diagnosis on the quality of life

Table 5. One-way ANOVA analysis of parents' quality of life

Domain	Group	Mean	Std. Deviation	95% CI for Mean		*F*-statistic	*p*-value
				Lower Bound	Upper Bound		
Physical health (mother)	HAs	14.53	1.19	13.88	15.19	.923	.409
	CIs	14.75	1.39	13.59	15.91		
	NH	15.25	1.04	14.38	16.12		
Physical health (father)	HAs	15.55	1.93	14.92	16.17	2.101	.149
	CIs	15.60	1.14	14.18	17.02		
	NH	16.57	1.27	15.39	17.75		
Psychological health (mother)	HAs	13.87	1.13	13.24	14.49	.480	.623
	CIs	14.00	1.41	12.82	15.18		
	NH	14.38	1.06	13.49	15.26		
Psychological health (father)	HAs	14.27	1.19	13.47	15.07	.152	.860
	CIs	14.40	1.14	12.98	15.82		
	NH	14.57	1.98	13.67	15.47		
Social relationships (mother)	HAs	13.60	1.77	12.62	14.58	1.596	.221
	CIs	14.25	1.58	12.93	15.57		
	NH	15.00	2.07	13.27	16.73		
Social relationships (father)	HAs	14.45	1.64	13.36	15.55	.813	.458
	CIs	15.20	1.48	13.36	17.04		
	NH	15.29	1.25	14.13	16.45		
Environment (mother)	HAs	14.40	1.45	13.59	15.21	2.236	.126
	CIs	15.13	1.46	13.91	16.34		
	NH	15.63	1.06	14.74	16.51		
Environment (father)	HAs	14.73	1.27	13.87	15.58	.754	.483
	CIs	15.00	1.71	14.12	15.88		
	NH	15.43	1.27	14.25	16.61		

Note. CI = confidence interval; HAs = hearing aid/s; CIs = cochlear implant/s; NH = normal hearing

implants. Furthermore, parents of children who were using cochlear implants had marginally lower QOL scores when compared to parents of children without a hearing loss. This implies that in the present study, the disparity in the satisfaction rating of the amplification device used did not significantly affect the QOL of parents in any of the four assessed domains.

The one-way ANOVA test was also used to compare the mean transformed QOL scores provided by mothers and fathers for another two independent groups. Parents were divided according to the gap between the day their child was diagnosed with a hearing loss and the day the researcher carried out the interview, either less than or equal to 24 months or more than 24 months (Table 6). Figure 5 shows a clustered bar graph of the mean QOL scores of mothers and fathers of the above mentioned groups. QOL scores were marginally higher for mothers and fathers whose gap between the day of diagnosis and the interview date was more than 24 months, when compared to parents whose gap was 24 months or less. However, statistically significant differences have only been observed in the mothers' physical health (p = .029) and social relationships (p = .049) domains. In fact, Meadow-Orlans (1995) states that when compared to fathers, mothers of children with hearing impairment are more likely to experience stress. This could be because mothers may take more responsibility for the daily needs of their children (Jaffe & Cosper, 2015).

These results should be interpreted in the light of the small samples involved and, thus, assumptions may not be generalisable to the whole population. The limited number of participants possibly does not make the sample representative of the population of parents of children with a hearing loss as well as parents of children without a hearing loss. More studies with larger sample sizes, which employ a more in-depth analysis covering wider age ranges, are required. Since QOL is influenced by a considerable amount of variables, longitudinal studies may provide a better understanding of the process of adaptation and transitions of families of children with hearing impairment. Analysis of the impact of a hearing loss on other family members such as grandparents and siblings may also be carried out.

4 Conclusions and Recommendations

The results of this study support the claims in the literature that early identification and intervention is crucial for children with hearing impairment. The guidelines proposed by the Joint Committee on Infant Hearing (2007) for screening, identification and amplification of children with hearing impairment, may never be followed unless a newborn hearing screening programme is implemented. The fact that Malta is a small country with a manageable population is one of the advantages which may facilitate the implementation of a newborn hearing screening programme (Grech, 1994). More awareness campaigns on childhood hearing loss may also be provided to the general public. Such campaigns may help parents identify the signs of a hearing loss at an earlier stage, which could be fruitful for the identification of both congenital and acquired hearing losses.

Table 6. One-way ANOVA analysis of the effect of time after diagnosis on parents' QOL

Domain	Group	Mean	Std. Deviation	95% CI for Mean		*F*-statistic	*p*-value
				Lower Bound	Upper Bound		
Physical health (mother)	≤ 24 months	14.08	0.79	13.58	14.59	5.477	.029
	> 24 months	15.18	1.40	14.24	16.12		
Physical health (father)	≤ 24 months	15.29	0.76	14.59	15.98	1.028	.328
	> 24 months	15.78	1.09	14.94	16.62		
Psychological health (mother)	≤ 24 months	13.50	1.00	12.86	14.14	3.263	.085
	> 24 months	14.36	1.29	13.50	15.23		
Psychological health (father)	≤ 24 months	13.86	1.22	12.73	14.98	2.143	.165
	> 24 months	14.67	1.00	13.90	15.44		
Social relationships (mother)	≤ 24 months	13.17	1.53	12.20	14.14	4.373	.049
	> 24 months	14.55	1.64	13.45	15.64		
Social relationships (father)	≤ 24 months	13.86	1.46	12.50	15.21	4.163	.061
	> 24 months	15.33	1.41	14.25	16.42		
Environment (mother)	≤ 24 months	14.25	1.55	13.27	15.23	1.975	.175
	> 24 months	15.09	1.30	14.22	15.96		
Environment (father)	≤ 24 months	14.29	1.38	13.01	15.56	3.227	.094
	> 24 months	15.22	0.67	14.71	15.73		

Note. CI = confidence interval; domains with a p-value < .05 are in boldface.

The importance of support for parents and caregivers of children with hearing impairment cannot be overrated. More intensive support may be indicated for parents (especially the mothers) whose children have been diagnosed with a hearing loss. Counselling enables caregivers to make informed decisions about their child's future. Supporting the family of a child with hearing impairment will likely result in family growth, which can in turn be beneficial for the child (DeConde Johnson, 1997).

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7 Conflicts of interest

The authors report no conflicts of interest.

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Research paper

THE INFLUENCE OF PERSONALITY TRAITS ON THE WELLBEING OF MALTESE UNIVERSITY STUDENTS: A QUANTITATIVE STUDY

Charlot Cauchi, Katya DeGiovanni
Department of Family Studies, Faculty for Social Wellbeing, University of Malta, Msida, Malta

Abstract. The purpose of this study was to examine the relationship between personality traits and health-related behaviours among Maltese university students. Based on the Five-Factor Model of personality, this study uncovered significant differences between low, medium and high levels of personality traits and their association to health-related behaviours. Data were collected from a sample of 576 students who completed a questionnaire comprising a personality inventory and wellbeing questions. The findings suggested that conscientiousness and agreeableness related to health-promoting behaviours like reduced binge drinking and drug use. Similarly, extraversion and openness related to increased fruit and vegetable consumption. However, health-deterring behaviours were also observed. These included drinking and driving, as well as unsafe sexual practices. Neuroticism was linked to health-deterring behaviours that included lack of exercise and drug use. This study adds to the existing evidence that shows personality traits to serve as both protective and deterring factors to health. The novel findings on Maltese students' personality-health trends reported in this paper could be used to develop health promotion strategies aimed at specific personalities.

Keywords: personality traits, wellbeing, university students, Five-Factor Model, health-related behaviours

1 Introduction

In a recent report, the World Health Organisation (2014) noted that the prevalence of current tobacco smoking is a central predictor of tobacco-related diseases, while alcohol abuse can lead to alcohol dependence, hepatic cirrhosis and other injuries. In this respect, studies on student populations have reported students to be non-adherent to several health-promoting behaviours (American College Health Association, 2008) such as abstinence from alcohol and drug use, increased physical exercise and healthy eating. To understand these behaviours, researchers have frequently focused on personality-health studies (Gray & Watson, 2002).

Personality can be understood as a dynamic organisation of characteristics typical of an individual, which influences behaviours, cognition and motivations (Ryckman, 2013). Personality traits can serve as both protective and deterring factors to health, as is evident in previous literature (Raynor & Levine, 2009). The Five-Factor Model of personality has become a widely accepted construct, which measures and categorises personality traits into five dimensions known as *conscientiousness*, *extraversion*, *neuroticism*, *agreeableness* and *openness* (John & Srivastava, 1999). *Conscientiousness* refers to being organised, thorough and planful, with substantial impulse control. *Extraversion* refers to being energetic, talkative, assertive and active. *Neuroticism* refers to easily experiencing negative emotions like feeling tense, anxious and nervous. *Agreeableness* refers to being sympathetic, kind and appreciative towards others. Finally, *openness* refers to having wide interests, imagination and intelligence (John, Naumann & Soto, 2008). The Five-Factor Model has been used in various studies and extensive data support its reliability and construct validity across ages and cultures (Rhodes & Smith, 2006).

Personality-health studies on college and undergraduate students have established several relationships between personality traits and health behaviours. Such findings have been frequently replicated. The literature consistently suggests that conscientiousness is related to increased health-promoting behaviours (e.g., exercise) and a decrease in health-deterring behaviours (Bogg & Roberts, 2004; Löckenhoff et al., 2012). Individuals scoring high on conscientiousness engaged more in regular exercise (Rhodes & Smith, 2006) and reported fewer health risk habits, ranging from decreased alcohol consumption (Ruiz, Pincus & Dickinson, 2003), decreased risky driving (Arthur & Doverspike, 2001) and safer sexual behaviours (Ingledew & Ferguson, 2007). Raynor and Levine (2009) linked high conscientiousness in college students to alcohol-related harm reduction strategies, increased fruit and vegetable consumption, increased seat belt use and decreased binge drinking. Dubey et al. (2010) found that non-substance abusers had higher levels of conscientiousness compared to substance abusers, while Benotsch et al. (2013) observed how conscientiousness in undergraduates served as a protective factor against prescription drug abuse. Characteristics of extraversion have been consistently related to both health-promoting and health-deterring behaviours. Raynor and Levine (2009) linked extraversion to an increase in strengthening exercises. However, extraversion in students has also been related to higher levels of binge drinking (Benjamin & Wulfert, 2005), increased risky driving (Dahlen & White, 2006) and increased substance abuse (Dordinejad & Shiran, 2011). Students scoring high on extraversion were also involved in riskier sexual behaviours, including increased sexual partners (Ingledew & Ferguson, 2007) and decreased condom use (Raynor & Levine, 2009). A consistent pattern was found between neuroticism and health-deterring behaviours. Indeed, neuroticism was associated with unfavourable behaviours, like decreased exercise adherence (Rhodes & Smith, 2006) and an increase in alcohol consumption in college students (Littlefield, Sher & Wood, 2009). Similar to extraversion, high neuroticism was related to increased substance use (Dordinejad & Shiran, 2011) and increased prescription drug abuse (Benotsch et al., 2013). Agreeableness was related to an increase in health-promoting behaviours. Raynor and Levine (2009) linked high agreeableness in college students to reduced binge drinking, increased alcohol-related harm reduction strategies and a decrease in the number of sexual partners. Finally, characteristics of openness were related to both health-promoting and health-deterring behaviours. In their sample of undergraduate students, Courneya and Hellsten (1998) linked openness

Correspondence to Katya DeGiovanni
(katya.degiovanni@um.edu.mt)
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to increased exercise engagement, while Raynor and Levine (2009) linked openness to higher fruit and vegetable consumption. In contrast, Benotsch et al. (2013) found associations between high openness and prescription drug abuse in their undergraduate sample.

Personality-health literature has been used to develop strategic interventions aimed specifically at individuals with a particular personality type. For example, Conrad, Castellanos and Mackie (2008) targeted 11- to 15-year-old adolescents scoring high on extraversion, a trait highlighted by sensation seeking, with the aim of reducing alcohol abuse. Cognitive behavioural exercises and personality psycho-education were developed to facilitate coping strategies for these individuals. The results showed that interventions reduced the likelihood of binge drinking by up to 50% for up to 12 months. These results suggest that personality-health studies could allow the development of health-promoting interventions that are designed specifically for individual personality types.

Personality health has been widely researched across Europe and the United States. The current study aims to understand the link between personality traits and health-behaviour variables among Maltese students. Besides the main objective of replicating previous studies and allowing comparisons, this study serves as a basis for future personality-health research directed at other Maltese populations, such as younger students and older adult populations. It also addresses the limited personality-health research based in Malta and adds to the local health literature.

2 Methods

The study adopted a quantitative approach to allow data collection from a wide population range. Quantitative research is concerned with quantifying phenomena; hence, it provides a more objective type of measure (Langdridge & Hagger-Johnson, 2009). This approach was chosen so that the results attained would enable comparisons to the existent literature.

2.1 Participants

A sample of 576 Maltese university students responded from a total of 11,067, which represents a 5.2% response rate. Participants included 166 males (29%) and 410 females (71%) aged between 17 and 62 years, with a mean age of 24 years.

2.2 Research design

The questionnaire used consisted of three main sections. The first section included questions about demographics. The second section comprised a personality inventory with statements that identified personality traits. Personality traits were measured using the Big Five Inventory (BFI) (John, Donahue & Kentle, 1991). This inventory consists of 44 statements which are rated on a five-point Likert scale, where 1 corresponds to 'Strongly Disagree' and 5 corresponds to 'Strongly Agree'. Despite being a short scale measure, the BFI presents with an average Cronbach's alpha of .83. This is a level of reliability similar to that of other personality inventories such as the Neuroticism-Extraversion-Openness-Five-Factor Inventory (NEO-FFI) (Costa and McCrae, 1992) and the Trait Descriptive Adjectives (TDA) scales (Goldberg, 1992), which have an average Cronbach's alpha of .81 and .84 respectively. Convergent validity with other personality inventories was also substantial. The BFI showed an overall convergence of .80 with the TDA and a .77 convergence with the NEO-FFI (John et al., 2008). These characteristics motivated choice of the BFI for use in this study. The BFI was also deemed more time-efficient, hence reducing the possible lack of responses due to a lengthy questionnaire. The third section of the questionnaire included questions measuring health-behaviour variables. Health-behaviour questions were constructed with reference to a research tool by Cefai and Camilleri (2009), with some questions being amended or omitted. Health-behaviour questions were clustered into four sections, namely 'general health habits' (e.g., dietary habits, physical exercise), 'alcohol use', 'drug use' and 'sexual behaviours'. Most of these questions were measured on a five-point Likert scale, where 1 corresponded to 'Strongly Disagree' and 5 corresponded to 'Strongly Agree'.

2.3 Data collection and analysis

The questionnaire was piloted on ten randomly selected Maltese university students to gain feedback about any ambiguities. The pilot questionnaire was then adjusted by rewording questions, adding options and including definitions for terms such as 'junk food' and 'binge drinking'. The final version of the questionnaire was sent by email through the Registrar's Office at the University of Malta, to ensure that it reached the entire student population. In turn, this ensured randomness in participant recruitment because every student received the questionnaire in the same period and had equal opportunity to respond. The data collected were coded as numeric values and analysed using the Statistical Package for the Social Sciences (SPSS) Version 20. The significance threshold was set at .05. Descriptive and inferential statistics were carried out using the one-way Analysis of Variance (ANOVA) test and the Student-Newman-Keuls post hoc test, while Cronbach's alpha provided a measure of internal consistency between personality items.

2.4 Ethical considerations

Potential participants were briefed through an information letter before they were directed to the online survey. The briefing section stated that participation was voluntary, responses were anonymous and participants could stop their survey participation at any time. All responses were treated with full confidentiality and only the authors were able to access the collected data.

3 Results

In order to have representative data that allowed comparisons, personality trait scores were recoded into grouped variables. After observing the cumulative percentage in frequency tables, personality trait results were divided into three score categories: low, medium, and high. The internal consistency of the BFI was measured and results are displayed in Table 1. Cronbach's alpha for personality traits resulted in a reliability coefficient of .73. This shows good internal consistency, meaning that the measure reflected the construct that it was measuring.

Table 1. Cronbach's alpha reliability for personality traits

Personality traits	Cronbach's alpha	No. of items
Conscientiousness	.79	9
Extraversion	.82	8
Agreeableness	.74	9
Neuroticism	.83	8
Openness	.71	10
All personality variables	.73	44

3.1 Conscientiousness

As seen in Table 2, statistically significant differences for conscientiousness were found in four health-behaviour variables, namely healthy breakfast consumption, junk food consumption, binge drinking and cannabis use. Students who scored high on conscientiousness reported a significant increase in healthy breakfast

consumption (mean = 2.71) in comparison to students scoring medium or low. Students scoring high on conscientiousness also reported a significant decrease in junk food consumption (mean = 3.96), binge drinking (mean = 1.60) and cannabis use (mean = 0.55) in comparison to students scoring medium or low on conscientiousness.

Table 2. One-way ANOVA test for significant differences between low, medium and high scores on conscientiousness and health-behaviour variables

Conscientiousness mean rating score		*F*-statistic	Degrees of freedom	*p*-value
'I have a healthy breakfast'				
Low	3.35			
Medium	3.11	7.806	2, 573	.001***
High	2.71			
'I consume junk food'				
Low	3.62			
Medium	3.95	5.703	2, 573	.004**
High	3.96			
'I engage in binge drinking'				
Low	2.48			
Medium	1.79	27.023	2, 573	.001***
High	1.60			
'When was the last time you ever used or took any of the following? Cannabis'				
Low	.90			
Medium	.76	3.145	2, 573	.044*
High	.55			

Note. Conscientiousness score refers to low (10-28), medium (29-33) and high (34-45); *p < .05, **p < .01, ***p < .001

Table 3. One-way ANOVA test for significant differences between low, medium and high scores on extraversion and health-behaviour variables

Extraversion mean rating score		*F*-statistic	Degrees of freedom	*p*-value
'How many servings of fruits and vegetables do you consume daily?'				
Low	1.98			
Medium	2.57	5.687	2,573	.004**
High	2.37			
'I consume junk food'				
Low	3.64			
Medium	4.08	7.469	2,573	.001**
High	3.88			
'Average daily caffeine intake – cup/s of coffee'				
Low	.63			
Medium	.71	5.134	2,573	.006**
High	.89			
'In the last month, how many alcoholic drinks did you have on average at social events?'				
Low	1.40			
Medium	1.65	3.282	2,513	.038*
High	1.59			
'In the last month, did you drive after having five or more drinks?'				
Low	.70			
Medium	1.07	4.498	2,513	.012*
High	.83			
'In the last year, have you unintentionally become or gotten someone pregnant?'				
Low	2.23			
Medium	2.09	3.815	2,320	.023*
High	2.12			

Note. Extraversion score refers to low (9-22), medium (23-26) and high (27-40); *p < .05, **p < .001

3.2 Extraversion

Table 3 shows that statistically significant differences for extraversion were observed in six health-behaviour variables, namely fruit and vegetable consumption, junk food consumption, coffee consumption, alcohol use, drinking and driving and unintentional pregnancy. Students who scored higher on extraversion reported a significant increase in fruit and vegetable consumption (mean = 2.37) while reporting a significant decrease in junk food consumption (mean = 3.88) in comparison to students scoring lower on extraversion. Students scoring higher on extraversion also reported a significant increase in coffee consumption (mean = .89), alcohol use (mean = 1.59), drinking and driving (mean = .83), and unintentional pregnancy (mean = 2.12) when compared to students scoring lower on extraversion.

3.3 Agreeableness

As reported in Table 4, statistically significant differences for agreeableness were found in six health-behaviour variables, namely junk food consumption, binge drinking, cannabis use, ecstasy use, cocaine use and inhalant use. Students who scored higher on agreeableness reported a significant decrease in junk food consumption (mean = 3.95) and binge drinking (mean = 1.79) in comparison to students scoring lower on agreeableness. Similarly, students scoring higher on agreeableness reported a significant decrease in recent cannabis use (mean = .58), ecstasy use (mean = .08), cocaine use (mean = .09) and inhalant use (mean = .03) in comparison to students scoring lower on agreeableness.

Table 4. One-way ANOVA test for significant differences between low, medium and high Scores on agreeableness and health-behaviour variables

Agreeableness mean rating score		*F*-statistic	Degrees of freedom	*p*-value
'I consume junk food'				
Low	3.67			
Medium	3.91	3.584	2,573	.028*
High	3.95			
'I engage in binge drinking'				
Low	2.26			
Medium	1.78	8.586	2,573	.001***
High	1.79			
'When was the last time you ever used or took any of the following? Cannabis'				
Low	1.00			
Medium	.66	4.937	2,573	.007**
High	.58			
'When was the last time you ever used or took any of the following? Ecstasy'				
Low	.38			
Medium	.12	8.603	2,573	.001***
High	.08			
'When was the last time you ever used or took any of the following? Cocaine'				
Low	.29			
Medium	.18	3.243	2,573	.04*
High	.09			
'When was the last time you ever used or took any of the following? Inhalants'				
Low	.25			
Medium	.14	4.659	2,573	.01*
High	.03			

Note. Agreeableness score refers to low (19-31), medium (32-35) and high (36-44); *p < .05, **p < .01, ***p < .001

3.4 Neuroticism

Table 5 shows that statistically significant differences for neuroticism were observed in two health-behaviour variables: healthy breakfast consumption and ecstasy use. Students who scored high on neuroticism reported a significant decrease in healthy breakfast consumption

(mean = 3.34) in comparison to students scoring medium or low on neuroticism. Students scoring higher on neuroticism also reported a significant increase in ecstasy use (mean = 0.24) in comparison to students scoring lower on neuroticism.

Table 5. One-way ANOVA Test for significant differences between low, medium and high scores on neuroticism and health-behaviour variables

Neuroticism mean rating score		*F*-statistic	Degrees of freedom	*p*-value
'I have a healthy breakfast'				
Low	2.80			
Medium	3.03	5.328	2,573	.005**
High	3.34			
'When was the last time you ever used or took any of the following? Ecstasy'				
Low	.05			
Medium	.26	4.574	2, 573	.011*
High	.24			

Note. Neuroticism score refers to low (9-23), medium (24-28) and high (29-40); *$p < .05$, **$p < .01$.

3.5 Openness

As seen in Table 6, statistically significant differences for openness were found in two health-behaviour variables: fruit and vegetable consumption and sexual partners. Students who scored high on openness reported a significant increase in fruit and vegetable consumption (mean = 2.66) in comparison to students scoring medium or low in openness. Students scoring high on openness also reported a significant increase in the number of sexual partners (mean = 1.41) in comparison to students scoring medium or low on openness.

Table 6. One-way ANOVA test for significant differences between low, medium and high scores on openness and health-behaviour variables

Openness mean rating score		*F*-statistic	Degrees of freedom	*p*-value
'On average, how many servings of fruits and vegetables do you consume daily?'				
Low	1.98			
Medium	2.23	8.035	2,573	.001**
High	2.66			
'How many sexual partners did you have within the last academic year?'				
Low	1.17			
Medium	1.34	3.252	2, 333	.04*
High	1.41			

Note. Openness score refers to low (19-33), medium (34-37) and high (38-50); *$p < .05$, **$p < .001$

4 Discussion

The aim of this study was to understand the link between personality trait scores and health-behaviour variables in undergraduate students. The results showed that significant differences were observed between different personality trait scores and both health-promoting and health-deterring behaviours. Results are discussed under three sub-sections. The first section discusses the results for conscientiousness and agreeableness, which were primarily linked to health-promoting behaviours. The second part discusses how results for extraversion and openness were linked to both health-promoting and health-deterring behaviours. The final section discusses neuroticism, which was exclusively linked to health-deterring behaviours.

4.1 Health-promoting traits

High conscientiousness scores underlined a significant increase in healthy breakfast consumption and, simultaneously, a significant decrease in junk food consumption. This result emphasises how self-discipline in conscientious individuals not only promotes healthier practices, but also reduces health-deterring lifestyles. Moreover, conscientiousness seems to protect against health-deterring behaviours as results suggest a significant decrease in alcohol use. In contrast, students scoring low on conscientiousness were significantly more likely to report drinking excessively. This finding is similar to that reported in an earlier study by Ruiz et al. (2003) and shows that conscientious individuals may be more self-controlling and, hence, better able at regulating alcohol consumption. Raynor and Levine (2009) found that binge drinking was reported significantly higher in lower conscientiousness students. An explanation for this could be that conscientious individuals are perhaps better able at controlling impulses, thus reducing the likelihood of binge drinking. Cannabis use was linked significantly less with students scoring higher on levels of conscientiousness. Hallucinogen use was reported significantly less by high conscientiousness students when compared to medium conscientiousness, although low conscientiousness in students did not result in increased hallucinogen use. This replicates a finding by Dubey et al. (2010) that showed lower conscientiousness scores to be more associated with drug users than non-drug users. Results for high agreeableness showed a significant decrease in junk food consumption. Furthermore, agreeableness also highlighted a significant decrease in binge drinking, which complements earlier studies (Raynor & Levine, 2009; Ruiz et al., 2003). Results for agreeableness were also linked to a significant decrease in drug use, including cannabis, ecstasy, cocaine and inhalants. These results suggest that high agreeableness students are perhaps more concerned about the welfare of others. Hence, they are less likely to engage in risky behaviours that negatively influence others.

4.2 Health-promoting and health-deterring traits

High extraversion scores were linked to a significant increase in fruit and vegetable consumption. Simultaneously, students reported a significant decrease in junk food consumption. This suggests the adoption of a healthier lifestyle by such individuals. Extraversion was also linked to a significant increase in coffee consumption, which highlights the facet of stimulation seeking. Indeed, extraverted students perhaps consume coffee more regularly since the caffeine acts as a stimulant, complementing their energetic personality. On the other hand, students scoring low on extraversion reported a significant decrease in drinking at social events compared to medium extraversion, although high extraversion did not underline a significant increase in alcohol use. This result replicates previous findings in which increased extraversion was linked to higher alcohol consumption and binge drinking (Benjamin & Wulfert, 2005). Furthermore, students scoring low on extraversion showed a significant reduction in drinking and driving behaviours compared to medium extraversion, although high extraversion students did not relate to increased drinking and driving behaviours. This complements earlier studies in which higher extraversion was related to an increase in risky driving (Dahlen & White, 2006). Notably, low extraversion seems to act as a protective factor against alcohol use. However, it is medium extraversion that increased alcohol use, rather than high extraversion. This suggests that the relationship between extraversion levels and alcohol use is complex. Nevertheless, these results underline how extraverted individuals somehow seek stimulating opportunities. Facets of extraversion, like being outgoing and enjoying the company of others, may contribute to an increased likelihood of engaging in excessive drinking. In contrast to conscientiousness, extraversion does not seem to protect against health-deterring behaviours. Unintentional pregnancy was reported significantly more in persons with higher extraversion scores. This corroborates previous studies in which extraversion was linked to

increased numbers of sexual partners (Ingledew & Ferguson, 2007) and decreased condom use (Raynor & Levine, 2009). Certainly, the combined facets of sociability and sensation seeking may explain why extraverted individuals are more likely to engage in risky behaviours, including unprotected sex. Students with high openness scores reported a significant increase in fruit and vegetable consumption, replicating a finding of an earlier study by Raynor and Levine (2009). This result suggests that students with high openness perhaps have a greater willingness to consider new ideas, such as adhering to a healthier lifestyle. Conversely, openness was also linked to a significant increase in sexual partners. This highlights other characteristics of openness, including unconventionality and curiosity, which perhaps explains why these individuals engaged in risky behaviours. Compared to the other personality traits, scores for openness produced only a few significant results with health-behaviour variables. This lack of significant findings for the openness trait is similarly observed in other personality-health studies (e.g., Raynor & Levine, 2009), which suggests that openness may play a minimal role in understanding health behaviours.

4.3 Health-deterring traits

Compared to all other personality traits, neuroticism was the only trait that showed a significant increase in most health-deterring behaviours. Students scoring high on neuroticism reported a significant decrease in healthy breakfast consumption, as well as a significant increase in ecstasy use, corroborating earlier research findings (Dordinejad & Shiran, 2011; Dubey et al., 2010). Indeed, the increase in drug use by such individuals may reflect a greater vulnerability to stress. This characteristic possibly explains why individuals with high neuroticism easily experience negative emotions like sadness and fear and hence, are more likely to use substances to deal with unpleasant situations.

4.4 Limitations

The main limitation of this study was the cross-sectional design in data collection. Although this allowed significant differences to be detected between groups, the results could not be used for inferring how personality traits could be directly causing variations in health behaviours. Another limitation was the lack of representation of the student population, which resulted in a lack of generalisability. Indeed, the sample was relatively homogenous, with participants being primarily females and under 20 years. Furthermore, health-behaviour responses were elicited through self-reported measures which could lead individuals to over- or under-report their behaviours. Finally, data were collected entirely through online questionnaire responses. Although the questionnaire was easily accessible, inexpensive and reached a large number of participants, the lack of personal contact may have led to participants not being entirely truthful in their responses. This highlights the need to be critical when interpreting online responses, due to validity issues.

5 Conclusion

This study has introduced personality-health studies within the Maltese context, with the results obtained extending the well-established body of personality-health literature. Findings suggest that patterns observed between personality and health-behaviour variables were rather similar to those identified in previous studies. Students scoring high in conscientiousness and agreeableness showed a significant increase in health-promoting behaviours, whereas neuroticism scores were linked to health-deterring behaviours. High scores in extraversion and openness highlighted both health-promoting and health-deterring behaviours. In general, conscientiousness and agreeableness served as protective traits against alcohol use, whereas extraversion highlighted a significant increase in alcohol use. Moreover, agreeableness and conscientiousness also served as protective traits against drug use, while neuroticism was associated with a significant increase in ecstasy use. Health-deterring behaviours such as drinking and driving, and unintentional pregnancy were mainly linked to extraversion, although openness showed similar results with an increase in the number of sexual partners.

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8 Conflicts of interest

The authors report no conflicts of interest.

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Short communication

ASSESSMENT OF THE UTILITY OF REPEAT STOOL TESTING FOR *CLOSTRIDIUM DIFFICILE* STOOL TOXIN USING ENZYME IMMUNOASSAY

Karl Galea,[1] Christopher Barbara,[1] Michael A. Borg[2]
[1]Department of Pathology, Mater Dei Hospital, B'Kara, Malta
[2]Department of Infection Control, Mater Dei Hospital, B'Kara, Malta

Abstract. The poor performance of toxin enzyme immunoassay (EIA) for laboratory testing for *Clostridium difficile* (*C. difficile*) infection (CDI) is well acknowledged. Guidelines published in recent years state that testing solely with EIA for detecting toxins A and B is sub-optimal. As a consequence, clinicians may lose confidence in the test and submit multiple samples to offset the poor sensitivity of the toxin EIA. This leads to waste of laboratory resources and is discouraged by recent guidelines. 2,489 requests for toxin EIA submitted during one year at a state general hospital in Malta were reviewed to assess the utility of repeat stool testing for *C. difficile* toxin detection using toxin EIA and also to gather data on the extent of repeat samples within 28 days of a positive test. There were a total of 1,970 diarrhoeal episodes, from which a total of 302 cases (15.3%) submitted more than one sample for repeated testing. Only 2% of these repeats tested positive after having an initial negative result for the *C. difficile* toxin EIA test. Most recent published practice guidelines recommend a two-step or three-step testing algorithm in the diagnosis of *C. difficile*-associated diarrhoea, which offers a marked increase in sensitivity when compared to that of toxin A and B EIA alone. A three-step protocol is proposed which should enable the discernment of the role of *C. difficile* in a diarrhoeal patient.

Keywords: *Clostridium difficile,* repeat stool testing, toxin enzyme immunoassay, algorithm

1 Introduction

Clostridium difficile (*C. difficile*) is a Gram-positive, spore-forming bacterium, spread by the faecal-oral route, which is an important cause of nosocomial diarrhoea in industrialised countries. The bacterium is non-invasive and only toxigenic strains cause disease, due to the production of toxins A and/or B. Carriage occurs in 5-15% of healthy adults, but may be as high as 88.4% in newborns and healthy infants, and up to 57.0% in residents of long-term care facilities (Surawicz et al., 2013).

C. difficile very rarely causes spontaneous disease in healthy young individuals. However, antibiotics may disrupt the normal flora of the gut, leading to *C. difficile* overgrowth and, subsequently, *C. difficile*–associated diarrhoea. The latter can be complicated by pseudomembranous colitis, megacolon, perforation of the colon and possibly death. Clinical disease as a result of *C. difficile* is described as *C. difficile* infection (CDI). The diagnosis of CDI is usually based on the clinical history of the patient in combination with laboratory tests.

For the past 30 years, the two primary reference tests were the *C. difficile* cytotoxin neutralisation assay (CCNA) and the toxigenic culture (TC). These two methods are time-consuming and require specific laboratory facilities as well as technical expertise. As a result, many clinical laboratories have replaced the use of these two methods with the enzyme immunoassay (EIA) technique, which is able to detect both toxins A and B while being less labour-intensive and more cost-effective. In addition, it allows for faster turn-around time from the receipt of the sample to the issuing of results. This test also has high specificity. However, specificity is a test characteristic derived from the proportion of true negative results out of the total number of negative results produced by the test and is not affected by the prevalence of disease in the tested population. Therefore, the high specificity of EIA toxin A/B is offset in settings where CDI is uncommon, resulting in a low positive predictive value (PPV), whereby persons who test positive are less likely to truly have the disease. The sensitivity of the toxin EIA method is 79-80%, due to low reproducibility compared to the cytotoxin assay (She, Durrant & Petti, 2009). These factors undermine the confidence clinicians have in tests for CDI detection, thus prompting them to order multiple samples per patient.

The practice guidelines published by the Society for Healthcare Epidemiology of America (SHEA) in 1995 (cited in Gerding et al., 1995) state that when *C. difficile* is clinically suspected, a single stool specimen should initially be sent for testing. If the result of this first test is negative, one to two additional stool samples should be sent for re-testing. Thus, up to three serial toxin EIA tests increase the diagnostic yield by as much as 8–10% if the initial test is negative (Deshpande et al., 2010). In view of this limited increase in diagnostic yield provided by repeat testing, the 2010 SHEA position paper suggested that repeat testing during the same episode of diarrhoea is of limited value and should be discouraged (Cohen et al., 2010). Tests of cure following a positive *C. difficile* result are not recommended. *C. difficile* toxin positive patients do not need to be retested for *C. difficile* toxin if still symptomatic within a period of 28 days unless symptoms resolve and then recur, pointing towards a need to confirm recurrent CDI (Department of Health and Health Protection Agency, 2008).

In a study carried out in 2008 by Aichinger et al. (2008), of 683 patients who had three or more repeat EIA tests performed within seven days, 605 (88.6%) had only negative results. Twenty patients (2.9%) had a negative result on the first test with subsequent positive results on the following tests. In 12 patients, a positive result was obtained by the second test, in three patients by the third test, in four by the fourth test and in one by the sixth test. The remaining 58 (8.5%)

Correspondence to Karl Galea
(karl.b.galea@gov.mt)
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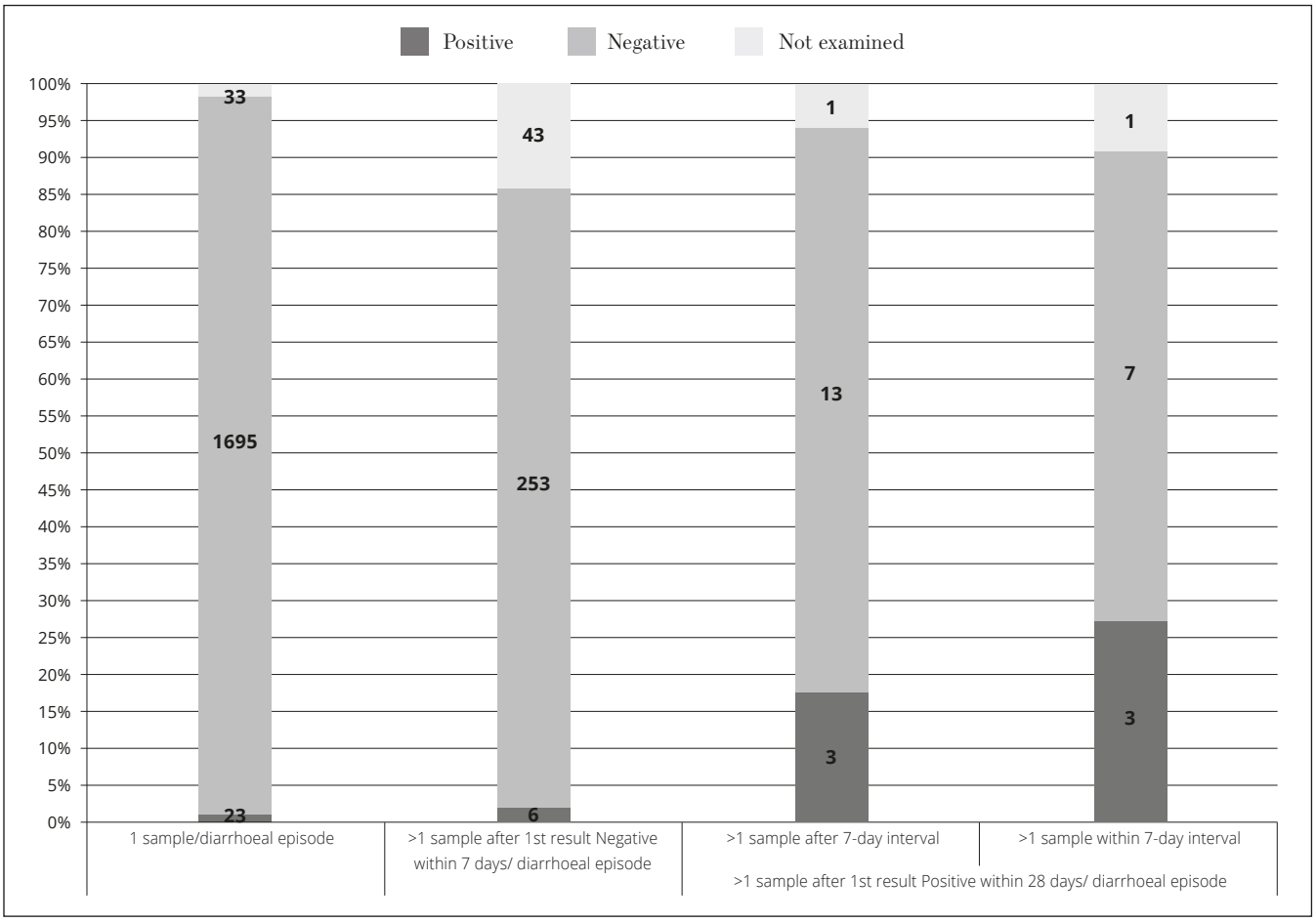

Figure 1. *C. difficile* stool testing outcome grouped according to number of samples sent per diarrhoeal episode

patients had a positive test followed by subsequent positive or negative tests (Aichinger et al., 2008). In a study by Nemat et al. (2009), out of 1165 positive cases, 1046 (89.8%) tested positive in the first test, 95 (8.2%) in the second, and only 24 (2.0%) tested positive in the third test. In the same study, a second test was ordered after an initial negative result in 1,934 cases, of which 95 (4.9%) tested positive, while in 793 episodes, a third test was ordered after two negative samples, of which only 24 (3.0%) resulted in toxin detection. These results highlight the ineffectiveness of repeat testing of stools for *C. difficile* toxin using EIA.

This study was carried out to assess the clinical value of the *C. difficile* toxin EIA technique, by determining the number of cases that had repeat stool tests performed during the same diarrhoeal episode, with a first negative stool toxin EIA result followed by a positive result. The study aimed to ascertain the diagnostic value of these repeat stool tests.

2 Methods

A retrospective study was carried out at the state general hospital in Malta, which is also a teaching hospital that covers the specialties of general medicine and surgery, geriatrics, paediatrics, nephrology, transplant, oncology and critical care. Permission to carry out the study was granted by the Chairman of the hospital's Pathology Department. The readily available and anonymous data was collected via a computer search using the laboratory information system. All requests for *C. difficile* toxin testing between 1st July 2013 and 1st July 2014 were included in the study.

All faecal samples submitted for routine testing for *C. difficile* were processed using RIDASCREEN® Clostridium difficile Toxin A/B (Product C0801; R-Biopharm AG, Darmstadt, Germany). Sequential samples collected during the same diarrhoeal episode and received at the laboratory within seven days of an initial negative test result were considered repeats. In addition, samples submitted for re-testing after an initial positive result within a 28-day time frame were also assessed and included in the study.

3 Results

During the study period (1st July 2013 – 1st July 2014), the laboratory received 2,489 requests for testing stool samples for *C. difficile* toxins. These had been collected from a total of 1,689 patients. Some patients suffered more than one episode of diarrhoea. Upon grouping the requests submitted by patient and by date of submission, a total of 2,053 episodes of diarrhoea were identified. In 1,751 of these diarrhoeal episodes, only one sample was collected, from which 23 (1.3%) samples yielded a positive result (see Figure 1). There were 302 diarrhoeal episodes that were repeatedly tested, which resulted in the submission of 738 samples; the mode of the number of samples submitted per diarrhoeal episode was 2. From these 302 diarrhoeal episodes, only 6 (2.0%) had a negative result on the first test, with subsequent positive

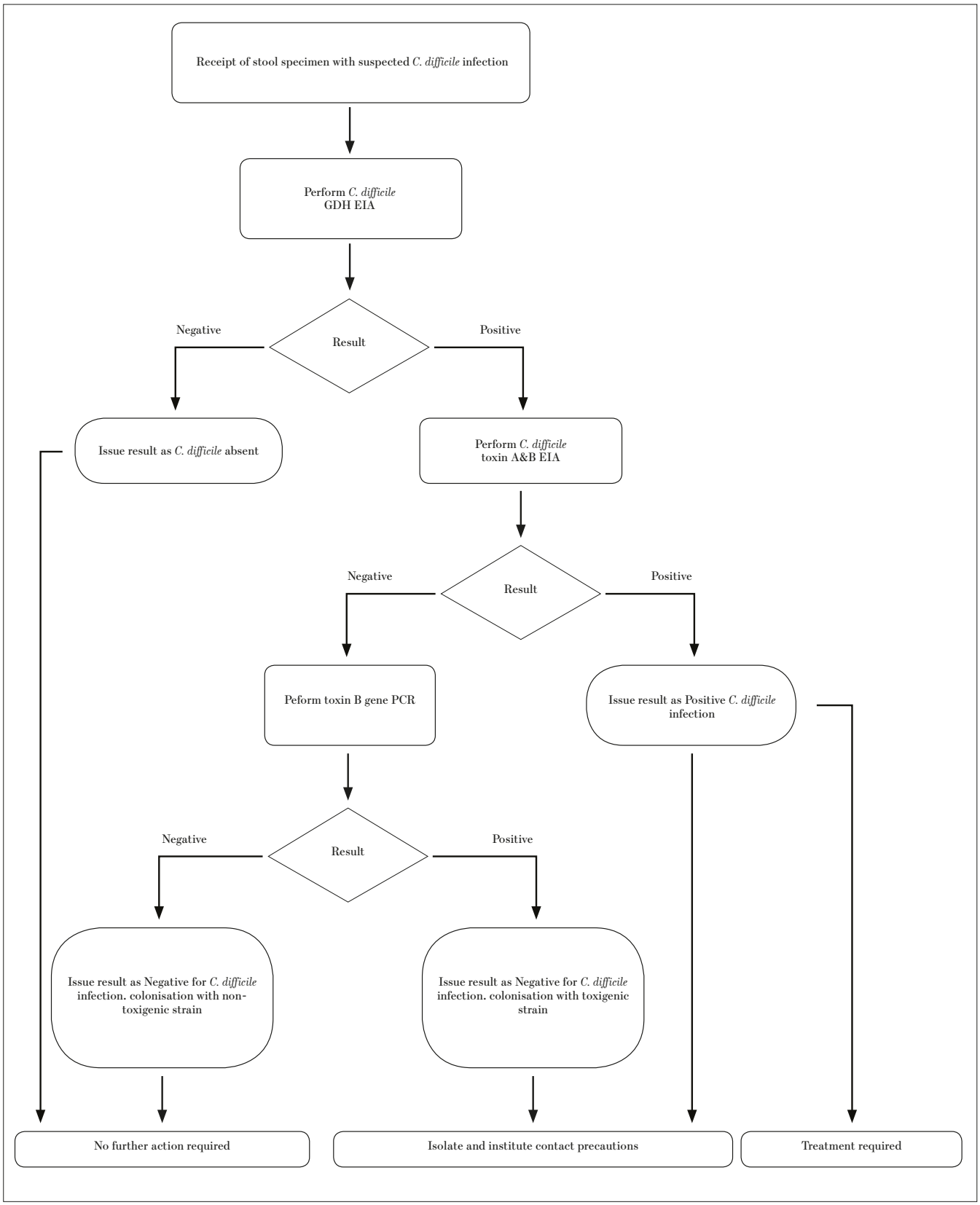

Figure 2. Proposed algorithm to diagnose *C. difficile* infection

results on a following test during the same episode. There were a total of 28 episodes in which the initial test resulted positive and repeat tests for toxin EIA were requested within a period of 28 days after the first positive result. In 11 of these episodes, the repeat test was requested within seven days of the first test.

4 Discussion

Across Europe, a daily average of 109 patients with CDI are undiagnosed due to lack of clinical suspicion or sub-optimal testing. This amounts to more than 39,000 cases which are potentially missed each year (Davies, Davis & Ashwin, 2014). Missed or imprecise diagnosis has implications for infection control practice and patient management.

The diagnosis of CDI is usually based on the clinical history in combination with laboratory tests. Various laboratory tests are currently available for the detection of *C. difficile* or its toxins. The diagnostic tests for *C. difficile* can be divided into (i) tests for *C. difficile* products (glutamate dehydrogenase (GDH), aromatic fatty acids, toxins A and/or B), (ii) culture methods for the detection of toxin-producing *C. difficile* (toxigenic culture), and (iii) tests for *C. difficile* genes polymerase chain reactions (PCR) for 16S ribonucleic acid (RNA), toxin genes, genes for GDH (Crobach et al., 2009).

Practice guidelines for the best testing strategy to diagnose *C. difficile* infection in a clinical laboratory suggest that *C. difficile* toxin EIAs are not suitable as standalone tests for the diagnosis of CDI in an endemic situation, due to the low prevalence rate that gives rise to low positive predictive values of diagnostic tests (Crobach et al., 2009). Various authors recommend a two-step method, whereby an initial highly sensitive and rapid screening test presumably detects all positive cases and is followed up by a second assay that identifies the true positive samples amongst all of the positive results detected during the screening test (Cohen et al., 2010; Crobach et al., 2009; De Silva, 2012; Surawicz et al., 2013).

The enzyme GDH is produced by all *Clostridium* species, including toxigenic and non-toxigenic strains of *C. difficile*, making it a good marker for the presence of *C. difficile* in stools. This is the basis for rapid detection methods using EIA. The negative predictive value (NPV) of the GDH test is comparable among populations having different *C. difficile* prevalence, thus making it a potential candidate for inclusion in a diagnostic algorithm for CDI (Crobach et al., 2009). A negative result would reliably exclude the presence of the organism in faeces. A positive GDH test has a very strong concordance with a positive culture but is not indicative of toxin production (Shetty et al., 2011). Positive GDH EIA requires further confirmatory testing as *C. difficile* infection is a toxin-mediated disease (De Silva, 2012).

Real-time PCR (RT-PCR) is unsuitable as a standalone test, as the lower 95% confidence interval for sensitivity is roughly 80–85% and that for specificity is about 93% (Planche & Wilcox, 2011). In settings of low prevalence, like in an endemic situation where the prevalence of CDI is expected to range between 5 and 10%, the PPV would be unacceptably low (Crobach et al., 2009). Thus, a highly specific test like RT-PCR in settings of low prevalence would lead to a high rate of false positive results. A false positive diagnosis could lead to the management and treatment of non-infected patients alongside true positive cases which, in turn, could potentially result in cross-infection of patients, thereby increasing the prevalence rate. Unnecessary treatment of CDI could be another outcome, potentially including the withdrawal of CDI-inciting antimicrobials that may be required to treat a concurrent infection. In addition, RT-PCR is significantly more expensive than EIA.

Therefore, an updated testing algorithm to conform to the recent guidelines is proposed (Figure 2). The current hospital guideline for the management of CDI would be updated to include guidance on test request submission, sample collection and result interpretation to complement the new testing algorithm, while discouraging repeat testing to ensure an efficient utilisation of laboratory resources. This approach must be accompanied by training to increase awareness of CDI, especially in view of recent insights into CDI epidemiology.

A number of limitations were present in this study. The type of stool sample submitted for testing was not recorded. Ideally, testing should be performed only on liquid specimens, thereby excluding testing on formed stool samples. Unfortunately, the number of tests carried out on formed stool samples is unknown. Moreover, data regarding the number of repeat stool tests performed until a positive test result was obtained following an initial negative result was not recorded. In addition, the time interval between the first negative test result and subsequent positive results was not documented.

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6 Conflicts of Interest

The authors report no conflicts of interest.

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Short communication

VALIDATION OF A POLYMERASE CHAIN REACTION TECHNIQUE FOR KIDD BLOOD GROUP GENOTYPING

Karl Xuereb[1], **Jesmond Debono**[2], **Joseph Borg**[1]
[1]Department of Applied Biomedical Science, Faculty of Health Sciences, University of Malta, Msida, Malta
[2]Blood Bank, Department of Pathology, Mater Dei Hospital, B'Kara, Malta

Abstract. The Kidd blood group antigens, Jk^a and Jk^b, are two of the main surface markers which are found on the membrane of red blood cells. The determination of whether a donor or a recipient has the Jk^a and/or the Jk^b antigens is crucially important to have a successful transfusion without the development of adverse incompatibility-related reactions. In Malta, routine serological-based tests are applied with the purpose of differentiating between homozygous and heterozygous states for the Jk antigens respectively. Although these tests are highly specific and sensitive, there are particular clinical scenarios where haemagglutination assays are not suitable for determining the individual's Kidd blood group status. Additionally, the alternative genotyping procedure has never been applied in Malta within the context of blood grouping. The current study was therefore carried out to determine whether a molecular-based technique such as Polymerase Chain Reaction – Restriction Fragment Length Polymorphism analysis (PCR-RFLP) is a suitable alternative procedure for distinguishing amongst the three different Kidd phenotypes. After extracting deoxyribonucleic acid (DNA) from 50 blood samples obtained from serologically-tested healthy blood donors who expressed at least one of the Kidd antigens, PCR-RFLP analyses were carried out. The results of the latter were then compared with those previously obtained with haemagglutination and a complete match was observed between the two. Therefore, this PCR-RFLP method was confirmed as a suitable alternative laboratory technique that can be used to determine efficiently the Kidd blood group of both donors and recipients, in an accurate manner without subjectivity as encountered in the case of haemagglutination. This research further facilitates the introduction of molecular-based techniques in molecular blood transfusion.

Keywords: Kidd, Jk^a, Jk^b, haemagglutination, PCR-RFLP, molecular transfusion

1 Introduction

Red blood cells have surface markers on their membrane, known as blood group antigens. The three main blood group antigens that are the most clinically significant out of the total 600 are the A and B antigens of the ABO blood group system and the Rh-D antigen of the Rh blood group system. Nonetheless, there are also other blood group antigens which are particularly important for a successful transfusion process. Two of these are the Jk^a and Jk^b antigens of the Kidd blood group system (Daniels & Bromilow, 2010).

The Kidd blood group system (ISBT 009; symbol *Jk*) was discovered in 1951 by Race et al. The antigens of the Kidd blood group system are expressed on the Kidd transmembrane glycoprotein, and the gene which is responsible for the production of the Jk antigens is the *SLC14A1* gene, which in old literature was referred to as *Jk, HUT11* (National Center for Biotechnology Information, 2014). The Jk gene is located on chromosome 18q11-q12 and has 11 exons spanning a total of 30kb (Geitvik et al., 1987).

There are three antigens within the Kidd blood group system: Jk^a, Jk^b and Jk^3. The Jk^a antigen is found in 77% of Caucasians, 92% of Blacks and 73% of Asians. The Jk^b antigen is detected in 74% of Caucasians, 49% of Blacks and 76% of Asians (Dean, 2005). The Jk^3 antigen is present on all the red blood cells which express the Jk^a and/or the Jk^b antigens, thus 99.8% of all the individuals have the Jk^3 antigen, regardless of their ethnicity. In fact, the Jk^3 antigen is only absent in 1% of Polynesians and Finns who do not express either Jk^a or Jk^b on their erythrocytes (Reid & Shine, 2012). Therefore, when it comes to determining the phenotype of the person, testing for the presence of the Jk^a and the Jk^b antigens is performed.

The Jk^a and Jk^b antigens are the products of the Jk-1 and the Jk-2 alleles of the *SLC14A1* gene respectively. When one examines the coding sequences of the mentioned co-dominant alleles, two fundamental differences can be detected; the wildtype Jk-1 allele has an Adenine at nucleotide 588 on exon 7 and a Guanine at nucleotide 838 on exon 9 whereas the mutant Jk-2 allele has a Guanine at nucleotide 588 with an accompanying Adenine at nucleotide 838 (Wester, 2010).

This Kidd blood group polymorphism was described for the first time almost two decades ago (Olivès et al., 1997). The 588A → G single nucleotide polymorphism (SNP) is considered to be silent since both resulting codons code for Proline (Pro) as amino acid 196 of the final gene product (Hong, Gong & Zhour, 2012). Conversely, the 838G → A missense SNP causes the change of amino acid 280 from Asparagine (Asn) to Aspartic acid (Asp) (Intharanut et al., 2013). Ultimately, this Asn280Asp amino acid substitution can be used to differentiate between the Jk^a and the Jk^b antigens (Olivès et al., 1997).

Since the Jk-1 and the Jk-2 alleles can be inherited in a co-dominant fashion, there are three possible phenotypes: homozygous for the Jk^a antigen Jk (a+b-), homozygous for the Jk^b antigen Jk (a-b+) or heterozygous for both antigens Jk (a+b+). In addition, there is also the null phenotype, depicted as Jk (a-b-), which is only expressed by 1% of Polynesians and Finns that lack both Kidd antigens (Irshaid et al., 2002).

In Malta, the three main Kidd phenotypes are differentiated from one another by use of the haemagglutination techniques which are carried out at the state general hospital's Blood Bank. During this serological procedure, a red cell suspension is prepared, which is then

Correspondence to Karl Xuereb
(karl.xuereb.11@um.edu.mt)
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divided into two aliquots and mixed with two anti-sera containing monoclonal IgM anti-Jk^a and anti-Jk^b antibodies. Then, depending on the resultant agglutination pattern, one would determine whether the individual has a homozygous or heterozygous expression of the Kidd blood group antigens. This assay relies on the concept of antigen-antibody reactions, which typically consist of the Red Cell Sensitisation phase and the Agglutination phase, to detect the antigens of interest (Overfield, Dawson & Hamer, 2011).

Haemagglutination is considered as the gold standard technique for Kidd phenotyping due to its high levels of specificity and sensitivity. Other advantages include the simplicity of the technique and its quick and rapid process which ranges from 25 minutes to a maximum of 40 minutes, leading to a fast turnaround time (Reid, 2009). However, like all laboratory techniques, serological haemagglutination has its own limitations. It is a highly subjective test and there is the possibility of having batch-to-batch variations of the anti-sera that are used.

Furthermore, serology-based investigations cannot be applied on blood samples obtained from recently transfused patients, massively transfused patients who had a severe haemorrhagic event and chronically transfused patients who suffer from thalassaemia, sickle cell disease or aplastic anaemias where regular blood transfusions are considered to be part of the treatment regime (Westhoff, 2006). These patients have the donor's red blood cells in their circulation, which would directly interfere with the test since haemagglutination techniques are incapable of differentiating between the patient's and the donor's red blood cells (Castilho & Pellegrino, 2004).

Moreover, individuals who suffer from some form of autoimmune haemolytic anaemias (AIHA) such as Cold Agglutinin Disease, Warm AIHA, Paroxysmal Cold Haemoglobinuria and Drug-Induced AIHA, cannot be typed by haemagglutination. These patients have circulating auto-antibodies which coat their red cells and cause spontaneous agglutination. Consequently, false positive reactions can be observed with haemagglutination-based tests since the observed agglutination would not have occurred due to a reaction between the red blood cells and the anti-sera reagents but due to the reaction that transpired between the erythrocytes and the individual's own auto-antibodies (Westhoff, 2006).

Thus, a number of conditions can result in a higher risk of Kidd blood group status mistyping. This may lead to transfusion with incompatible red cells, therefore increasing the risk of haemolytic transfusion reactions (HTR) (Overfield, Dawson & Hamer, 2011). In order to overcome these limitations, genotyping procedures are being introduced to complement the routine serological techniques.

In Malta, the antigens of the Kidd blood group system have never been genotyped by molecular techniques, due to the fact that they are phenotyped on a routine basis with serological tests. The purpose of this study was therefore to validate Polymerase Chain Reaction – Restriction Fragment Length Polymorphism (PCR-RFLP) analysis, a molecular-based procedure which is specific, sensitive and capable of differentiating between the Jk^a and Jk^b antigens and their respective genotypes in a non-subjective manner.

2 Methods

2.1 Selection of samples

Fifty volunteer, regular, healthy Maltese blood donors from Malta's national blood transfusion centre were recruited for this study. These blood donors were chosen on the premise that they had already been tested by haemagglutination during past donations and thus, their phenotype for the Kidd blood group antigens was known. Donors were selected in such a way as to have the three different Kidd phenotypes represented in the study. Other selection criteria, such as age and gender, were not considered. Once each chosen donor voluntarily underwent the donation process, 9 mL of blood were transferred from the blood donation bag's sample diversion pouch into a 10 mL purple-capped vacutainer containing ethylenediaminetetraacetic acid (EDTA) anticoagulant. Approval for the collection of blood samples was obtained from the University of Malta's Research Ethics Committee.

2.2 Deoxyribonucleic acid (DNA) extraction

The salting out technique (Miller, Dykes & Polesky, 1988) was implemented to extract the deoxyribonucleic acid (DNA) from the 50 whole blood samples. The extracted DNA was dissolved in sterile Tris EDTA buffer (TE) and quantified using the Nanodrop 2000c UV-VIS Spectrophotometer. The samples were diluted to a concentration of 50 ng/μL in a total volume of 20 μL using TE buffer. In addition, a random batch of DNA samples was analysed by 1% agarose gel electrophoresis to re-confirm the quality of the extracted DNA.

2.3 Polymerase Chain Reaction (PCR)

The JK1S forward primer [*5'-TGAGATCTTGGCTTCCTAGG-3'*] and the JK2 reverse primer [*5'-ATTGCAATGCAGGCCAGAGA-3'*] were the two published primer sequences which were used to target the Kidd blood group system gene (Denomme, Rios & Reid, 2000).

The Master Mix that was prepared during the optimised PCR runs is shown in Table 1, whereas Table 2 shows the thermal cycler's profile which was used to carry out the amplification of the Kidd gene. The resultant PCR products were qualitatively checked with 2% agarose gel electrophoresis.

Table 1. PCR components and Master Mix volumes

PCR components	Amount per reaction	Master Mix volumes
2x ReddyMix Master Mix	5 μL	280 μL
JK1S forward primer (50 μM)	0.1 μL	5.6 μL
JK2 reverse primer (50 μM)	0.1 μL	5.6 μL
Genomic DNA (50 ng/μL)	1 μL	/
Sterile Water	3.8 μL	212.8 μL
Final Volume	*10 μL*	*504 μL*

Table 2. PCR Mastercycler® profile for the amplification of the Kidd blood group gene

Number of cycles	PCR step	Temperature	Time
1 cycle	Initial hot start	95°C	5 minutes
28 cycles	Denaturation	95°C	50 seconds
	Annealing	56°C	30 seconds
	Extension	72°C	45 seconds
1 cycle	Final extension	72°C	10 minutes
1 cycle	Incubation period	4°C	∞

2.4 Restriction enzyme digestion

Based on the same published work from which the primers' sequences were obtained, *MnlI* was the restriction enzyme that was chosen to carry out digestion of the PCR products (Denomme, Rios & Reid, 2000). A Master Mix was prepared accordingly, as shown in Table 3, and the

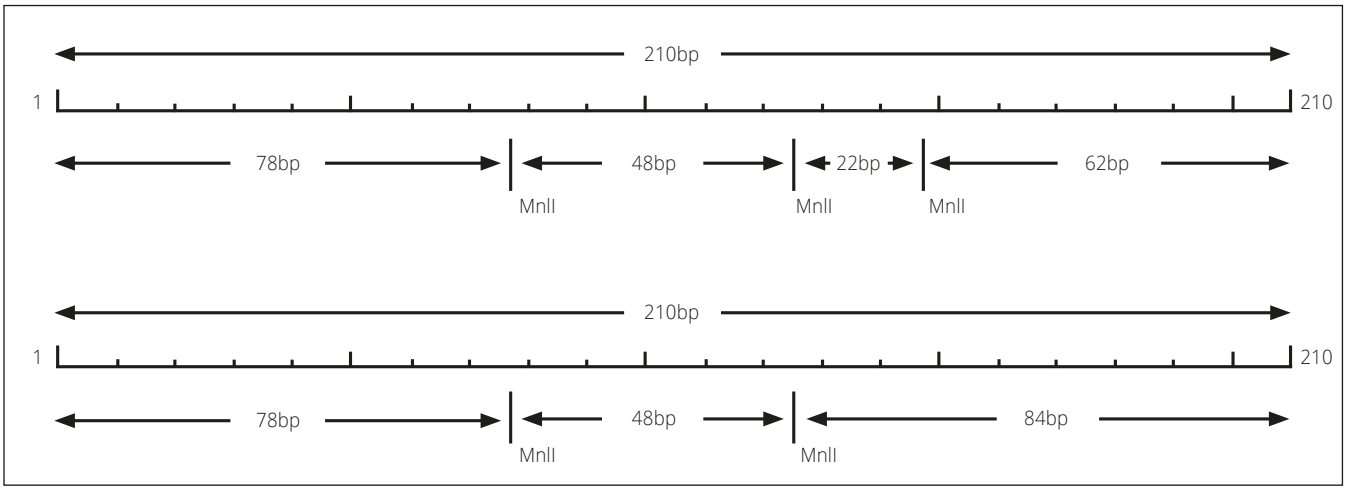

Figure 1. Kidd blood group gene restriction maps; the top image shows a graphical representation of the four DNA fragments obtained from the Jk^a-encoding Jk-1 wildtype allele following digestion by the restriction enzyme *MnlI*. In this case, the enzyme recognised three 5' - CCTC $(N)_7$ ↓ - 3' recognition sites. The bottom image shows a graphical representation of the three DNA fragments obtained from the Jk^b-encoding Jk-2 mutant allele which had two recognition sites, with one of the original three recognition sites being abolished due to the 838G → A missense SNP.

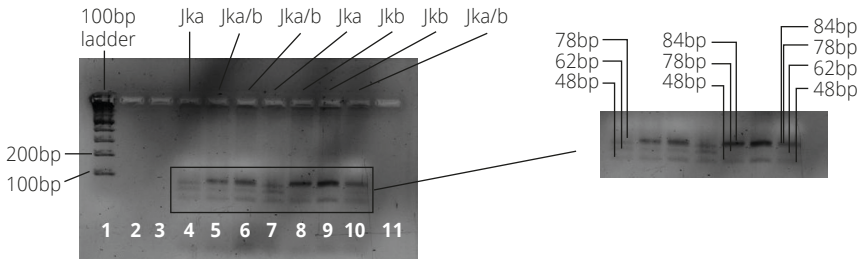

Figure 2. Restriction enzyme digest result; gel image of the *MnlI*-digested Kidd blood group gene when run on 3% Micro ABgarose gel electrophoresis. Lanes 4 and 7 show the homozygous Jk^a genotype, lanes 8 and 9 show the homozygous Jk^b genotype and lanes 5, 6, and 10 show the heterozygous $Jk^{a/b}$ genotype.

PCR products were digested overnight with *MnlI* enzyme (New England Biolabs, Hertfordshire, United Kingdom).

The expected fragment sizes for each genotype were obtained via online analysis with the NEB cutter website (http://nc2.neb.com/NEBcutter2/) and are shown in Table 4 and in Figure 1**.**

Table 3. Restriction enzyme digestion reaction and Master Mix volumes

Digest component	Amount per reaction	Master Mix
NEBuffer 4	2 µL	112 µL
MnlI Enzyme (5,000 U/mL)	0.9 µL	50.4 µL
Sterile water	12.1 µL	677.6 µL
PCR products	5 µL	/

Table 4. The expected sizes of the DNA fragments (in base pairs) of the three Kidd blood group genotypes

Homozygous Jk^a/Jk^a	Homozygous Jk^b/Jk^b	Heterozygous Jk^a/Jk^b
-	84	84
78	78	78
62	-	62
48	48	48
22	-	22

2.5 Final Micro ABgarose gel electrophoresis

The digested PCR products were separated via 3% Micro ABgarose gel electrophoresis and analysed with a UV-transilluminator Bio-Doc-It® Imaging System.

3 Results

On direct comparison, all PCR-RFLP results matched completely with the haemagglutination results, as shown in Table 5.

Table 5. Haemagglutination results compared with PCR-RFLP results

Number of samples	Haemagglutination	PCR-RFLP	Match percentage
11	Jk^a + / Jk^b -	Jk^a	100%
14	Jk^a - / Jk^b +	Jk^b	100%
25	Jk^a + / Jk^b +	$Jk^{a/b}$	100%

Additionally, Figure 2 shows the digested PCR fragments and the three Kidd blood group genotypes.

4 Discussion and Conclusion

This study confirmed that the three different Kidd blood group genotypes, homozygous Jk^a, homozygous Jk^b and heterozygous $Jk^{a/b}$, can be successfully differentiated from one another by PCR-RFLP. This validated PCR-RFLP technique can therefore be applied to blood samples obtained from recently transfused patients, massively transfused patients, chronically transfused patients and individuals who have AIHA, ensuring that these persons who cannot be tested by serology, due to potential interferences from either the donor's transfused red blood cells or their own auto-antibodies respectively, can have their Kidd blood group status genotyped correctly.

Furthermore, given that this study was the first successful undertaking within the field of molecular transfusion, further research is warranted to determine whether this genotyping technique can be used to differentiate the genotypes of other clinically significant blood groups. This is especially relevant in view of the fact that molecular genotyping of the other blood group antigens such as the A, B and O antigens of the ABO blood group system, the C, c, D, E and e antigens of the Rh blood group system, the Fy^a and Fy^b antigens of the Duffy blood group system, the K antigen of the Kell blood group system and the M, N, S and s antigens of the MNS blood group system, amongst others, has never been carried out in Malta.

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7 Conflicts of Interest

The authors report no conflicts of interest.

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