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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 Nursing, Midwifery, Nursing, Occupational Therapy, Physiotherapy, Podiatry and Radiography.

Submitted manuscripts undergo independent blind peer review, typically by two reviewers with relevant expertise. All manuscripts are reviewed as rapidly as possible and an editorial decision is generally reached within approximately two months of submission. Authors of manuscripts that require revisions will have two weeks to submit their revised manuscripts. No manuscript that has already been published or is under consideration for publication elsewhere will be considered.

Types of manuscripts

Review articles will present a substantial survey with an adequate historical perspective of the literature in an area of Health Sciences. The manuscript should not exceed the limit of 5,000 words.

Research papers should be manuscripts comprising complete reports of original, scientifically sound research. They must contribute new knowledge, be prepared for a wide readership and should not exceed 4,000 words.

Short communications should be brief reports of scientifically sound research, but of limited scope e.g. limited amount of data, that contribute new knowledge. They should not exceed 2,000 words.

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Letters to the editor should provide further debate on a topic addressed by a paper published in the Journal. Authors of the manuscript will be sent an edited copy of the letter and they will have the right to reply. They should not exceed 1,000 words.

Book reviews should be timely and objective. They should discuss and assess the main ideas and major objectives of the book and how effectively these are accomplished. Finally, discussions could also be combined. Repetitions between sections should be avoided. Abbreviations and acronyms may be used where appropriate but must always be defined at their first mention.

Discussion This should provide an interpretation of the results without repeating them.

Conclusions The main conclusions of the paper are to be presented in this section.

Acknowledgements (if relevant) Collate acknowledgements in a separate section at the end of the article before the references.

References Authors are responsible for the accuracy of references. All references must appear both in the text and the reference list. References should follow the Harvard referencing system. In the text, the information required is: the surname of the authors; the year of publication;
the page number (direct quotes only).

Commas are used to separate information within parentheses e.g. (author’s name, year of publication). Full stops are always used after the abbreviation p. for page number e.g. p.34 (or pp. for multiple pages). If the citation is at the end of the sentence, it is considered to be part of the sentence, so the full stop is placed after the closing parenthesis.

Examples

Book (1 author)

In text:
Neville (2010) argues that...

Book (2 to 3 authors)

In text:
Bradbury, Boyle and Morse (2002)...

Journal article (printed)

In text:
Trefts and Blacksee (2000) argue that....

Journal article (electronic/online)

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References should be arranged first alphabetically and then further sorted chronologically if necessary. More than one reference from the same author(s) in the same year must be identified by the letters “a”, “b”, “c” etc., placed after the year of publication.

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Number tables consecutively with Arabic numerals (1, 2, 3 etc.), in the order in which they appear in the text. Each table should have a short descriptive title, be self-explanatory and not duplicate data given in the text or figures. Ensure that all columns and rows are properly aligned. Include horizontal rules at the top and bottom of a table and one below the column headings. If a column heading encompasses two or more subheadings, then the main headings and subheadings should be separated by a single short rule. No other rules should be included, neither horizontal nor vertical. Appropriate space should be used to separate columns. Rows should be double-spaced.

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(ii) judge its significance by evaluating the importance of the findings;
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This work was supported by XXX [grant number xxx].

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As we launch Issue 2 of the Malta Journal of Health Sciences (MJHS), I would like to share with you some thoughts on one particular topic which I hold very much at heart, namely children’s language development. In particular, I would like to highlight the importance of social interaction in helping children to develop into effective communicators.

The complex process of language learning is grounded in the child’s social experiences. In the months preceding the emergence of language, babies thrive on the attention, smiles, language input and physical contact of their caregivers. Early communicative exchanges drive them to channel their reflexive actions and vocalisations into purposeful ones. In early language learning, the child depends on social interactions with significant others to be able to forge links between the speech sounds and meanings they receive in their input. Language needs to be experienced naturally, as it is used to identify and describe entities, persons, objects and experiences in daily settings. Interactive experiences help the child to crack the language code and realise the value of language in enabling a wide range of communicative functions. As children’s language capacity expands, communicative exchanges allow them to practise available language skills and develop new ones.

Experiencing the communicative potential of language is price-less for language-learning children. It is not merely a question of exposing the child to the ambient language. While technology may assist the child’s language learning, it is no substitute for one-to-one interactions in encouraging the uptake of language. Strikingly, children are extremely adept at using technological devices and applications, to the extent that they often become engrossed in them and ignore other forms of input. Although such devices can be useful and educational in their own right, they surely cannot make up for the rich language input that may be derived from meaningful one-to-one or group interactions. These not only support language development but also assist the learning of social rules for communication. The sheer beauty and effectiveness of simple, face-to-face exchanges tends to be under-estimated. Communicative exchanges in everyday contexts provide children with the necessary tools for learning language and developing social skills. Through social interactions, infants and toddlers learn the basic components of language and test their newly-acquired skills in real-life settings. Involvement in social exchanges enables older children to learn the rules for social language use and to practise organising multiple thoughts in a coherent sequence prior to verbalising them. Participation in conversations equips them with skills for understanding and using shades of humour and sarcasm, among others. Importantly, they also develop an appreciation of others’ perspectives and emotions, while developing their own. These are but a few of the advantages that may be derived from encouraging children’s involvement in social exchanges.

In today’s day and age, isolation from face-to-face communication has become acceptable. However, we cannot afford to have our children detach themselves from social exchanges. To reap the full benefits of the technological facilities available today, they can be focused on and explored in interactional exchanges. Children can be encouraged to share, describe and discuss their favourite gadgets, programmes and applications. This should go a long way in encouraging them to appreciate the social dimension of language and become truly effective communicators.

Having aired my views on the importance of social engagement for children’s language and communication development, I now invite you to explore the articles in this issue. The diversity of topics should make for interesting and intriguing reading. I trust that these papers will spur us on to engage in further health science research.
Review paper

KEY ENVIRONMENTAL STRESS BIOMARKER CANDIDATES FOR THE OPTIMISATION OF CHEMOTHERAPY TREATMENT OF LEUKAEMIA

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Abstract. The impact of fluctuations of environmental parameters such as oxygen and starvation on the evolution of leukaemia is analysed in the current review. These fluctuations may occur within a specific patient (in different organs) or across patients (individual cases of hypoglycaemia and hyperglycaemia). They can be experienced as stress stimuli by the cancerous population, leading to an alteration of cellular growth kinetics, metabolism and further resistance to chemotherapy. Therefore, it is of high importance to elucidate key mechanisms that affect the evolution of leukaemia under stress. Potential stress response mechanisms are discussed in this review. Moreover, appropriate cell biomarker candidates related to the environmental stress response and/or further resistance to chemotherapy are proposed. Quantification of these biomarkers can enable the combination of macroscopic kinetics with microscopic information, which is specific to individual patients and leads to the construction of detailed mathematical models for the optimisation of chemotherapy. Due to their nature, these models will be more accurate and precise (in comparison to available macroscopic/black box models) in the prediction of responses of individual patients to treatment, as they will incorporate microscopic genetic and/or metabolic information which is patient-specific.

Keywords: leukaemia, environmental stress, hypoxia, autophagy, chemotherapy, cellular biomarkers

1 Introduction

Leukaemia is a severe cancer of the haematopoietic system characterised by the incapability of blood progenitors to mature normally, leading to the accumulation of immature white blood cells, the so-called blasts, in the bone marrow (BM) (Beutler et al., 2001). Alternatively, this disease can be viewed as the formation of an abnormal haematopoietic tissue, the initiation of which is the result of the function of a small amount of Leukaemic Stem Cells (LSCs) (Passegué et al., 2003). Depending on the decrease of the normal blood cell populations, the disease symptoms can consist of fatigue, haemorrhage, infections and fever. The health condition of the patient depends on the amount of normal blood cells compared to the amount of those that are leukaemic. According to Cancer Research U.K. (2014), 8,616 people in the U.K. were diagnosed with leukaemia in 2011, with 4,503 deaths from this disease occurring in 2010. Moreover, 82,300 new cases of leukaemia were diagnosed in 2012 within the European Union. Leukaemia can be divided into different types, depending on the haematopoietic lineage in which the proliferation disorder occurs. More specifically, myeloid leukaemia occurs in the myeloid lineage and lymphocytic leukaemia occurs in the lymphoid lineage. Depending on the speed of evolution of the disease, leukaemia can be divided into acute, whereby the number of blasts increases rapidly, leading to a faster disease evolution, and chronic i.e. the progress of the disease is slower as there is production of partly mature but not fully functional white blood cells. Based on this categorisation, the following four general types of leukaemia can be found: Acute Myeloid Leukaemia (AML), Chronic Myeloid Leukaemia (CML), Acute Lymphocytic Leukaemia (ALL) and Chronic Lymphocytic Leukaemia (CLL).

Acute Myeloid Leukaemia (AML) is one of the most aggressive types of leukaemia. According to Cancer Research U.K. (2014), approximately 2,921 cases of AML occurred in 2011 in the U.K.
AML is a type of leukaemia which is characterised by an accumulation of immature blasts in the myeloid lineage and, as a consequence, insufficient blood cell production. These blasts have low proliferation capability. However, a small population of cells with higher proliferation capacity and self-renewal potential, the LSCs, are the key component for maintenance of the disease (Bonnet & Dick, 1997). AML is usually the result of somatic mutations in either a pluripotent haematopoietic stem cell or in a slightly differentiated progenitor cell, leading to a deregulation and/or inhibition of normal haematopoiesis due to space restrictions and inhibitory and clonal factors specific to the disease (Lowenberg, Downing & Burnett, 1999; Lichtman, 2001; Panoskaltsis, Reid & Knight, 2003).

Chronic Myeloid Leukaemia (CML), alternatively known as Chronic Granulocytic Leukaemia (CGL), is a stem cell disease - of the myeloid lineage - characterised by an exaggerated granulocytosis, anaemia, granulocytic immaturity, basophilia and splenomegaly. There is an extreme cellular accumulation in the BM. From a genetic point of view, 90% of cases show a reciprocal translocation between chromosomes 9 and 22, the so-called Philadelphia (Ph) chromosome translocation. In many cases, the disease can progress into a very high speed phase, resembling AML (Lichtman, 2001). CML is a rather rare cancer of the blood. According to Cancer Research U.K. (2014), 675 patients were diagnosed with CML in 2011 in the U.K.

Acute Lymphoblastic Leukaemia (ALL) (alternatively known as Acute Lymphocytic Leukaemia) is a type of leukaemia which mostly affects children. Approximately 4,000 cases of ALL are reported on an annual basis in the U.S.A., of which two thirds are children (Pui & Evans, 2006). According to Cancer Research U.K. (2014), 654 cases of ALL were diagnosed in 2011 in the U.K. ALL is a leukaemia in which the cellular deregulation occurs in the lymphoid lineage of the haematopoietic system. More specifically, an abnormality in the lymphocytes takes place, leading to an accumulation of non-functional white blood cells (blasts) in the BM as a result of abnormal cellular proliferation, blocking of cellular differentiation and increased resistance to apoptosis i.e. cell death (Pui, 2009).

Chronic Lymphocytic Leukaemia (CLL) is a type of leukaemia which is characterised by an accumulation of immature lymphocytes (of the B-cell lineage) in the human BM, peripheral blood and the lymphoid tissues (Kipps, 2001). The progress of CLL is much slower than that of ALL and it affects older adults, i.e. people over 51 years of age. According to the National Health Service (NHS) U.K., about 2,400 people in the U.K. are diagnosed with CLL on an annual basis and the disease affects 2.7 persons per 100,000 in the U.S.A. (Kipps et al., 2001). According to Cancer Research U.K. (2014), 3,235 people were diagnosed with CLL in the U.K. in 2011.

The most common treatment for all types of leukaemia is chemotherapy (Cancer Research U.K., 2014). A variety of chemotherapy drugs are generally used, depending on the type of leukaemia. The most commonly used drugs for treatment of leukaemia are cytarabine (cytosine arabinose or ara-C), which is an antimetabolite targeting to block the DNA/RNA replication by attacking the cells that are in the S-phase of the cell cycle, and the anthracycline drugs (such as doxorubicin) which attack cells that are in the G1-phase of the cell cycle (American Cancer Society, 2013). Current chemotherapy treatment protocols are designed on the basis of pre-clinical animal experiments and empirical clinical trials, as well as the acquired experience of specialist physicians. The design parameters for these protocols consist of the patient BM aspirate examination (blasts percentage, immunophenotype, cytogenetic and molecular analysis) and physiological patient characteristics (height, weight) for the normalisation of the dose applied on the body surface area (BSA).

2 Towards a Personalised Chemotherapy Treatment: integrating in vivo, in vitro and in silico knowledge

As mentioned in the previous section, traditional clinical diagnosis and further treatment of leukaemia focuses on each patient's clinical symptoms and signs (such as sex and family history) and laboratory imaging evaluation. This process is a reactive approach to the disease, initiating after the disease symptoms appear. Moreover, in the past, drug development by pharmaceutical industries was based on empirical observations. However, nowadays, with significant progress constantly taking place in the areas of genomics, proteomics and metabolomics, it is believed that specific information related to the genetic characteristics and proteomic and metabolomic profile of an individual patient could be used for tailored medical care, or personalised medicine. Some of the challenges in the delivery of personalised medicine lie in (a) the fidelity and validity of current experimental procedures, (b) in vitro, systems used to investigate human disease, (c) the integration of patient-specific and disease-specific datasets i.e. in silico and (d) the application of these models in clinical practice to identify simple targets and more efficient, yet less toxic, therapies for a specific condition, i.e. in vivo. Therefore, 'closing the loop' from in vivo to in vitro and in silico is a first step towards optimisation and consequently, personalisation of chemotherapy treatment. Moving in that direction, an appropriate platform has been developed at the Centre for Process Systems Engineering at Imperial College London, consisting of three main blocks i.e. in vivo-in vitro-in silico which, upon completion, will allow the prediction of optimal drug dosages for each individual, based on specific (personalised) characteristics (Vellion et al., 2014a). The main advantage of this platform is the dynamic interaction of its three robust building blocks. As opposed to existing computational or experimental studies, our platform consists of the interaction of both: data from our in vitro platform are used as an input for the development of mathematical tools. These mathematical tools are further validated with patient data. Figure 1 describes the different parts of the integrated platform that is currently being designed at the Centre for Process Systems Engineering at Imperial College London (Vellion et al., 2014a). Development and optimisation of the in vitro and in silico blocks will eventually bridge the gap between laboratory experimentation, mathematical modelling and in vivo optimal chemotherapy treatment for a specific individual.

A very innovative in vitro tool has been developed and is being used for the ex vivo long-term patient sample cultivation at the Centre for Process Systems Engineering (Mortera-Blanco et al., 2010). This tool is a three-dimensional highly porous polyurethane-based matrix coated with collagen type I, therefore mimicking the porosity as well as the extracellular matrix present in the human BM. This three-dimensional system allows the long-term (up to 6 weeks) cultivation of patient cells in an environment that mimics the in vivo one.

An in silico tool, or mathematical model, which enables the estimation/design of chemotherapy protocols for a specific individual based on the personalisation of the drug schedule has been developed by Pefani et al. (2013). This model consists of a Pharmacokinetic (PK) and a Pharmacodynamic (PD) part. The PK part consists of mass balances of the drug distribution in different body organs such as the heart, the liver and the BM. The PD part calculates the effect of the drug on both normal and abnormal cells in the BM, which is the location of the tumour. The model input is the treatment inflow, which is calculated based on the drug administration route and the injection rate. The latter is a function of body characteristics such as height and weight.
Key environmental stress biomarker candidates for the optimisation of chemotherapy treatment of leukaemia

For cases in which the initial drug dose proposed clinically failed to eliminate the cancerous population, an optimisation process was followed in order to re-determine the optimal drug dose on the basis of clinically relevant constraints. Remarkably, the model output treatments suggest drug dosages similar to those used clinically but, for example, the scheduling is different. The optimised treatment would have much better outcomes with respect to the elimination of the tumour (Pefani et al., 2014). Furthermore, a more detailed cell cycle model based on the work of García-Müñoz et al. (2013, 2014) has been developed, consisting of a multi-stage population balance model (MS-PBM) (Fuentes-Garí et al., 2014). This model is distributed on cell cycle progress-related events i.e. cyclin or DNA expression (different cyclins are produced at different phases of the cell cycle and DNA is produced at the S-phase of the cell cycle). Cell cycle kinetics are tracked not only across different cell cycle phases but also within each phase, thus allowing a more efficient monitoring of the evolution of the leukemic population throughout the cell cycle. The latter is of high importance for chemotherapy optimisation since, as mentioned previously, most chemotherapeutic drugs are cell-cycle specific, targeting cells that are present in specific phases of the cell cycle. This PBM has been successfully validated with experimental data (Fuentes-Garí, 2014).

Moving towards the delivery of optimal chemotherapy protocols for each individual, it is important to understand and further incorporate in the mathematical models the impact of environmental stress. More specifically, the effect of alterations of environmental parameters (oxygen, temperature, nutrients) on the evolution and further response of leukaemia to chemotherapy have to be incorporated (Velliu et al., 2014a, b). Fluctuations of those parameters can take place either across different patients or within the same patient. For example, glucose levels in the blood can differ across a diabetic, a hypoglycaemic and a normal patient. During chemotherapy treatment, it is possible that a patient may have fever; high body temperature can be experienced as a temperature shock by the cancerous population. Oxygen levels can fluctuate in different body compartments e.g. the BM and peripheral blood. The impact of those environmental factors on the leukemic evolution is further discussed in the following section.

2.1 Environmental Stress and Leukaemia

Fluctuations in the micro-environmental conditions of the BM i.e. oxygen concentration, composition and concentration of nutrients such as glucose, cytokines or other growth factors, may be experienced as an environmental stress. As a consequence, these fluctuations can highly affect the normal and abnormal haematopoietic proliferation, metabolic activity as well as drug resistance and further evolution. For example, several researchers have shown that oxidative stress i.e. the increase in the concentration of Reactive Oxygen Species (ROS), leads to activation of survival pathways and is a key factor that promotes progression of cancerous stem cells as well as resistance to chemotherapy (see as examples Adbal Dayem et al., 2010; Fruehaufl & Mayskens, 2013; Liu et al., 2009; Lyu et al., 2008).

Especially in the case of the abnormal haematopoietic situation of leukaemia, alterations of the oxygen and glucose concentration in the different body compartments e.g. in the BM and the peripheral blood or the liver, and, on the other hand, between patients, i.e. individual cases of hypoglycaemia or hyperglycaemia, may lead to a different stress adaptation of the leukaemic population. The latter will most likely affect the cancer growth and inactivation kinetics, as well as the response to a chemotherapeutic drug in vitro. A variety of research studies have revealed the strong correlation between resistance i.e. longer survival and increased proliferation of haematopoietic and/or leukaemic cells and/or resistance to chemotherapy, with (1) oxygen or (2) starvation stress in vitro and/or in vivo. For example, Fecteau et al. (2013) observed an increased in vitro survival of cells from BM aspirates of patients with CLL in 5% O₂ compared to 20% O₂. This increased survival under hypoxic conditions was a result of the Mesenchymal Stromal Cells (MSC’s) increased proliferation and the production of soluble pro-survival factors i.e. CXCL12. Interactions between CLL and (increased) MSCs lead to enhanced CLL resistance.

Wilkinson, Tome and Briehl (2012) state that chronic oxidative stress may contribute to increased resistance of lymphoma patients to chemotherapy. This is due to the fact that oxidative stress leads to alteration of the mitochondria i.e. release of intermembrane proteins, which leads to an increased permeabilisation of the outer mitochondrial membrane and further resistance to apoptosis. Zhou et al. (2010) pointed out the possible relation between the relapse of AML and increased oxidative stress in vivo. Specifically, parameters related to oxidative stress e.g. activities of adenosine deaminase and xanthine oxidase, antioxidant capacity (T-AOC), levels of human thioredoxin (TRX) and indoleamine 2,3-dioxygenase, as well as expression of specific genes related to oxidative stress, were monitored in patients with AML for a time period between a primary and a relapsed status. Low T-AOC and up-regulated TRX expression led to a relapse of the disease, indicating a strong correlation between oxidative stress and AML development and relapse.

Lodi et al. (2011) observed that hypoxia is a key factor that affects metabolic activity i.e. adaptation of phospholytic and glycolytic metabolism, and evolution of KG1a and K562 leukaemic cell lines. Mitochondrial respiration remained unaltered for both cell lines, indicating the ability of these leukaemic cell lines to increase their resistance under oxidative stress.

Giuntoli et al. (2011) studied the effect of the level of glucose on the growth and proliferation of K562 cell lines, U937 cell lines or primary CML cells under hypoxic conditions i.e. 0.1% O₂, as well as under normoxia i.e. 21% O₂. Although in general, slower growth was observed for lower glucose concentrations in hypoxia as well as in normoxia, glucose shortage in hypoxia led to increased size of the leukaemic population compared to the normal haematopoietic one.

Herst et al. (2011) have pointed out a possible relation between the level of glycolytic metabolism of AML blasts and resistance to chemotherapy. Analysis of 26 BM aspirates showed that AML cells with higher glucose consumption were more tolerant to chemotherapy. Analysis of 26 BM aspirates showed that AML cells with higher glucose consumption were more tolerant to chemotherapy.

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All previously mentioned studies point out that there is a very strong relation between oxidative or starvation stress and the leukæmic evolution i.e. growth, kinetics or resistance to chemotherapy. Therefore, the possible oxidative and starvation cellular stress should be systematically followed and/or quantified experimentally (in vitro) and eventually further incorporated in mathematical models (in silico). Efficient monitoring of the level of oxidative and metabolic stress and further adaptation can take place via the selection and further quantification of specific biomarker(s) i.e. intracellular molecules, the expression and/or concentration of which may alter depending on the fluctuation of oxygen and glucose in the system. In order to select ‘stress’ biomarker(s), an in-depth investigation of possible mechanisms of oxidative and starvation stress cellular responses is needed (Vellion et al., 2014a). Hereafter, autophagy, a crucial mechanism which is activated in the absence of nutrients and low oxygen levels, is described and potential biomarker molecules related to the cell stress response are summarised.

2.2 (Macro-)Autophagy: the cellular response to metabolic stress and hypoxia

Autophagy is a cellular mechanism which aims at the maintenance of homeostasis of a normal cell, via degradation of organelles and cellular components by the lysosomes (Banerji & Gibson, 2012; Choi, Ryter & Levine, 2013; Kongara & Karantza, 2012, Levine & Kroemer, 2008; Lozy & Karantza, 2012). Autophagy is activated as a result of exposure to a stress factor and most probably in the absence of nutrients i.e. glucose starvation, as well under hypoxic conditions (Lum et al., 2005; Scherz-Shouval et al., 2007). Degradation of damaged mitochondria as well as aggregation of proteins and other cellular organelles via the autophagic mechanism protect the cells from apoptotic signalling (Jin & White, 2007; Moore, Allen & Sommerfield, 2006). Autophagy may be important in the regulation of cancer development and in the determination of the response of cancer cells to chemotherapy (Degenhardt et al., 2006; Hippest, O’Toole & Thorburn, 2006; Wilkinson, Tone & Briehl, 2012).

Several researchers have shown that autophagy plays a crucial role in maintenance of normal haematopoiesis and function of Haematopoietic Stem Cells (HSCs) (Kundu et al., 2008; Warr et al., 2013). Others have shown that autophagy may lead to an increased resistance to chemotherapy and survival of several cancers, including haematological malignancies. For example, Mortensen, Watchon & Simon (2011) observed that loss of autophagy resulted in loss of normal function of murine HSCs, leading to the expansion of a progenitor cell population in the BM which has as a consequence a severe myeloproliferation. This myeloproliferation strongly resembled human AML, indicating a possible link between maintenance of autophagy and avoidance of malignancies such as AML. Wallington-Beddoe et al. (2011) showed that activation of autophagy (induced by the FTY720 drug) leads to increased survival of ALL cells.

2.3 Biomarker Candidates for the Prediction of the Disease Evolution under Stress

2.3.1 (Autophagic) Biomarker candidates

For ‘switching on’ and maintenance of the autophagic response, a variety of genes are over-expressed and proteins, mainly kinases, are activated and/or de-activated, depending on whether they have a positive or negative regulatory role in autophagy. Therefore, in order to monitor autophagy in an in vitro system, many different biomarkers of genomic and/or protein level can be considered (see Table 1 for an overview).

Table 1. Stress biomarker candidates for leukaemia

<table>
<thead>
<tr>
<th>Biomarker</th>
<th>Genomic</th>
<th>Protein</th>
</tr>
</thead>
<tbody>
<tr>
<td>LKB1-AMPK</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>AMPK</td>
<td>+</td>
<td></td>
</tr>
<tr>
<td>ULKI</td>
<td>+</td>
<td></td>
</tr>
<tr>
<td>P53</td>
<td>+</td>
<td></td>
</tr>
<tr>
<td>PTEN</td>
<td>+</td>
<td></td>
</tr>
<tr>
<td>Atg7</td>
<td>+</td>
<td></td>
</tr>
<tr>
<td>HIF</td>
<td></td>
<td>+</td>
</tr>
<tr>
<td>FOXO3A</td>
<td>+</td>
<td></td>
</tr>
<tr>
<td>FUMH</td>
<td></td>
<td>+</td>
</tr>
</tbody>
</table>

A possible candidate biomarker is the serine threonine kinase ULK1, which is a key initiator of autophagy. Its activation is essential for clearance of cellular mitochondria and ribosomes (Kundu et al., 2008). This kinase is activated under growth factor deprivation and leads to activation of the glycogen synthesis kinase 3 (GSK-3). The latter phosphorylates the acetyltransferase TIP60 which in turn acetylates and activates ULK1 (Lin et al., 2012). Another crucial kinase which is directly related to autophagy is the mTOR kinase which, under normal nutrient concentrations, binds and phosphorylates the ULK1, therefore repressing autophagy. Under nutrient deprivation, the activation of the P53 gene enables activation of the ULK1 and autophagy (Feng et al., 2005). Another possible candidate biomarker is the Atg7 gene, which is an essential gene for activation of autophagy and further regulation of HSC maintenance (Kundu et al., 2008; Mortensen et al., 2011; Mortensen & Simon, 2010). It has been shown that deleting this gene in murine HSC leads to death as a result of an accumulation of mitochondria and ROS, increased proliferation and DNA damage (Mortensen et al., 2011; Mortensen & Simon, 2010). FOXO3A can also be a possible biomarker candidate. It has been found to have a critical role in autophagy induced in mice in a cytokine-free environment (Warr et al., 2013a).

Several researchers have reported a tumour suppressor role of autophagy. More specifically, it has been shown that activation of the AMPK pathway has a suppressor role in AML. AMPK is a protein kinase which regulates protein and energy homeostasis at an intracellular level via autophagic recycling of intracellular components. Practically, AMPK acts as a metabolic sensor of alteration of the intracellular lipid composition and restores energy by maintaining the balance ATP vs AMP, through the LKB1-AMPK activation. LKB1-AMPK is a tumour suppressor in AML (Green et al., 2010). Activation of the P53/PTEN genes and autophagy (Feng et al., 2005), which is a key initiator of autophagy. Its activation is essential for clearance of cellular mitochondria and ribosomes (Kundu et al., 2008). This kinase is activated under growth factor deprivation and leads to activation of the glycogen synthesis kinase 3 (GSK-3). The latter phosphorylates the acetyltransferase TIP60 which in turn acetylates and activates ULK1 (Lin et al., 2012). Another crucial kinase which is directly related to autophagy is the mTOR kinase which, under normal nutrient concentrations, binds and phosphorylates the ULK1, therefore repressing autophagy. Under nutrient deprivation, the activation of the P53 gene enables activation of the ULK1 and autophagy (Feng et al., 2005). Another possible candidate biomarker is the Atg7 gene, which is an essential gene for activation of autophagy and further regulation of HSC maintenance (Kundu et al., 2008; Mortensen et al., 2011; Mortensen & Simon, 2010). FOXO3A can also be a possible biomarker candidate. It has been found to have a critical role in autophagy induced in mice in a cytokine-free environment (Warr et al., 2013a).

2.3.2 (Non-autophagic) Stress biomarker candidates

A biomarker related to oxidative stress is the Hypoxia Induced Factor, HIF, which is the central regulator of oxygen homeostasis. More specifically, HIF1α regulator is overexpressed in many cancer types and HIF proteins mediate cell adaptation to hypoxia (Birner et al., 2000; Talks et al., 2000, Warr & Passagué, 2013b; Zhong et al., 1999).

A possible cell biomarker related to starvation stress is the metabolic enzyme fumarate hydratase (FUMH) which converts fumarate to malate. This enzyme highly controls the intracellular levels of fumarate, with silencing of the expression of FUMH leading to fumarate intracellular accumulation (Ratcliffe, 1999).

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3 Conclusion

The impact of environmental stresses such as oxidative and starvation stress on the leukaemic cell evolution i.e. growth and/or resistance to drugs, has been discussed in depth. It is clear that the effect of these factors on the cancer evolution should be taken into consideration for the accurate prediction of optimal chemotherapy protocols for specific individuals. Furthermore, several cellular components that are related to the cell response to the stress have been analysed. Quantitative information on these key biomarkers could serve as an appropriate input for the construction of more detailed predictive models for the in silico description of the leukaemic evolution. More specifically, from a detection/quantification point of view and as a first step, application of techniques such as next generation sequencing (NGS) or deep sequencing will enable the selection of biomarker application of techniques such as next generation sequencing from a detection/quantification point of view and as a first step, taken into consideration for the accurate prediction of optimal chemotherapy protocols for specific individuals. Furthermore, overall, quantification of appropriate (intra-) cellular biomarkers that are related to the leukaemic in vitro kinetics can enable the combination of macroscopic kinetics with microscopic information, leading to the construction of more detailed models. The (micro) cellular information that these models account for will allow a much more accurate prediction of the cancer response, eventually leading to delivery of personalised chemotherapy protocols for the treatment of patients with leukaemia.

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5 Conflicts of Interest

The authors report no conflicts of interest.

References

Kongara, S. & Karantza, V. (2012) The interplay between au-
Correspondence to: King, 2004). For this reason, research on burnout syndrome is focused on the nursing, physiotherapy and occupational therapy professions, with very few studies within the podiatric profession exploring this concept. The purpose of this paper is to compare the degree of burnout among Maltese podiatrists working in two different settings, namely the public health service and private practice. A quantitative non-experimental study, involving a convenience sample, was conducted. The Copenhagen Burnout Inventory (CBI) was used to evaluate levels of burnout in the two participating groups. No statistical difference was found between the overall mean burnout scores for Maltese podiatrists employed in the public health service and those working in private practice ($p = 0.067$, mean = 48.89 vs 31.84). Although the level of burnout did not differ between podiatrists working within the different settings, podiatrists working in the public sector in general reported higher mean scores for each subscale of the CBI than podiatrists working in private practice. More research is necessary to build a better understanding of burnout among podiatrists in parallel with other health care professionals, in order to help prevent or alleviate this phenomenon.

Keywords: occupational burnout, podiatrists, public health service, private practice, performance, job satisfaction

1 Introduction

The literature suggests that burnout is a serious problem which needs to be addressed appropriately (Akroyd, Caison & Adams, 2002; Akgun, Al-Assaf & Bakan, 2008). Burnout emerged as an important concept in the 1970s and continues to do so today (Schaufeli, Leiter & Maslach, 2008). Burnout is regarded as a multidimensional syndrome which comprises three components, namely emotional exhaustion, depersonalisation and reduced personal accomplishment (Brenninkmeijer & Van Yperen, 2003). Burnout syndrome has never been fully and deliberately understood for health care professionals, their patients and the organisation (Lloyd & King, 2004). For this reason, research on burnout syndrome is increasing, particularly in the health service professions. There has been a continuous search among researchers to develop improved burnout measures to study this concept. These include the Maslach Burnout Inventory (Schaufeli et al., 1996), the Oldenburg Burnout Inventory (Demerouti et al., 2001) and the more recent Copenhagen Burnout Inventory (CBI) (Kristensen et al., 2005). All these instruments embrace the assumption that the burnout concept comprises all the three components discussed above.

Burnout is a distinct kind of occupational stress which generally occurs among health care professionals, mainly due to their continual exposure to patients’ problems, although the point at which burnout begins is unclear (Manely & Tinley, 2004). This in turn results in emotionally drained and burnt out professionals (Akroyd et al., 2002). Burnout is known to result in social and medical problems in the professional, such as dissatisfaction, frustration, anxiety, detachment, psychological fatigue, headaches, sleep disorders, intention to quit the job, depression and substance abuse (Akgun et al., 2008; Schaufeli & Enzmann, 1998). Angerer (2003) reported that this concept, which can lead to cynicism in the individual’s work performance, is correlated with absenteeism, job turnover and low productivity, reduced job satisfaction and decreased commitment to the job. Failing to identify burnout among workers will inevitably lead to considerable financial, social and psychological costs (Maslach & Leiter, 1997; Glasberg, Norber & Soderberg, 2007).

In Malta, the podiatric profession is still relatively new when compared to other local health care professions. Podiatry exists as a clinically independent profession involving the diagnosis and treatment of foot problems. The profession has grown considerably since the patient population is also growing, with 52% to 90% of older people complaining of a foot problem. In Malta, podiatry is providing its services to over 60,000 people annually through the public health service only (The Association of Podiatrists of Malta, 2007). A podiatrist is a qualified health professional who specialises in the prevention, diagnosis, treatment and rehabilitation of disorders of the foot and ankle. Underlying lower limb structures, such as the knee and the upper third of the lower limb, may also be examined by a podiatrist since these structures might be related to a foot or ankle problem. The complex mechanics of the feet (52 bones, a network of muscles, joints and ligaments) and a wide range of foot problems (over 200 identified conditions) demand a separate and autonomous profession which provides high levels of training and professional expertise in the field of podiatry (The Association of Podiatrists of Malta, 2007). As a result, the podiatrist’s scope of practice is diverse and has expanded to include curative and preventive strategies in paediatrics, diabetes, sports injuries, structural and biomechanical problems, geriatrics, vascular and podiatric surgery (The Associ-
Degree of burnout among Maltese podiatrists working in two different settings

2 Methods

A quantitative non-experimental study design was employed. Ethical approval was sought and later granted by the University of Malta Research Ethics Committee. Convenience sampling was adopted. The Register of the Council for the Professions Complementary to Medicine was used to identify potential participants. An information letter was sent to all Maltese podiatrists working either in private practice or within the public health service. The aim of this study was to compare levels of burnout among Maltese podiatrists working in two different settings, namely the private or the public sector only. The nature of work undertaken by each group was fundamentally the same; it was the environment that the profession was practised in which differed between the two groups. The null hypothesis which was formulated for this study suggested no difference in the levels of burnout between Maltese podiatrists working in the public health service and those working in private practice only.

2.1 The Copenhagen Burnout Inventory (CBI)

The CBI is a questionnaire that measures the degree of psychological fatigue experienced in three sub-dimensions of burnout: personal, work-related and patient-related. The personal burnout scale is generic, in that it can be answered by any individual. The work-related burnout scale assumes that the person undertaking the questionnaire has some kind of paid employment, while the patient-related burnout scale assumes that the individual's work involves patients (Kristensen et al., 2005). This inventory is unique since its scales were developed in such a way that they should be able to indicate whether individual feelings of burnout are due to personal or work-related factors (Kristensen et al., 2005). Translations of the CBI into eight languages are available, allowing its use in many studies worldwide. Results support the reliability and validity of the tool, with high internal consistencies ranging from 0.85 to 0.87 (Borritz et al., 2006). The CBI questionnaire consists of 19 items with five response categories for each question: Always, Often, Sometimes, Seldom, Never. For some of the questions, the response categories are adjusted as follows: To a very high degree, To a high degree, Somewhat, To a low degree, To a very low degree. Participants are required to choose only one answer for every question. The response categories are re-scaled to a 0-100 metric (0 = Never/Almost Never/To a very low degree, 25 = Seldom/To a low degree, 50 = Sometimes/Somewhat, 75 = Often/To a high degree, 100 = Always/To a very high degree). If less than three questions are answered from scales 1 and 3, or less than four questions are answered from scale 2, the respondent is classified as a non-responder. A mean rating score greater than 50 indicates a high degree of burnout, whilst a mean rating score smaller than 50 implies a lower degree of burnout. The CBI is relatively easy to complete, requiring approximately 15 minutes to be filled in. It is a public domain questionnaire, unlike other measures of burnout which are protected by copyright and distributed by a commercial publisher, thus making it more accessible to researchers. Permission was obtained from the authors of the CBI (Kristensen et al., 2005) to employ the original English version of the CBI in the present study. Evidence confirming the reliability and validity of the instrument enhanced the study's methodological rigour.

3 Results

Twelve male and six female podiatrists working on a full-time basis either in private practice or in the public health service participated in this study. Participants were aged between 25 and 45 years, with 10 participants aged between 25 and 35 years and eight participants aged between 36 and 45 years. Sixty-one percent of the sample was married. Seventy-two percent (N = 13) of the participants worked within the public health service and 27% (N = 5) worked in private practice. Ninety-five percent of the respondents reported seeing approximately 31 to 40 patients per week.

3.1 CBI Scores

The Kolmogorov-Smirnov test showed collected data to be normally distributed. The one-way analysis of variance (ANOVA) test was used to confirm or reject the null hypothesis which was formulated for this study in conformity with the hypothesis. Results supported the null hypothesis with at least one mean rating score for the CBI differed significantly, thus leading to rejection of the null hypothesis. No statistical difference was found between the overall mean burnout scores of podiatrists working in the public health service and private practice (p = 0.067). Moreover, the overall mean burnout score was less than 50 for each group, implying a low degree of burnout being present among Maltese state-registered podiatrists (mean score for public sector podiatrists = 48.87; mean score for private practice podiatrists = 38.34). Individual mean burnout scores were higher for podiatrists working within the public sector when compared to podiatrists working in the private sector for nearly all items of the three subscales of the CBI, as evidenced by the means of several independent samples to make assumptions on the population means (Bowling, 2002). When the p-value was found to be less than the 0.05 level of significance, it was deduced that the mean rating score for the CBI differed significantly, thus leading to rejection of the null hypothesis.

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Table 1. CBI Scale 1 - Personal Burnout: means and standard deviations for the participants (N = 18).

<table>
<thead>
<tr>
<th></th>
<th>Mean</th>
<th>Std. Deviation</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>How often do you feel tired?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>61.54</td>
<td>24.185</td>
<td>0.769</td>
</tr>
<tr>
<td>Private sector</td>
<td>65.00</td>
<td>13.693</td>
<td></td>
</tr>
<tr>
<td>How often are you physically exhausted?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>55.77</td>
<td>27.298</td>
<td>0.687</td>
</tr>
<tr>
<td>Private sector</td>
<td>50.00</td>
<td>25.000</td>
<td></td>
</tr>
<tr>
<td>How often are you emotionally exhausted?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>57.69</td>
<td>25.789</td>
<td>0.104</td>
</tr>
<tr>
<td>Private sector</td>
<td>35.00</td>
<td>22.361</td>
<td></td>
</tr>
<tr>
<td>How often do you think 'I can't take it any more'?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>48.08</td>
<td>29.689</td>
<td>0.039</td>
</tr>
<tr>
<td>Private sector</td>
<td>15.00</td>
<td>22.361</td>
<td></td>
</tr>
<tr>
<td>How often do you feel worn out?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>57.69</td>
<td>23.683</td>
<td>0.083</td>
</tr>
<tr>
<td>Private sector</td>
<td>35.00</td>
<td>22.361</td>
<td></td>
</tr>
<tr>
<td>How often do you feel weak and susceptible to illness?</td>
<td>50.00</td>
<td>22.822</td>
<td>0.014</td>
</tr>
<tr>
<td>Private sector</td>
<td>20.00</td>
<td>11.180</td>
<td></td>
</tr>
</tbody>
</table>

Table 2. CBI Scale 2 - Work Burnout: means and standard deviations for the participants (N = 18).

<table>
<thead>
<tr>
<th></th>
<th>Mean</th>
<th>Std. Deviation</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Is your work emotionally exhausting?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>51.92</td>
<td>39.319</td>
<td>0.190</td>
</tr>
<tr>
<td>Private sector</td>
<td>30.00</td>
<td>27.386</td>
<td></td>
</tr>
<tr>
<td>Do you feel burnt out because of your work?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>57.69</td>
<td>23.683</td>
<td>0.104</td>
</tr>
<tr>
<td>Private sector</td>
<td>35.00</td>
<td>28.504</td>
<td></td>
</tr>
<tr>
<td>Does your work frustrate you?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>32.69</td>
<td>27.735</td>
<td>0.862</td>
</tr>
<tr>
<td>Private sector</td>
<td>30.00</td>
<td>32.596</td>
<td></td>
</tr>
<tr>
<td>Do you feel worn out at the end of the working day?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>59.62</td>
<td>26.098</td>
<td>0.250</td>
</tr>
<tr>
<td>Private sector</td>
<td>45.00</td>
<td>11.180</td>
<td></td>
</tr>
<tr>
<td>Are you exhausted in the morning at the thought of another day at work?</td>
<td>40.38</td>
<td>29.823</td>
<td>0.057</td>
</tr>
<tr>
<td>Private sector</td>
<td>10.00</td>
<td>22.361</td>
<td></td>
</tr>
<tr>
<td>Do you feel that every working hour is tiring for you?</td>
<td>40.38</td>
<td>21.743</td>
<td>0.074</td>
</tr>
<tr>
<td>Private sector</td>
<td>15.00</td>
<td>33.541</td>
<td></td>
</tr>
<tr>
<td>Do you have enough energy for family and friends during leisure?</td>
<td>67.31</td>
<td>27.735</td>
<td>0.877</td>
</tr>
<tr>
<td>Private sector</td>
<td>65.00</td>
<td>28.504</td>
<td></td>
</tr>
</tbody>
</table>

4 Discussion

The aim of this study was to compare the degree of burnout among Maltese podiatrists working in two different settings (private versus public sector). It is important to note that this study captured the views of all Maltese podiatrists who were working in either the private or public sector only at the time of the investigation (100% response rate).

Identification of burnout using the CBI requires participants to obtain high scores on all three subscales, namely personal burnout, work-related burnout and patient-related burnout (Borritz et al., 2006). No statistical difference between mean burnout scores of podiatrists working in the public health service and private practice was identified. Moreover, the mean burnout scores for each subscale of the CBI were less than 50 for each group, implying a low degree of burnout among Maltese state-registered podiatrists. Previous study results on burnout among podiatrists in other populations are not congruent with the results of this study. Podiatrists in the United Kingdom and Australia were reported to exhibit high levels of emotional exhaustion and depersonalisation, as well as low levels of personal accomplishment when using the Maslach Burnout Inventory and the Work Stress Inventory to measure burnout levels (Mandy, 2000; Mandy & Tinley, 2002, 2004). However, this is the first study to have explored burnout within the Maltese context. Furthermore, the tool used to measure burnout in this specific population of podiatrists is differ-

http://dx.medra.org/10.14614/BURNPOD.2.38 http://www.um.edu.mt/healthsciences/mjhs
ent from the ones employed in the studies mentioned above. It could be argued that the limitations of this study include that all data were collected using a self-report questionnaire, raising the possibility of response set tendencies. Furthermore, the data were collected at one point in time, making it difficult to determine causality. Participants' personality and past or recent traumas/losses were not taken into consideration.

When comparing the overall mean burnout scores between the two groups, podiatrists working in the public sector demonstrated a higher score than those working in the private sector. Research carried out among various health care professionals has led to the conclusion that health care workers within the public sector exhibit a higher degree of burnout when compared to those in private practice (Mrayyan, 2005; Pillay, 2009; Pinto, 2009). Various reasons have been given for this outcome, including that the private sector is less afflicted by rules, regulations and restrictive work, and that motivation in this sector is highly based on profits (Macklin, Smith & Dollard, 2006; Cherniss, 1995).

Consideration of the Personal Burnout CBI subscale demonstrates that podiatrists working within the public sector reported feeling more physically and emotionally exhausted and more worn out than podiatrists working privately. On the Work Burnout subscale, podiatrists employed within the public sector reported being more emotionally exhausted at work, feeling more burnt out as a result of their work, feeling more frustrated with their work, feeling more worn out at the end of a working day and also feeling more exhausted at the thought of another day at work when compared to podiatrists working in the private sector. However, it is interesting to note that despite these negative responses, podiatrists working in the public sector reported more energy for family and friends after work than those working in the private sector. As for the Patient Burnout scale, podiatrists working within the public sector reported that, when working with patients, they found it harder to work, were more frustrated, felt more drained and felt that they gave more than they got back when compared to podiatrists working within the private sector.

The patient-clinician relationship can differ across the public and private sectors. A number of common themes have been identified across studies, explaining why podiatrists working within the public sector tend to be more burnt out than podiatrists working in the private sector. These include lack of professional status among colleagues and patients, as well as labeling by patients as being unproductive, inefficient, distant and poorly motivated (Ferrinho et al., 2004). Long waiting lists are attributed to health care professionals working inside the public sector, leading them to being viewed as incompetent and disrespectful (Forrest et al., 2002). Over-demanding patients, shortage of staff and professional isolation have also been reported to result in professional burnout (Mandy & Tinley, 2004). Moreover, patients attending private services considered their practitioners to be more understanding than practitioners working in the public sector and reported that clinicians had more time available to spend with them (Bjorngaard et al., 2008). Feedback from patients is important for any health care professional to feel appreciated (Mrayyan, 2005). Maslach and Leiter (1997) argue that feedback from patients attending public health care services is either non-existent or almost always negative. Patients attending the public sector do not pay directly for services rendered and thus are more likely to neglect and ignore advice given to them by their clinicians. This in turn leaves health care professionals feeling frustrated, tired and exhausted.

Further research investigating differences in burnout between different podiatric clinical environments in this specific population is warranted if a better service is to be offered to patients attending podiatric services. A better understanding of the burnout concept could be developed through the use of longitudinal research designs that also address off-job experiences in the context of burnout research, particularly addressing indicators of psychological well-being as predictors of work outcomes, in addition to the three CBI subscales (Sonnenstag, 2005). Such studies should enable a better understanding of burnout and why it happens. Results could also provide information on how to cope with it, prevent it or combat it (Schaufeli et al., 2008). Prevention and management of burnout is of utmost importance since it is not just a personal problem but it could also have significant implications for organisations' performance. More motivated podiatrists have a better quality of life, which in turn translates into improved patient care (Mandy & Tinley, 2004).

### Table 3. CBI Scale 3 - Patient Burnout: means and standard deviations for the participants (N = 18).

<table>
<thead>
<tr>
<th></th>
<th>Mean</th>
<th>Std. Deviation</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Do you find it hard to work with patients?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>34.62</td>
<td>26.098</td>
<td>0.134</td>
</tr>
<tr>
<td>Private sector</td>
<td>15.00</td>
<td>13.693</td>
<td></td>
</tr>
<tr>
<td>Do you find it frustrating to work with patients?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>32.69</td>
<td>21.371</td>
<td>0.044</td>
</tr>
<tr>
<td>Private sector</td>
<td>10.00</td>
<td>13.693</td>
<td></td>
</tr>
<tr>
<td>Does it drain your energy to work with patients?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>48.08</td>
<td>29.689</td>
<td>0.256</td>
</tr>
<tr>
<td>Private sector</td>
<td>30.00</td>
<td>27.386</td>
<td></td>
</tr>
<tr>
<td>Do you feel that you give more than you get back when you work with patients?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>61.54</td>
<td>24.185</td>
<td>0.082</td>
</tr>
<tr>
<td>Private sector</td>
<td>40.00</td>
<td>13.693</td>
<td></td>
</tr>
<tr>
<td>Are you tired of working with patients?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>30.77</td>
<td>25.318</td>
<td>0.241</td>
</tr>
<tr>
<td>Private sector</td>
<td>15.00</td>
<td>22.361</td>
<td></td>
</tr>
<tr>
<td>Do you sometimes wonder how long you will be able to work with patients?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Public sector</td>
<td>40.38</td>
<td>33.132</td>
<td>0.794</td>
</tr>
<tr>
<td>Private sector</td>
<td>45.00</td>
<td>32.396</td>
<td></td>
</tr>
</tbody>
</table>

The results of this study have shown no difference in burnout between Maltese podiatrists working in the public and private sectors. However, the podiatrists working in the public sector obtained higher mean burnout scores for most items of the CBI.
sub scales when compared to the practitioners working privately. More research is warranted in this area of study in Malta to contribute towards a better understanding of burnout among podiatrists in parallel with other health care professionals, in order to help prevent or alleviate this phenomenon. Furthermore, results of this study encourage further research with the purpose of establishing the CBI’s validity and reliability in the local context.

6 Acknowledgements

The authors would like to thank all podiatrists who agreed to participate in this study. The authors are also grateful to Marianne Borritz who gave them permission to use the English version of the CBI.

7 Funding

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8 Conflicts of Interest

The authors report no conflicts of interest.

References


Abstract. The purpose of this study was to evaluate the effectiveness of a 10-week Social Communication Group (SCG) programme involving six children diagnosed with Autism Spectrum Disorder (ASD). The sessions were carried out at the mainstream school attended by the children. This study evaluated the effectiveness of the programme in enhancing specific social communication skills in each participant, as opposed to previous research on social skills groups where results were more general. The children’s social communication skills were rated pre- and post-therapy by the first author, parents and Learning Support Assistants (LSAs) to allow comparison. Feedback forms filled in by the LSAs and the first author’s own observations allowed ongoing evaluation of each child’s performance throughout sessions. Questionnaires given pre- and post-therapy to the parents and LSAs were expected to provide evidence of generalisation of social communication skills at home and at school. Qualitative and quantitative data obtained from this small group study indicated that this method of intervention was effective in enhancing the social communication skills of children with ASD. Although all the participants showed an improvement in their overall performance, specific objectives targeted through structured activities that were motivating and repetitive elicited a higher percentage improvement. Factors such as the children’s primary language, pre-therapy status and their understanding at conversational level seemed to have an impact on their individual improvement. Generalisation of skills acquired during therapy was limited since it depended on the commitment of parents and LSAs in implementing the goals of the programme at home and at school.

Keywords Autism Spectrum Disorder, social communication, group therapy, mainstream school, case study

1 Introduction

The development of social communication is necessary for the adequate use of verbal and non-verbal communication for social purposes, across various contexts and with different communication partners (Prizant & Wetherby, 2005). According to the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-V) (American Psychiatric Association, 2013), one of the criteria necessary for diagnosing Autism Spectrum Disorder (ASD) is an impairment in social communication and social interaction, including deficits in social-emotional reciprocity, deficits in non-verbal communicative behaviours used for social interaction and deficits in developing and maintaining relationships. There are two main theories which attempt to explain the social deficits in ASD, namely Theory of Mind and social motivation theory. Previous studies have supported the hypothesis that Theory of Mind is impaired in children with ASD (Baron-Cohen, 2000). Theory of Mind is required for the understanding of the mental states of others, including their beliefs, desires, intentions and emotions (Baron-Cohen, 2001). In other words, children with ASD find it challenging to infer the thoughts of others. An impairment in Theory of Mind would result in difficulties during social communication (Cummings, 2009). In contrast, some theories have suggested that the social communication deficit arises from lack of social motivation rather than an impairment in social cognition. Social motivation models indicate that due to their impairment in early social attention, the social learning experience of children with ASD would be limited during development. Therefore, as a result of their preference for non-social stimuli, the impairment in social skills and social cognitive development would be more significant (Brodkin et al., 2012).

Social skills intervention must be part of the therapeutic programme of children with ASD since it is unlikely that these skills will improve spontaneously (Flood et al., 2010). Furthermore, the impairment in interacting with others limits these children’s development of additional social communication abilities. Intervention targeting social communication should focus on encouraging the acquisition of skills, enhancing performance of skills, eliminating competing negative behaviours and facilitating generalisation (Gresham, Horner & Sugai, 2001). According to Forness et al. (1999), intervention is more effective when it targets specific skills rather than when it is general. Moreover, frequent therapy sessions would produce more positive results (Gresham et al., 2001).

Social communication skills are generally targeted using a behavioural approach which involves modelling, prompting and reinforcement (Baker, 2010). These strategies could be implemented in a Social Communication Group (SCG) programme. Intervention within a small group of individuals having social communication difficulties would give them an opportunity to interact during conversations and games while enabling them to form friendships (Baker, 2010). Generalisation in children with ASD does not occur spontaneously and must be incorporated as part of the therapy programme (Flood et al., 2010).
Research on SCGs for children with ASD has shown inconsistent results due to the diverse presentation of participants as well as the different methods and measurements used across studies (e.g., Barry et al., 2003; Chung et al., 2007; Dittner et al., 2006; Fombonne et al., 2007; Jones et al., 2004). Evidence regarding the effectiveness of SCGs is necessary to prove that resources are being put to good use (Cicchetti et al., 2009). Research should aim to establish an intervention method suitable for individuals with ASD to avoid treatments with additional risks (Offit, 2008).

The purpose of the current study was to investigate the effectiveness of SCGs for children with ASD who attend mainstream schools. A case study approach was adopted, thus enabling a detailed group evaluation with reference to individual characteristics of each child. Apart from identifying the specific communication skills that were enhanced in children after participating in the SCG programme, the study evaluates the factors which might have affected improvement on an individual basis. It also seeks evidence of generalisation of the skills acquired during the sessions to everyday settings.

2 Methods

2.1 Participants

The subjects were recruited from the caseload of speech-language pathologists (SLPs) working in a local Health Centre. The sample consisted of six students who had been diagnosed with ASD and attended a mainstream school, where they had a Learning Support Assistant (LSA). The participants were aged between 8:00 and 9:03 years, with a mean age of 8:06 years. This age range was selected since the chosen SCG programme, designed by Agius (2007), was formulated for children of this age. The six participants are referred to in the present text using a number from 1 to 6, with Child 1 being the youngest child. Although the participant selection criteria did not include gender specifications, the children whose parents consented to their participation in the study were all males.

2.2 Procedure

The main goals of the programme were to enhance basic skills such as attention, listening and greeting, to improve the participants’ conversation skills, to enable them to be aware of different emotions, to enhance their friendship skills and to encourage them to take part in activities requiring teamwork. The programme consisted of i) an initial meeting with the parents during which they were provided with detailed information on the programme ii) 10 group intervention sessions with the six participants and iii) a final meeting with the parents and LSAs during which they were given reports summarising each child’s progress. The intervention sessions were carried out at the participants’ school, which was considered to be a clinical setting for the purposes of this study. Parents and LSAs were advised to attend the sessions, which took place once a week and lasted one hour. Each session targeted a specific set of social skills. Since the children were bilingual, both Maltese and English were used when targeting the SCG goals.

Across sessions, the participants were provided with a series of worksheets, which formed part of the SCG programme. Parents and LSAs were advised to use these at home and at school to encourage generalisation of skills targeted during therapy.

A detailed overview of the programme construction is found in Agius (2007), who describes how the activities for each session were planned following a review of relevant literature and research into different social skills programmes (e.g., Rinaldi, 1992; Arons and Gittens, 1998). Available resources such as handouts were included among the session materials (e.g. Schroeder, 1996; Barratt et al., 2000; Mortimer et al., 2005). Emotions were addressed during every session using The Transporters, a series of short animated films produced by the Autism Research Centre at Cambridge University (2006). Finally, the guidelines of the programme were adapted from Rinaldi’s (1992) Social Use of Language Programme.

2.3 Measures

The effectiveness of the intervention programme was measured using pre- and post-treatment comparison. The programme’s Observation Schedule and Checklist (refer to Appendix 1) was used to measure the children’s performance in various social communication skills. The Checklist content was based on research into the various areas typically targeted during therapy with children with ASD, as well as other social skills programmes and observation schedules (see Agius, 2007). In completing the Checklist, the children’s social communication skills were assigned a ‘pre-rating’ and ‘post-rating’ score before and after therapy. Table 2 exemplifies use of the Checklist rating scale with the conversational skill of ‘Looking (Eye Contact)’ (Section 1).

Baseline and outcome measures were supplemented by field notes on individual participants’ performance on tasks, based on ongoing observation and evaluation. The pre- and post-treatment measures together with the field notes were collected by the first author. In addition, LSAs were requested to complete a feedback form during each session, thus providing another ongoing measure...
Table 2. Rating of ‘Looking (Eye Contact)’ on the Observation Schedule and Checklist.

<table>
<thead>
<tr>
<th>Rating Level</th>
<th>Description</th>
<th>Example</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Not evident</td>
<td>No eye contact</td>
</tr>
<tr>
<td>2</td>
<td>Emerging, but need improvement</td>
<td>Eye contact inconsistent</td>
</tr>
<tr>
<td>3</td>
<td>Appropriate level</td>
<td>Eye contact present even</td>
</tr>
<tr>
<td></td>
<td></td>
<td>during conversations</td>
</tr>
</tbody>
</table>

Since children with ASD find it difficult to generalise skills acquired during therapy to other settings such as home and classroom (Keen & Rodger, 2012), the effectiveness of the programme at home and at school was also considered. The parents and LSAs of the participants were requested to fill in a Questionnaire on their opinion of the child’s communication skills before the therapy programme (refer to Appendix 2). An identical Questionnaire was completed upon completion of intervention. Both the Checklist and the Questionnaire were divided into five main sections evaluating conversation skills, emotions, games, teamwork and friendship skills.

Reliability and validity of measures were not addressed directly in the methodological design due to time restrictions. However, use of the Checklist in previous research (Agius, 2007) showed that it effectively evaluated skills targeted during programme sessions. Moreover, it was explicitly designed to accompany this particular therapy programme. The specificity of measures to the participants reduced the generalisability of results, limiting their external validity (Jewell, 2014). Measurement error in pre- and post-treatment scores was minimised since the same observer assigned all ratings. The Questionnaire items targeted the same skills addressed in the Observation Schedule and Checklist, enhancing the validity of results. However, completion of the Questionnaire by different individuals may have introduced observer bias in the responses.

Data collected were analysed using both quantitative and qualitative methods. Data obtained from the Checklist and Questionnaire was analysed quantitatively using descriptive statistics. The ongoing observations of children’s performance during tasks and the feedback forms completed by the LSAs for every session were analysed qualitatively.

2.4 Ethical considerations

Ethical approval from the University of Malta’s Research Ethics Committee was obtained prior to data collection. The Committee’s ethical guidelines were followed throughout the research study.

3 Results

3.1 Analysis of Performance within the Clinical Setting

Of the 10 sessions making up the SCG programme, each child attended at least eight. Average attendance was 92% of the total number of sessions. The average attendance of relatives (mothers and grandmothers) and LSAs was 66% and 90% respectively. Pre- and post-rating Checklist scores were compared to calculate the percentage improvement in social communication skills for each child and the mean percentage improvement for the group. Skills not requiring further intervention at the pre-therapy stage were identified through the Checklist. Skills which were not evident or were still emerging at the onset of therapy were grouped according to whether they improved or not by the end of the programme. Improvement was accounted for when skill ratings moved up by one or two ranking points. The overall improvement of each child and the mean improvement of all six children was calculated by obtaining a percentage of the total number of enhanced skills in relation to the skills that were not at an appropriate level pre-therapy. This is represented in Figure 1.

Figure 1: Overall percentage improvement of all children compared to the mean.

Figure 2: Mean percentage improvement of the main areas of social communication skills.

The mean percentage improvement of the six participants in conversation skills, emotions, games, teamwork and friendships skills was calculated and represented in Figure 2. A qualitative description of relevant results is provided in the next sections.

3.1.1 Conversation Skills

The main conversation skills targeted during the SCG included greetings, eye contact, turn taking, listening, topic maintenance,
initiation and voice volume.

- **Greetings:** The six children all showed improvement in greeting by saying “hello” and “goodbye” more spontaneously.

- **Eye contact:** None of the children showed adequate eye contact for conversational purposes pre-therapy, but 50% of them improved this skill. Child 4, 5 and 6 improved by maintaining more eye contact. Child 1 was able to maintain eye contact when listening but not when speaking. Child 2 avoided looking at the listener’s face when speaking so that eye contact was not evident whereas Child 3 made eye contact when greeting but not during conversation.

- **Turn taking:** Five of the children did not show appropriate turn taking skills pre-therapy and only Child 4 did not show improvement since he did not wait for his turn to speak during conversation.

- **Listening:** Five of the children presented with difficulties in listening attentively during conversation since they interrupted the speaker or appeared to be distracted. Child 4 was the only child who did not show improvement in active listening due to his tendency to vocalise when someone else was speaking.

- **Topic maintenance:** Improvement in topic maintenance was observed in all the children since they were able to speak for a longer time about a particular subject.

- **Initiation:** The six children presented with difficulties in initiating a conversation. Five of them showed improvement by introducing a topic or by asking questions.

- **Volume:** Of the four children who presented with an inappropriate volume level, three showed improvement. Child 2 increased the volume level of his voice while Child 4 and 6 decreased their loudness. Child 5 did not use adequate volume since it was either too high or too low.

### 3.1.2 Emotions

The emotions targeted were happy, sad, angry, afraid, excited, worried, surprised, disappointed, tired, unfriendly, sorry, kind and helpful. Five of the participants had presented with difficulties in explaining and/or recognising emotions in themselves pre-therapy. They all showed improvement in this area. In addition, all six children presented with difficulties in explaining and/or recognising emotions in others. Only Child 2 did not show any evident improvement in the latter. By the end of the programme, 50% of the children, including Child 1, 3 and 5, also exhibited an understanding of complex emotions.

### 3.1.3 Games

Games included in the SCG focused on enhancing participation, following and accepting rules, attending as well as showing appropriate behaviour when winning or losing. Five of the children presented with difficulties in participation during games and an improvement was observed in that they became more attentive. This was evidenced by an increase in their listening and/or looking behaviours. All of the children showed an improvement in collaboration during games, which was observed through turn taking, understanding and/or following instructions and accepting and/or following rules. Although all the children behaved appropriately when playing a game, four of them had difficulties when losing. Three of the latter showed improvement in behaving appropriately in this area.

### 3.1.4 Teamwork

Teamwork activities included in the SCG focused on encouraging collaboration, sharing, commenting and giving assistance. Although the children were encouraged to work together, two of them preferred working alone throughout all the sessions. Improvement was noted in the skills listed below.

- **Collaboration:** The children who tended to take a leadership role improved by allowing others to contribute, while those who preferred working alone improved by joining the rest of the group. Child 2 showed progress by simply participating, given that in earlier sessions he refused to take part in teamwork activities. Child 6 was the only participant who did not demonstrate any difference in collaboration pre- and post-therapy since he preferred working individually.

- **Sharing materials:** Four of the children were not observed to share materials pre-therapy. Three of them improved by sharing the materials used during the crafts activities with the members of the team. This was not observed in Child 2.

- **Commenting:** Discussion of ideas was not evident in the initial session. Four of the children improved since they started discussing ideas with the group while the others just followed the rest of the team.

- **Giving assistance:** Only one of the children was observed helping others during the initial session. However, by the final session, three other children started giving assistance by helping others find pictures, helping them to finish their puzzle or preparing for the activities.

- **Leadership:** Although not targeted directly, three out of the six children improved in leadership skills by accepting leadership, speaking for the team and accepting ideas of others.

### 3.1.5 Friendship skills

Friendships skills targeted through role play and brainstorming included asking another child to play, asking a question, introducing oneself, controlling anger, practising independence and solving problems. Improvement observed in friendship skills is described below.

- **Welcoming others:** The participants became more welcoming towards their peers as the sessions progressed, except for Child 5. This trend was evident as participants accepted peers joining them when asked to play with them. Willingness to work with others was also shown during teamwork activities. Participants also started showing a preference for particular peers when choosing teams.

- **Introducing self:** All children improved in introducing themselves by using greetings, saying their name and their age, and mentioning their hobbies.

- **Reciprocity:** Four of the participants took part in more reciprocal conversations. However, this improvement was not observed in Child 1, who continued talking in a monologic style most of the time, and in Child 2, who did not take part in conversations.

- **Controlling Anger:** Child 4, 5 and 6 became more able to control their anger when waiting for their turn during games and when disagreeing with a peer. Child 1 and 2, who also presented with difficulties in controlling their anger, did not show any improvement in this regard.

- **Independence:** Child 1, 3 and 5 became more independent when making decisions while the other participants did not show any improvement in this area.

- **Solving problems:** Although all of the children presented with difficulties in solving problems during social situations, only Child 1 and 3 showed progress in this skill.

- **Making friends:** All the children improved in making friends except for Child 2 who, even though frequently encouraged, was rarely seen interacting with the others.

### 3.2 Evaluation of Performance at Home and at School

The pre-therapy and post-therapy Questionnaires given to parents and LSAs were compared. Figures 3 and 4 show percentage change in rating of skills according to whether parents and LSAs reported improvement, no change or regression. The mean percentage improvement of skills at home and at school was 20% and 19% respectively. Most of the skills remained the same after therapy, both at home and at school. According to the Ques-
4 Discussion

The purpose of this study was to evaluate improvement in the social communication skills of six children with ASD after attending a 10-week SCG programme in a mainstream school. Based on observations within the clinical setting and parents’ and LSAs’ ratings, improvement was evident in the social communication of all six children taking part in the study. However, a 100% improvement was not shown by any of the children, implying that further therapy on social communication was required. Social interaction and pragmatic skills are influenced by cultural factors (Bratanic, 2007; Harzing & Neyer, 2008). It should therefore be taken into consideration that difficulties in social communication might also be observed in typically-developing school-aged children, as in the inability to wait for a turn. In addition, personality has an impact on reaction to social situations even in typically-developing children (Mischel & Shoda, 2008), as in acceptance or otherwise of losing. These variables might therefore account, at least in part, for lack of improvement in the skills targeted by the programme. However, it could not be established whether limited progress was specific to children with ASD since a control group of typically-developing children was not included in the study.

Attendance did not seem to affect the children’s individual improvement. However, it might have had an impact if they missed more than 80% of the sessions. More improvement was shown in children who a) were English-speaking, b) had more age-appropriate skills before therapy and c) had adequate understanding at conversational level. English-speaking children possibly showed most improvement since they tended to participate and lead more in activities, resulting in more use of English during sessions which in turn encouraged their participation further. Nevertheless, this improvement could be a result of their increased willingness to participate, which led to more opportunities for practising their skills. Another factor which might have had an impact on this result is the fact that the DVDs targeting emotions were in English. Although Maltese was also used during the sessions, the use of English in the previously mentioned task might have been a limitation for the Maltese-speaking children. Those children having a more positive pre-therapy status showed increased improvement since they presented with fewer skills requiring intervention. Another possible reason is that they had more skills which were already emerging pre-therapy, rather than skills which were not evident. Therefore, they required intervention on improvement of skills as opposed to others who necessitated intervention on acquisition. Finally, the comprehension difficulties exhibited by Child 2, 4, 5 and 6 seemed to have affected performance during tasks requiring conversational understanding, namely free conversation with peers, show-and-tell and teamwork activities. Improvement in each of the main skills addressed by the SCG programme is discussed in the following sections.

4.1 Conversation skills

The results obtained for conversation skills correspond to those reported in the study by Barry et al. (2003), where improvement was mostly shown in greetings rather than in other conversation skills. However, Barry et al. (2003) did not evaluate improvement in specific conversation skills such as eye contact, turn taking, listening, topic maintenance, initiation and voice volume. In Cragar and Horvath’s (2003) case study of a 10-year-old boy attending group therapy, improvement in conversations was evident in that the child interrupted the speaker less. Yet, lack of progress in the child’s understanding of non-literal language and humour was reported, similar to findings emerging from the Checklist and Questionnaire data in the current investigation. Several studies have reported an improvement in the initiation of social interactions as a result of group therapy (e.g. Bauminger, 2002; Crooke, Hendrix & Rachman, 2008; Kroeger, Newsome & Schultz, 2007). In this study, only Child 2 did not improve in initiating conversations. At the start of the programme, this child did not participate in interactions, so that for him, progress entailed maintaining a conversation rather than initiating it. In the four case studies evaluated by Chung et al. (2007), all children progressed when making initiating comments. However, in contrast to the current study, Chung et al. (2007) also involved typically-developing peers who modelled adequate use of social skills to the participant group.

4.1.1 Emotions

The children showed more improvement in recognising and explaining emotions as experienced by themselves rather than by others. This ties in with the proposed difficulties in Theory of Mind (Baron-Cohen, 2000), which prevents children with ASD from understanding the perspective of others, including their feelings. Improvement in the understanding of complex emotions was only observed in three of the children. This was expected since studies such as Balconi and Carrera’s (2007) reported that chil-
dren with ASD had the most difficulty in recognising complex emotions. Baron-Cohen et al. (2007) found that difficulties in recognising complex emotions might persist through adulthood. However, there is evidence that intervention may encourage the development of these emotions. In fact, Baumering’s (2002) study on the effectiveness of cognitive behavioural intervention for children with ASD in a group setting reported improvement in the production of complex emotions.

4.2 Games

The highest percentage improvement in skills was observed in games. This was probably due to the fact that games were more structured when compared to the tasks used to target the other objectives. In addition, the prospect of winning acted as a motivator, which reinforced the children’s participation and collaboration during games. This aspect of reinforcement forms part of the behavioural approach which encourages learning in children with ASD (Baker, 2010). There are very few studies that have evaluated the effectiveness of games as part of a SCG programme. Barry et al. (2003) found that children showed more improvement in play than in the other areas of intervention, suggesting that the children improved in their ability to play with others. Targeting social skills through games, Jones et al. (2004) observed improvement in all the skills addressed, although progress in the children’s actual performance during games was not evaluated.

4.3 Teamwork

There was a general improvement in collaboration, sharing materials, commenting, giving assistance and leadership. This corroborates the findings of Jones et al.’s (2004) study, in which participants were reported to start offering help and encouragement to others. An improvement in the sharing of ideas was observed in four of the children in the current study. Improvement in this area was similarly reported by Jones et al. (2004). Leadership skills were observed in the children who were more willing to collaborate in the earlier sessions of the programme. Dittner et al. (2006) found no significant improvement in leadership skills in their investigation. However, the scales they used to assess performance might not have been sufficiently valid since they included general social skills and did not evaluate the abilities targeted during sessions.

4.4 Friendship skills

Friendship skills exhibited the least improvement, possibly because the relevant intervention was less structured and more unpredictable, excluding the friendship skills targeted during brainstorming activities and role play. The lack of structure in intervention was due to the fact that it depended on situations presenting themselves during the sessions, which would differ from one group to another. Five of the participants improved in making friends, but Child 2 failed to interact with others through to the final session. In Fombonne et al.’s (2007) study, 12 out of the 13 adolescents with Asperger’s Syndrome and High-Functioning Autism who gave feedback on their ability to make friends reported an improvement. However, this outcome was based on self-report rather than the researchers’ observations, suggesting that the participants’ subjectivity in reporting may have influenced results. In addition, the prospect of winning acted as a motivator, which reinforced the children’s participation and collaboration during games. This aspect of reinforcement forms part of the behavioural approach which encourages learning in children with ASD (Baker, 2010). There are very few studies that have evaluated the effectiveness of games as part of a SCG programme. Barry et al. (2003) found that children showed more improvement in play than in the other areas of intervention, suggesting that the children improved in their ability to play with others. Targeting social skills through games, Jones et al. (2004) observed improvement in all the skills addressed, although progress in the children’s actual performance during games was not evaluated.

4.5 Generalisation

The limited evidence of generalisation could have resulted from the lack of implementation of worksheet activities at home or at school. Although parents and LSAs were encouraged to use these worksheets, it was reported that they did not have time to implement them. Most children with ASD are not able to generalise skills outside the clinical setting without additional support, suggesting that this aspect must form an integral part of any SCG programme (Flood et al., 2010). Previous studies have also reported limited generalisation skills. For example, Barry et al. (2003) found greetings to be generalised as opposed to conversation and play skills. Generalisation was reported by the parents taking part in Fombonne et al.’s (2007) study, but no evidence of generalisation was present in the investigation carried out by Jones et al. (2004). A decrease in aggression, increased self-control and improved interaction were reported by Dittner et al. (2006). In the latter study, however, parents participated directly during sessions and the measurement scales were based on their reports. Cragar and Horvath (2003) reported an increase in generalisation after a nine-month follow up. When comparing pre- and post-therapy Questionnaires completed by the parents and LSAs, regression was observed in specific skills which differed across participants. This outcome could have been due to a change in the parents’ and LSAs’ perceptions of the children’s skills. In turn, the latter could have resulted from a recent incident involving the child, such as difficulties with conflict resolution emerging during an argument with a peer. Regression might have also been an outcome of parents’ and LSAs’ increased awareness of social communication deficits: upon completion of the programme, they were likely to be more knowledgeable in the requirements of each skill. Another possibility could be that parents and LSAs compared their child with other participants in the group and subsequently rated his skills as less appropriate. In addition, the pre-therapy Questionnaire was completed at the beginning of the scholastic year, when LSAs might have not yet had sufficient experience with the children to judge whether specific skills were at an appropriate level pre-therapy. Furthermore, reporting bias in parents and LSAs may have resulted in lack of objectivity in the generalisation data. The latter was manifested in discrepancies in the ratings of individual children’s behaviours by the two observers.

4.6 Limitations

The findings of this study cannot be generalised to the broader population of children with ASD due to the small sample size. The data were obtained from a case study of a single group, so that the results are not necessarily applicable to other Maltese children attending a different mainstream school. Regrettably, the study lacked a control group consisting of children not receiving group therapy. This would have allowed comparison with the experimental group, providing more objective results regarding the effectiveness of the SCG. However, the ethical issue of limiting therapy to the experimental group led to exclusion of a control group from the study’s methodological design. Nonetheless, improvements in social communication do not usually occur spontaneously (Flood et al., 2010), which implies that unless the participants were attending additional therapy, improvement shown could be attributed to the SCG. Indeed, at the beginning of the programme, none of the parents reported that their children were receiving intervention targeting their social skills. It is also acknowledged that the Checklist and Questionnaire measures depended on the perspective of the individuals completing these instruments. Yet, subjectivity should have been minimised since forms were completed by the same individuals pre- and post-
therapy. Ensuring the validity and reliability of the Checklist and Questionnaire data would have increased the objectivity and accuracy of results. In addition, skills emerging during the group sessions might have been restricted by the structured nature of the clinical setting, limiting the representativeness of the data.

5 Conclusion

The results of this study support the claims in the literature that SCGs are effective in enhancing the social communication skills of verbal children with ASD. The percentage improvement resulting in the main skills targeted highlights the importance of structured activities, motivation through rewards and repetition of tasks for effectiveness in the intervention of children with ASD. Although differences in children’s primary language, pre-therapy status and understanding at conversational level had an impact on their individual improvement, further investigation is required regarding the effects of these specific factors. Results showed limited evidence of generalisation, but the parents and LSAs still reported an improvement in each child’s specific deficits. These positive results should encourage the implementation of SCGs in mainstream schools, although further research on their effectiveness with a more representative sample is recommended.

6 Acknowledgements

We would like to thank the participants, their families and their LSAs for taking part in the study.

7 Funding

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8 Conflicts of interest

The authors report no conflicts of interest.

References


Appendix 1

SOCIAL COMMUNICATION GROUPS
OBSERVATION SCHEDULE AND CHECKLIST

Name: [ ]
Group: [ ]
Number of sessions attended: [ ]

Date Pre-Rating: [ ]
Date Post-Rating: [ ]

Rating
1 = Not evident
2 = Emerging but needs improvement
3 = Appropriate

Note: Positive changes in post-rating scores are in bold

<table>
<thead>
<tr>
<th>Skill</th>
<th>Pre-Rating</th>
<th>Post-Rating</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Conversation:</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Initiation</td>
<td>Greeting</td>
<td></td>
</tr>
<tr>
<td>Looking (eye contact)</td>
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</tr>
<tr>
<td>Introducing a topic</td>
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<tr>
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<tr>
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<td>Attention</td>
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<tr>
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<tr>
<td>Turn taking</td>
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<tr>
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<td>Flexibility of topic</td>
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<tr>
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<td>Closure</td>
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<tr>
<td>Understanding humour</td>
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1
The effectiveness of social communication groups in mainstream schools

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<td>Tone of voice</td>
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<td>In others</td>
<td>In self</td>
<td></td>
</tr>
<tr>
<td>Explaining emotions</td>
<td>In self</td>
<td></td>
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<tr>
<td>In others</td>
<td>In others</td>
<td></td>
</tr>
<tr>
<td>Showing emotions</td>
<td>Expressing verbally</td>
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<td>Appropriateness</td>
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<tr>
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<td>3. Games:</td>
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<tr>
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<td>Attention</td>
<td></td>
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<tr>
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<tr>
<td>Looking</td>
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<tr>
<td>Collaboration</td>
<td>Turn taking</td>
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<tr>
<td>Understanding instructions</td>
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<td>Following rules</td>
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<td>Losing</td>
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<td>Explaining ideas</td>
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<td>Coping with being interrupted</td>
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<td>Choosing friends with similar interests</td>
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<td>Adapting to character of others</td>
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<td>Assertiveness</td>
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<td>Ability to say ‘no’</td>
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<td>Conflict resolution</td>
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<td>Accepting mistakes of others</td>
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<td>Coping with change</td>
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<td>Avoiding provoking others</td>
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<td>Recognising boredom</td>
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<td>Inhibiting offensive comments</td>
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<td>Ending</td>
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<td>Saying goodbye</td>
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Appendix 2

Sample questions of Parents’ and LSAs’ Social Communication Questionnaire

Section A: Communication

1. Initiating Conversation

1A. Does the child use greetings to initiate conversation such as says ‘hello’?
- Yes, most of the time
- Yes, but not always
- Never

1B. Does the child maintain eye contact when speaking with others?
- Yes, most of the time
- Yes, but not always
- Never

1C. Does the child manage to initiate a conversation by introducing a new topic?
- Yes, most of the time
- Yes, but not always
- Never

1D. Does the child initiate conversation by asking questions?
- Yes, most of the time
- Yes, but not always
- Never

2. Maintaining a conversation

2A. Is the child attentive to the speaker during a conversation?
- Yes, most of the time
- Yes, but not always
- Never

2B. Is the child an active listener thus being able to give feedback to the speaker by either non-verbal or verbal conversation?
- Yes, most of the time
- Yes, but not always
- Never
2C. Is the child able to wait for his turn during a conversation?

☐ Yes, most of the time
☐ Yes, but not always
☐ Never

2D. Is the child able to maintain speaking about a particular topic for some time even if the topic is chosen by another person?

☐ Yes, most of the time
☐ Yes, but not always
☐ Never

2E. Is the child able to change the topic during the same conversation?

☐ Yes, most of the time
☐ Yes, but not always
☐ Never

2F. Is the child able to be part of a conversation when the topic is not chosen by him?

☐ Yes, most of the time
☐ Yes, but not always
☐ Never

2G. Does the child ask for clarification when he does not understand what the speaker is saying?

☐ Yes, most of the time
☐ Yes, but not always
☐ Never

2H. Does the child respond appropriately during a conversation?

☐ Yes, most of the time
☐ Yes, but not always
☐ Never

2I. Is the child able to agree with the speaker?

☐ Yes, most of the time
☐ Yes, but not always
☐ Never
2J. Is the child able to disagree with the speaker?
☐ Yes, most of the time
☐ Yes, but not always
☐ Never

2K. Does the child show interest by asking questions?
☐ Yes, most of the time
☐ Yes, but not always
☐ Never

2L. Does the child comment appropriately?
☐ Yes, most of the time
☐ Yes, but not always
☐ Never
THE LIVED EXPERIENCE OF PERSONS WITH AN AMPUTATION

Chanelle Grech, Roberta Farrugia Debono
Department of Psychology, Faculty for Social Wellbeing, University of Malta, Msida, Malta

Abstract. This paper presents an exploratory study of Maltese persons living with an amputation. Semi-structured interviews were conducted with four participants. Three of the participants had a lower limb amputation, while the fourth had an upper limb amputation. Interpretative phenomenological analysis (IPA) was used in the analysis phase. The three core superordinate themes that emerged from the data revolved around the emotional reactions towards the amputation, the numerous changes it brought to the participants’ lives, and the support which enabled amputees to cope with their situation. The study highlights the range of emotions experienced by the participants on having an amputation and the changes in their lifestyle. Support from family, friends, health professionals and other amputees was identified as a factor that helped participants cope and adapt to their new condition.

Keywords: amputation, lived experience, prosthesis, social support, qualitative research, interpretative phenomenological analysis

1 Introduction

Limb amputation is considered to be one of the oldest surgical procedures with a history of more than 2,500 years (Van der Meij, 1995). It is defined as a medical procedure in which there is "partial or complete removal of a limb for a variety of reasons, including tumours, gangrene, intractable pain, crushing injury or uncontrollable infection" (Fremgen & Frucht, 2009, p.10). Amputation is usually considered as a last resort in the case of life endangerment (Paudel, Shrestha & Banskota, 2005), since it brings about drastic social, economical and psychological changes to both the amputees’ and their families’ lives (Sinha, van den Heuvel & Arokiasamy, 2011). Local statistics indicate that there were approximately 88 Maltese patients who had undergone one or multiple major amputations over a one-year time frame, between 1st June 2011 and 31st May 2012 (Cassar, 2012). Fortunately, the number of major limb amputations above or below the knee in Maltese patients has decreased by around 33% between 2003 and 2010, as a result of early detection, timely intervention and improvement in hospital and medical facilities (Government of Malta, 2011).

Various studies on the experiences of amputees (e.g. Desmond & MacLachlan, 2006; Liu et al., 2010; Senra et al., 2012) focus on three broad aspects: the psychological impact, the processes of adjustment linked to the change in physical image and pain, as well as the impact of social factors such as social support and education. Furthermore, most of the available research (Horgan & MacLachlan, 2004; Senra et al., 2012) focuses on the lived experience of persons having an amputation of the lower limb. This study aims to contribute to this literature by exploring the lived experience of Maltese individuals with a lower or upper limb amputation.

2 Methods

2.1 Participants

Four Maltese participants were interviewed regarding their lived experience of being an amputee. Interpretative phenomenological research is usually conducted using small samples (three to six participants) since a detailed analysis of each case is necessary (Smith, Flowers & Larkin, 2009). The purposive sample consisted of two male and two female amputees whose ages ranged between 30 and 80 years. Participants were recruited from the Amputees4Amputees Association, which is a support group for Maltese amputees. They were informed about the study by the Association through an information letter which contained details on the nature, aims and requirements of the study. Those persons who were willing to participate informed the designated Association member, who then forwarded their contact details to the first author. Criteria for inclusion in the study included being over 18 years of age and having had an amputation for at least 4 years prior to the study, to ensure that emotional distress levels would have returned within the norm (Horgan & MacLachlan, 2004). A profile of the participants is presented in Table 1.

2.2 Data Collection

The interview schedule consisted of ten semi-structured and open-ended questions. Questions were open-ended to provide participants with an opportunity to elaborate on their responses. Their semi-structured nature allowed for probing and a more conversation-like interview, rather than a rigid question-and-answer approach (Langridge & Hagger-Johnson, 2009). The participants could opt to be interviewed in Maltese or English. In fact, all opted for the Maltese language. The duration of the tape-recorded interviews was 40-60 minutes and took place in a
The lived experience of persons with an amputation

Table 1. Participant characteristics.

<table>
<thead>
<tr>
<th>Pseudonym</th>
<th>Gender</th>
<th>Status</th>
<th>Type of amputation</th>
<th>Reason</th>
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<tbody>
<tr>
<td>Jessica</td>
<td>Female</td>
<td>Single</td>
<td>Below the knee</td>
<td>Birth defect - limb malformation</td>
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<tr>
<td>Maria</td>
<td>Female</td>
<td>Married</td>
<td>Above the knee</td>
<td>Gangrene</td>
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<tr>
<td>Joseph</td>
<td>Male</td>
<td>Married</td>
<td>Upper Limb</td>
<td>Accident</td>
</tr>
<tr>
<td>Mario</td>
<td>Male</td>
<td>Married</td>
<td>Below the knee</td>
<td>Accident</td>
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</table>

Note: Participants’ ages are not included in order to preserve their anonymity.

Table 2. Super-ordinate themes.

<table>
<thead>
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<th>Super-ordinate themes</th>
<th>Sub-themes</th>
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<td>Impact on self</td>
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<tr>
<td></td>
<td>Feelings of significant others</td>
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<tr>
<td>A different life</td>
<td>Changes in lifestyle</td>
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<tr>
<td></td>
<td>Prosthesis and discomfort</td>
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<td></td>
<td>Searching for meaning</td>
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<td>Social connections and support</td>
<td>Family and friends</td>
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<td></td>
<td>Professionals as a source of information and care</td>
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setting suggested by the interviewee. Three core areas were explored, namely participant pre-amputation and post-amputation experiences, as well as external factors such as social support and social integration. Interviews were conducted by the first author between December 2012 and January 2013.

2.3 Data Analysis

Interpretative phenomenological analysis (IPA) was used, following procedures outlined by Smith et al. (2009). This approach was found to be the most suitable to examine personal experiences as it involves a detailed analysis of each case (Smith et al., 2009), and therefore the researcher must take on the role of an interpreter (Smith & Osborn, 2003). IPA aims to engage with people’s reflections on their subjective experiences, paying particular attention to what the experience was like for every participant and how s/he made sense of it (Smith et al., 2009). Studies using IPA are often concerned with existential issues in relation to major life-changing or threatening events, as in the case of the loss of a limb (Smith, 2004).

2.4 Ethics

Ethical approval to conduct this study was obtained from the University of Malta Research Ethics Committee (UREC). All participants were asked to sign a consent form prior to the study. They were informed that they could terminate the interview at any point, or decline to answer any question without the need to provide a reason. Pseudonyms were assigned to each participant, and transcripts and audio-recordings were stored in a locked cabinet to safeguard participant confidentiality.

3 Results and Discussion

Three super-ordinate themes emerged from participant accounts. These were: a) the Initial Emotional Reaction, b) a Different Life and c) Social Connections and Support. In the following sections, each super-ordinate theme is divided into sub-themes, which are discussed with the use of excerpts from participant accounts and compared to existing literature.

3.1 An Emotional Reaction

Participants gave details of their own personal reactions to the amputation, as well as the reactions of family members and significant others.

3.1.1 Impact on self

Most of the research available on the experience of amputation is related to one’s reaction to his or her own amputation, which is dependent on a number of factors such as age, level (below or above the knee/elbow) and time since amputation (Horgan & MacLachlan, 2004). The participants in this study described their initial reaction to their own amputation, occurring as a consequence of an accident or gangrene, in a negative manner. Mario explained:

“Initially it was a huge shock. I was crushed.”

Maria stated:

“I really cried.”

Similarly, participants in a study conducted by Senra et al. (2012) expressed feelings of sadness, shock and surprise. However, Jessica, who had her limb amputated due to a malformation in her leg (birth defect), perceived it in a positive light, as exemplified in the following excerpt.

“The iron rods around my [malformed] leg were so ugly and so uncomfortable. In fact, the first thing I asked her [my mother] as a child, was whether the amputation would allow me to wear heels and any kinds of shoes. In that aspect I can say it was more of an improvement for me.”

This demonstrates that the cause of an amputation may influence one’s reaction towards it (Horgan & MacLachlan, 2004). Participants in this study denied experiencing any long-lasting emo-

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tional distress such as depression and anxiety, even though psychopathological symptoms following amputation are commonly cited in the literature (see Hamill, Carson & Dorahy, 2010). This could be due to “heroic cheerfulness” and denial of any problems (Thomson & Haran, 1984, cited in Horgan & MacLachlan, 2004). Another possible reason could be the presence of sufficient support, a possible mediator between amputation and the psychological outcome (Hamill et al., 2010).

All the amputees in this study voiced their preoccupation that the loss of a limb would affect their physical appearance. A research study by Miller and Deathe (2004) highlighted the effect of an amputation on aesthetics. The ability to sustain femininity and wear heels and skirts, was also of particular importance to the females interviewed in this study. In contrast, male participants seemed to be more concerned over the loss of their breadwinner status.

“Even aesthetically, as a woman, I want to make sure that it [the prosthesis] looks good, especially if I’m wearing a dress.” (Jessica)

Joseph also described how the amputated limb affected his self-pride.

“... it made me feel inferior, because I could no longer be my family’s breadwinner.”

This concern finds support in the literature. Men place more importance on the functional aspect of the limb and their ability to financially provide for the family (Murray & Fox, 2002). During each of their interviews, participants expressed similar opinions that certain character traits such as courage, confidence and being positive helped them to cope, as exemplified by the following excerpt.

“... but every human mind is different, not everyone thinks alike. Some people get disheartened... There were some who gave up and passed away within a fortnight.” (Maria)

In a study about positive adjustment to lower limb amputation, Unwin, Kacperek & Clarke (2009) suggested that positive mood was a strong contributor towards a good early adjustment.

3.1.2 Feelings of significant others

Participants also described the emotional responses of significant others in their lives. Maria reported how her granddaughter, with whom she had a close relationship, reacted to the amputation. She explained how initially her granddaughter appeared to be afraid of her, but soon realised that her grandmother was still the same person. Jessica, who had undergone her amputation during childhood, mentioned how preoccupied her mother was with her amputation. She explained how initially her granddaughter appeared to still be the same person. Jessica, who had undergone her amputation. This in turn affected their sexual relationship.

Another participant, Maria, described the change in her intimate relationship with her husband following her amputation. She explained that her husband had a fear of illness and was greatly upset by her amputation. This in turn affected their sexual relationship.

“...It’s very difficult! You have to depend on many people. I hate it when I can’t do something myself.” (Joseph)

Changes in lifestyle were specifically cited by participants who previously enjoyed sports and physical activities.

“...certain things like running, sports and football... The worst thing of them all is not being able to run and play with my children.” (Mario)

According to Asano et al. (2008), mobility was the second best predictor of quality of life among individuals who had a lower limb amputation. In this study, participants highlighted the changes in their everyday life brought about by the amputation. Similar to what was reported by Liu et al. (2010), three of the participants described feelings of frustration as a result of their loss of independence and increased dependence on caregivers.

Another participant, Maria, described the change in her intimate relationship with her husband following her amputation. She explained that her husband had a fear of illness and was greatly upset by her amputation. This in turn affected their sexual relationship.

“...it made me feel inferior, because I could no longer

3.2 A Different Life

Findings from the present study showed that living with an amputation may bring about several changes in one’s life. The following section provides details on the participants’ perceived changes in their life, as a result of their amputation.

3.2.1 Changes in lifestyle

All participants had similar views on the fact that the amputation had an effect on their independence, posing certain limitations and at times making them dependent on others.

“...It’s very difficult! You have to depend on many people. I hate it when I can’t do something myself.” (Joseph)

Changes in lifestyle were specifically cited by participants who previously enjoyed sports and physical activities.

“...certain things like running, sports and football... The worst thing of them all is not being able to run and play with my children.” (Mario)

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3.2.2 Prosthesis and discomfort

All participants described how the use of a prosthesis involved a process of acceptance and adjustment, as demonstrated in the following two excerpts.

“...you start adjusting to the prosthesis by wearing it for half an hour. Then you start going for very short walks.” (Mario)

“...You must trust your body, you must trust the leg so that you can stand on it and walk. You can’t remain dependent on crutches or a wheelchair.” (Jessica)

These comments highlight the importance of the adjustment process of the amputee to his/her prosthetic limb. Saradjian, Thomson and Datta (2008) showed that adjustment to a prosthesis is a personal and individual process. All the participants had different ways of adjusting, with some even customising their prosthesis to suit their own personal needs or wearing it initially for short periods of time. Participants also complained about the discomfort and increased sweating induced by wearing the prosthesis in the hot Maltese summer, as well as the burden of its weight. The former factor was also cited in Saradjian et al.’s (2008) study, in which amputees spoke about the sweating and skin sores caused by wearing a prosthesis in warm temperatures. Although the amputees in the current study did not mention any phantom limb pain, they complained of residual pain and pain in the lower back. Edeh et al. (2000) state that this could be due to altered gait patterns to accomodate the prosthesis.

3.2.3 Searching for meaning

Participants were consistent in reporting that the amputation had resulted in a change in how they viewed their own lives. They explained how they had begun to appreciate life and had learned to live on a day-to-day basis, rather than focusing on the future. It was more evident in those participants who had lost their limb in an accident, as exemplified by the following statement.

“Nowadays, I don’t take anything for granted...because I know I could leave here in five minutes, a minute or a second, and my life could end, and so your priorities do change.” (Joseph)

A common question asked by all the participants was “Why me?”, representing an effort to understand why the amputation had happened to them. Similarly, Dossey and Dossey (1998) had identified the need for participants in their study to arrive at an understanding to this question. This was reported to often result in spiritual discomfort, which could in turn lead to the complete disintegration of God in some amputees (Dossey & Dossey, 1998). This was clearly evident in Mario’s statement.

“I don’t believe in anything anymore. When I look at people going to church nowadays, they remind me of when we used to watch Red Indian movies, in which people adored wood or stones...People go there because...they are obsessed and blind.” Conversely, Maria described a very different experience.

“It [the experience of an amputation] brought me closer to God. It made me realise God was by my side all along.”

Maria goes on to explain how her experience encouraged her to join her local parish group. A relationship with God/higher power has been demonstrated to offer security and strength (Baldacchino et al., 2013) and to serve as a means of positive coping. Joseph also described how his initial reaction was that of anger towards God. However, over time he realised that God had nothing to do with the incident that befell him.

3.3 Social Connections and Support

Participants all highlighted the importance of social support, be it from family and friends, from professionals and staff at hospital, or from other amputees. Furthermore, the importance of support from family and friends was highlighted as one of the most important aspects in their experience.

3.3.1 Family and friends

Participants did not report any change in the way they interacted with others. However, they highlighted the importance of re-integration and remaining socially active by joining organisations, or simply by spending time with friends and family. Williams et al. (2004) reported that this is an important aspect of adjusting to life after the loss of a limb. As explained by Mario:

“The way you re-integrate in the first few months...if you take long to re-integrate, yes, I think you’ll suffer. I was meeting my friends and going to the disco when I was still in a wheelchair. Maintaining your social life is very important!”

Furthermore, family members also served to encourage the amputee during the rehabilitation process. For example:

“You need to let go. You need to let go of your walking aids. In fact, the more time passed, the more I felt pressured by those around me [to trust the prosthesis].” (Jessica)

Other family members improvised ways of enabling the amputee to cope in his/her situation, as described by Joseph:

“I was really lucky that my son was able to modify my prosthesis...he attached an electric motor which helps me a lot.”

Although support from family and friends is considered to be of importance following an amputation (Liu et al., 2010), at certain times, participants felt that they were being over-protected by their families. For instance, Jessica recounted the following episode.

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“Last time my mam saw me climbing up the ladder, to change a light bulb. She started panicking and asked me to get down [due to fear of a fall].”

The participants in Hamill et al.’s (2010) investigation similarly identified this over-protective behaviour as frustrating for amputees.

The amputees in the current study spoke about the need to form relationships with others in the same situation as themselves. They also highlighted the importance of being able to share and encourage each other throughout the experience. One of the participants had been introduced to another amputee her age before the amputation.

“When they introduce you to an amputee who is already walking, you get to see the final results.”

Similarly, Joseph explained:

“That [meeting another amputee] helped me so much, because automatically, when you see someone who has been in the same situation, you can’t help listening to them.”

Other participants managed to meet up with other amputees through information meetings and sessions held by local associations for amputees or when visiting the Orthotic and Prosthetics Unit or Physiotherapy Outpatients Department following their amputation. These experiences tie in with Liu et al.’s (2010) finding that peer support is helpful in providing relief from emotional distress, and reducing isolation.

3.3.2 Professionals as a source of information and care

Health professionals, such as doctors, nurses and physiotherapists, were also perceived as an important source of support. Participants seemed to mention different aspects of support when describing their experience in hospital. Joseph described the support that he received from hospital staff as follows:

“I cannot complain about anything. When I was in hospital, I had good support because I had doctors, nurses...they were all supportive. They were always there to help change my dressings [following the amputation] and provided for my needs.”

In contrast, Maria held a very different viewpoint.

“Nothing! While in hospital I found no emotional support at all. They give you no information on the procedure, the way it will affect your life and how to cope.”

Despite the mixed reactions regarding the support given during their stay in hospital, all participants agreed on the quality of support given at the Orthotic and Prosthetics Unit, which they had been making use of following their amputation.

“They all give their 100% to see you satisfied...they provide the support that you need. They see you as a person and not a number.”

In their review on rehabilitation following major traumatic amputations, Eldar and Jelić (2003) mentioned the importance of early involvement of rehabilitation health professionals who can provide useful advice on rehabilitation, prosthetic options and ways on maintaining muscle strength, as well as self-care skills.

4 Limitations

The present study addresses the lacuna in local literature on the lived experiences of persons with lower and upper limb amputations. There is, however, a need for further research which is longitudinal in nature and explores the lived experiences of study participants over a period time. The limited time frame for this study posed limitations on the number of participants with whom interviews were conducted, and on the possibility of second interviews. Also, the fact that only one of the four participants had an upper limb amputation limits the ability of this study to delve into the experience of amputees of the upper limb. A more diverse sample that included more upper limb amputees would have allowed further understanding of the experiences of persons having different types of amputation.

5 Conclusions

This study explored the lived experience of four Maltese amputees. It identified the various reactions and changes which occur following the loss of a limb. Being specific to the Maltese population, it contributes to a better understanding of the experiences of persons with an amputation in the local context. It also explores the needs of amputees in Malta as well as the type of services provided. Furthermore, it highlights the importance of family members’ and friends’ psychological and physical support as an essential part of individuals’ adjustment to their new life as amputees and the use of their prostheses.

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8 Conflicts of Interest

The authors report no conflicts of interest.

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In order to receive language input, children mainly rely on their

1 Introduction

In order to receive language input, children mainly rely on their
hearing abilities. If the latter are impaired, a temporary or per-
manent hearing difficulty may result. Sensorineural hearing loss
(SNHL) is a permanent type of hearing impairment that has of-
ten led to fitting of hearing aids. However, depending on the
severity of the hearing loss, the latter may not be ideal because

they amplify but do not clarify incoming sound for the hearing-
impaired individual (Welling, 2010). Thus, in cases where the
individual is unable to comprehend spoken language through con-
ventional hearing aids, cochlear implants (CIs) might be an al-
ternative (Roland, 2000). Indeed, CIs are a major technological
attainment to treat deafness (Niparko et al., 2010). They allow
the development of auditory skills in naturalistic, albeit supported
environments and are able to support the acquisition of speech,
language and communication (Archbold, 2002).

Children with a unilateral CI are reported to have difficulty un-
derstanding speech in noise and localising sound (Scherf et al.,
2009). Bilateral implantation can overcome these limitations.
Various researchers (e.g. Battista & Highhouse, 2011; Dunn et
al., 2008; Litovsky et al., 2006; Sammeth, 2007; Therres, 2012)
assigned three effects to binaural listening as a result of bilateral
implantation, namely the head shadow effect, the binaural sum-
mation effect and the binaural squelch effect. In the head shadow
effect, both ears are active. The ear with the more favourable
signal-to-noise ratio (SNR) is immediately accessible and the per-
son can selectively attend to this ear (Therres, 2012). Binaural
summation enables the individual to perceive the sounds presented
to both ears louder. Specifically, an improvement of 3 dB is known
to occur for binaural listeners, leading to better speech perception
(Therres, 2012). Furthermore, when input is received from both
ears, the brain and the auditory system will be able to combine
inputted information from both ears and produce a better cen-
tral representation, providing a clear separation of the speech and
noise signals (Therres, 2012). This is referred to as the binaural
squelch effect. With these reported benefits, the cochlear implant
enhances the individual’s hearing experience which, as a result,
would help the acquisition of speech and language.

Individuals with bilateral implants are implanted either sequen-
tially or simultaneously. Sharma et al. (2007) compared the P1
latency, an evoked potential which analyses the auditory cortical
response to auditory stimuli, in 20 children who received a bilat-
eral implantation during the first 3;06 years of life. Ten children
had a simultaneous implantation while the rest received sequen-
tial bilateral implants. In both groups, the average P1 latency was
measured on implant activation and at two-month intervals post-
implantation up to 15 months. The P1 latency for both groups
was within normal limits by 3.5 months post-implantation. This
suggests that any bilateral implantation that occurs within the
first 3;06 years takes place within a highly plastic central auditory
nervous system.

Despite the sensitive time proposed by Sharma et al. (2007),
several studies have shown that there should be no age limit for
the second implantation. Scherf et al. (2009) observed the audi-
tory abilities and speech performance of 35 children with sequen-

Abstract. The purpose of this study was to document the
speech and language skills of three Maltese children with bilateral
cochlear implants. One child was simultaneously implanted and
had a hearing age (HA) of 2;10 years at the time of testing, an-
other was sequentially implanted and had a HA of 3;06 years,
while the third child was sequentially implanted and had a
HA of 5;03 years. Maltese standardised speech and language
assessments were used to gather information on the children’s
speech and language skills, with data being collected during
their speech and language therapy sessions. Following data
transcription and analysis, the participants’ speech and language
abilities were compared to those of their HA- and chronological
age (CA)-matched peers using available norms for Maltese
children. Additional information regarding the children’s speech
and language history prior to and post-implantation was also
collected, providing a holistic overview of the participants’ speech
and language development. Results indicated that the children
presented with speech and language delay when compared to
their CA-matched peers. Variations across the participants
were found in specific language skills. Similarities in language
patterns were also noted, including expressive abilities in advance
of receptive skills. These findings extend the limited data on
the speech and language skills of Maltese children with cochlear
implants, with comparison to norms for typically-developing
children being a novel approach to research in this area.

Keywords speech, language, hearing loss, Maltese chil-
dren, cochlear implants, bilingualism

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tial bilateral CIs following three years of bilateral implant use. CIs offered improved auditory skills to all children, even those sequentially implanted after the age of 6:00 years, although it emerged that the latter took longer than those implanted before the age of 6:00 years to demonstrate bilateral benefit. The older children, that is, those implanted after the age of 6:00 years, obtained better speech recognition levels. Additionally, at two to three years post-bilateral implantation, almost all children benefited in noisy situations.

In another study, Kim, Kim and Jeong (2013) investigated the functional benefits of sequential bilateral cochlear implantation in 42 children having a mean inter-stage interval of 5:06 years between the two implants. Average ages were 4:02 years at the first implant and 9:07 years at the second implant. The participants were grouped according to the inter-stage intervals. The second and third group, those with an inter-stage interval of 5:00 and 6:09 years, and 7:00 and 9:09 years respectively, obtained comparable results in both quiet and noise tests to the first group, who had a shorter interval of 3:00 and 4:09 years, two years following bilateral use. Additionally, speech performance in noise improved even in the older children following prolonged use of bilateral CIs. These results show that the binaural hearing experience plays an important role in the speech outcome.

On the other hand, simultaneous implantation enhances children’s speech and language acquisition and decreases their ability difference in comparison to their chronological age (CA)-matched peers. Wie (2010) investigated a group of 21 children with cochlear implants two years post-simultaneous implantation. The majority of these acquired receptive and expressive language skills at a faster pace than expected in comparison to their hearing age (HA)-matched peers. Apart from the type of implant, language skills depend on the severity of the SNHL, the age of identification and implantation, as well as the child’s cognitive abilities (Welling, 2010). For example, Niparko et al. (2010), who studied 188 children who underwent cochlear implantation before the age of five, reported greater rates of improvement in children with better residual hearing and higher socioeconomic status. Additionally, Geers, Nicholas and Sedey (2003), who studied 181 8:00- and 9:00-year-old children who also received a cochlear implant by the age of five, found that the child’s learning ability contributed greatly to the linguistic outcome. Participants with average learning ability were reported to show language levels similar to those of their HA-matched peers. Similar findings were reported for two Maltese children with a cochlear implant (Tabone, 2004). However, Geers et al. (2003) further stated that children with average or above average intelligence performed similarly to their CA-matched peers. Together, these results show that the linguistic outcome is multifactorial.

Speech production is influenced by the child’s age at onset of deafness, time of implantation as well as the mode of communication and rehabilitation (Robbins, 2000). CIs improve speech perception and intelligibility (Robbins, 2006), both of which eventually improve speech production. In the two Maltese children with CI studied by Tabone (2004), speech development was identified as delayed, implying typical developmental stages emerging with a time lag, rather than as deviant. Furthermore, the Maltese-speaking child observed by Xuereb (2004) at 4:06 years post-implantation experienced speech and language delay when compared to her CA-matched peers on tests of speech and language. However, the tests used in both case studies were not standardised on the Maltese-speaking population, so interpretations of results need to be considered as tentative.

Importantly, speech and language assessments standardised on the Maltese hearing population have recently become available, enabling re-evaluation of the speech and language abilities of children with CIs in relation to those of Maltese hearing children. Such a comparison was the purpose of the current study. The following research questions were addressed:

1. What speech and language skills are manifested by three Maltese children with bilateral CIs?
2. Are the speech and language skills of these three Maltese children comparable to those shown by HA- or CA-matched typically-developing children?

More specifically, the study documented the participants’ phonetic inventory, phonological error patterns, inconsistency in word production, verbal comprehension and expression, aiming to identify their strengths and weaknesses. It also sought to determine whether their skills were the same as, similar to or different from the speech and language abilities documented in a normative sample of Maltese children. This information was needed to indicate whether the speech and language development of the three Maltese participants with CIs was age-appropriate, delayed or deviant in relation to hearing children, while acknowledging the limited generalisability of results to other Maltese children with CIs.

2 Methods

2.1 Participants

All speech-language pathologists working in the public health service were asked to distribute an information sheet among potential participants in their current or previous caseloads. Children considered as potential participants had bilateral cochlear implants, a HA ranging between 2:00 and 6:00 years and were Maltese-English bilinguals. Children reported to have other developmental disabilities were excluded. Three children having a HA in the range of 2:10-5:03 years were identified as participants. Table 1 lists background information on each child.

2.2 Research design

A multiple case study design was implemented. Since this design is known to enable researchers to investigate real-life events holistically (Yin, 2009), its implementation in this study allowed the chosen participants to be studied in depth. The status of their speech and language development was assessed through the use of standardised assessments and results were analysed quantitatively and qualitatively. Furthermore, previous audiology and speech and language progress reports were accessed for background information that could help in the interpretation of results.

2.3 Method

All data were collected by the first author. Background information was obtained by reviewing the participants’ medical and audiology file. Details on their hearing assessment results and implantation were noted. Their HA was subsequently calculated. Speech and language data were then collected through the use of two standardised assessments, outlined below, which enabled each participant’s scores to be compared with population norms.

The Maltese-English Speech Assessment (MESA) (Grech, Dodd & Franklin, 2011) assesses speech and taps into articulation, phonology, consistency, diadochokinesis, consonantal clusters and multisyllabic words, as well as single oral movements and sequenced movements. In the present study, only the articulation, phonology and consistency subtests were administered. The articulation and phonology subtests involved naming 42 coloured pictures eliciting all the English and Maltese vowels and consonants (Grech & Dodd, 2008). This enabled compilation of their phonetic inventory and the tabulation of phonemes. Percentage Consonants Correct (PCC) and Percentage Vowels Correct (PVC) scores were calculated and error patterns were described and analysed. In the consistency subtest, the participants were to name 17 pictures, three times on separate trials. An inconsistency score was then computed.

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The Language Assessment for Maltese Children (LAMC) (Grech, Franklin & Dodd, 2011) examines language skills, tapping into verbal comprehension, narrative vocabulary and grammar, sentence imitation, phonological awareness and pragmatics. It also includes a voice and fluency checklist. Participants were administered the verbal comprehension, narrative vocabulary and grammar subtests. They were first asked questions related to the story previously narrated by the assessor using a picture stimulus book. They were subsequently encouraged to retell the story with the help of the stimulus book.

Prior to commencing data collection, the study was approved by the University of Malta Research Ethics Committee, ensuring that the children's rights were safeguarded. The legal guardians of the potential participants were given an information sheet through the speech-language pathologist, explaining the children’s potential involvement in the study. For Child 3, the information sheet was passed on through the speech-language pathologist previously working with the child. Upon understanding the children's involvement in the study and agreeing to their participation, parents completed a consent form.

### 3 Results

Descriptive statistics, including raw scores, means, standard deviations, z-scores and percentiles for each participant on every speech and language subtest, were calculated. The children’s individual performance was compared to that of typically-developing HA- and CA-matched peers involved in assessment standardisation. However, the available norms for both assessments cover the 3:00-6:00-year age group, with the exception of the verbal comprehension subtest, for which norms begin at 2:06 years. Grech, Franklin and Dodd (2011) explain that children aged 2:00-2:06 years carried out only a small part of the language assessment. The resulting data did not warrant further analysis. Consequently, given Child 1’s actual HA of 2:10 years, he was compared to HA-matched peers aged 3:00 years. Analagously, Child 2 and Child 3, whose actual CAs were 6:10 and 7:05 years respectively, were both compared to CA-matched peers aged 6:00 years, as this was the ceiling age of both assessments.

When the MESA was standardised, children were divided into monolingual and bilingual groups. As defined by Grech, Dodd and Franklin (2011), the monolingual group consisted of children exposed to Maltese at home and to English at school, whilst children in the bilingual group had simultaneous exposure to both languages at home. Hence, the results of Child 1 and 2 were compared to the monolingual group norming data, whilst Child 3’s data were compared to the bilingual group.

Each participant’s phonetic inventory was determined first. According to Ingram (1981, cited in Grech, 2006), a criterion for a sound to become part of the child’s inventory is its frequency, in that it has to occur at least once in a mixture of 25 words selected at random. The phonetic inventory was noted from the articulation subtest of the MESA by listing the sounds that the child produced. These sounds are hereafter referred to as phones. The number of phonemes was subsequently counted from performance on the phonology subtest. For a sound to be considered a phoneme it should be used regularly and contrastively and it should occur in minimal or near minimal pairs (Grech, 2006). The phonology subtest also enabled calculation of the PCC and PVC scores. Finally, the inconsistency score was derived from the participants’ inconsistent production of words on the consistency subtest.

### 2.4 Ethical considerations

Prior to commencing data collection, the study was approved by the University of Malta Research Ethics Committee, ensuring that the participants’ rights were safeguarded. The legal guardians of the potential participants were given an information sheet through their speech-language pathologist, explaining the children’s potential involvement in the study. For Child 3, the information sheet was passed on through the speech-language pathologist previously working with the child. Upon understanding the children's involvement in the study and agreeing to their participation, parents completed a consent form.
### Table 2. Child 1’s PCC, PVC and inconsistency scores (means and standard deviations (SD) quoted from the MESA manual) with the equivalent z-scores and percentiles derived for HA- and CA-matched peers

<table>
<thead>
<tr>
<th>Raw Score</th>
<th>Mean Quoted</th>
<th>SD Quoted</th>
<th>Derived z-score</th>
<th>Derived Percentile</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>PCC</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 2;10 years</td>
<td>91.45 89.6</td>
<td>9.20 0.201</td>
<td>58</td>
<td></td>
</tr>
<tr>
<td>CA: 4;08 years</td>
<td>91.45 97.23</td>
<td>2.68 -2.157</td>
<td>2</td>
<td></td>
</tr>
<tr>
<td><strong>PVC</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 2;10 years</td>
<td>98.99 99.18</td>
<td>3.29 -0.058</td>
<td>48</td>
<td></td>
</tr>
<tr>
<td>CA: 4;08 years</td>
<td>98.99 99.89</td>
<td>0.35 -2.609</td>
<td>0.5</td>
<td></td>
</tr>
<tr>
<td>PVC without outlier</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 2;10 years</td>
<td>100 99.18</td>
<td>3.29 0.250</td>
<td>60</td>
<td></td>
</tr>
<tr>
<td>CA: 4;08 years</td>
<td>100 99.89</td>
<td>0.345 0.320</td>
<td>63</td>
<td></td>
</tr>
<tr>
<td><strong>Inconsistency Score</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 2;10 years</td>
<td>29.41 15.05</td>
<td>18.59 0.748</td>
<td>77</td>
<td></td>
</tr>
<tr>
<td>CA: 4;08 years</td>
<td>29.41 4.03</td>
<td>7.79 3.258</td>
<td>100</td>
<td></td>
</tr>
</tbody>
</table>

### Table 3. Child 1’s phonological error processes, frequency of occurrence and age at which they are expected, based on data by Grech, Dodd and Franklin (2011)

<table>
<thead>
<tr>
<th>Phonological Error Processes</th>
<th>Error Processes</th>
<th>Frequency</th>
<th>Typically expected</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Systemic Error Processes</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Voicing</td>
<td>2</td>
<td>3.00 - 3.05 years</td>
<td></td>
</tr>
<tr>
<td>Backing</td>
<td>2</td>
<td>3.00 - 3.05 years</td>
<td></td>
</tr>
<tr>
<td>Fronting</td>
<td>2</td>
<td>3.00 - 6.00 years</td>
<td></td>
</tr>
<tr>
<td>Lateralisation of /r/</td>
<td>2</td>
<td>3.00 - 4.11 years</td>
<td></td>
</tr>
<tr>
<td>Nasalisation</td>
<td>1</td>
<td>MEP*</td>
<td></td>
</tr>
<tr>
<td><strong>Structural Error Processes</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Weak syllable deletion</td>
<td>1</td>
<td>3.00 - 3.05 years</td>
<td></td>
</tr>
<tr>
<td>Final consonant deletion</td>
<td>1</td>
<td>3.00 - 4.11 years</td>
<td></td>
</tr>
<tr>
<td>Initial consonant deletion</td>
<td>1</td>
<td>3.00 - 3.11 years</td>
<td></td>
</tr>
<tr>
<td>Compensatory vowel lengthening</td>
<td>1</td>
<td>MEP*</td>
<td></td>
</tr>
<tr>
<td>Consonant harmony</td>
<td>1</td>
<td>MEP*</td>
<td></td>
</tr>
<tr>
<td>Epenthesis</td>
<td>1</td>
<td>MEP*</td>
<td></td>
</tr>
<tr>
<td>Gemination reduction</td>
<td>1</td>
<td>MEP*</td>
<td></td>
</tr>
<tr>
<td>Cluster formation</td>
<td>1</td>
<td>MEP*</td>
<td></td>
</tr>
</tbody>
</table>

*MIniminally used error pattern (MEP): an error pattern used by less than 10% of children between 3:00 and 6:00 years

### 3.1 Child 1

Child 1 produced a complete phone repertoire, 22 phones, which included all the Maltese consonants documented by Azzopardi (1981) (cited in Borg & Azzopardi-Alexander, 1997). Given the child’s CA (4:08 years) and Grech and Dodd’s (2008) MESA results indicating that both sequential and simultaneous Maltese-English bilingual children produce all phones by 4:00 years, this was an expected finding. Therefore, Table 2 shows Child 1’s raw scores on the MESA speech subtests in comparison to his HA- and CA-matched peers and Table 3 lists his phonological error processes. In the light of his CA, Child 1 should have mastered the majority of the adult phonemes (Grech, Dodd & Franklin, 2011). The child did show a complete phonemic inventory by producing 22 phonemes. Yet, his phonological system was still developing, with his performance similar to that of his HA-matched peers. On PCC, he scored close to the average of his HA-matched peers yet well below average in comparison to his CA-matched peers. A similar performance was noted on PVC. Although Child 1 performed below average in PVC when compared to both groups, there was a slight difference between his raw score and the mean score for both age-matched groups. This was due to an omitted vowel. Without this outlier, the child would have obtained a full score. Also, the possibility of an inconsistent phonological disorder was eliminated as the child obtained an inconsistency score lower than 40% (see Grech, Dodd & Franklin, 2011). However, the discrepancy between the child’s score and the mean score for both age groups seems to suggest that he was still learning the Maltese and English phonological systems. As discussed by Grech and Dodd (2008), inconsistent production increases with initial exposure to two languages.

Child 1 produced five systemic and eight structural error processes. None were considered frequently-used error patterns. Yet, they are potential error patterns. When compared to his HA-matched group, seven error processes used by the child were all typically used at 3:00 years. Alternatively, when compared to his CA peers, voicing, backing, weak syllable deletion and initial consonant deletion should have been eradicated by the age of 4:08 years. Furthermore, half the error processes used by Child 1 (see minimally used error patterns

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Table 4. Child 1’s scores on verbal comprehension and narrative ideas (means and standard deviations (SD) quoted from the LAMC manual) with the equivalent z-scores and percentiles derived for HA- and CA-matched peers

<table>
<thead>
<tr>
<th>Raw Score</th>
<th>Quoted Mean</th>
<th>Quoted SD</th>
<th>Derived z-score</th>
<th>Derived Percentile</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Verbal Comprehension Score</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 2;10 years</td>
<td>11.50</td>
<td>10.61</td>
<td>3.38</td>
<td>0.263</td>
</tr>
<tr>
<td>CA: 4;08 years</td>
<td>11.50</td>
<td>16.80</td>
<td>1.87</td>
<td>-2.834</td>
</tr>
<tr>
<td><strong>Narrative Ideas Score</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 2;10 years</td>
<td>14</td>
<td>0.92</td>
<td>1.44</td>
<td>9.083</td>
</tr>
<tr>
<td>CA: 4;08 years</td>
<td>14</td>
<td>9</td>
<td>1.44</td>
<td>9.083</td>
</tr>
</tbody>
</table>

Table 5. Child 2’s PCC, PVC and inconsistency scores (means and standard deviations quoted from the MESA manual) with the equivalent z-scores and percentiles derived for HA- and CA-matched peers

<table>
<thead>
<tr>
<th>Raw Score</th>
<th>Quoted Mean</th>
<th>Quoted SD</th>
<th>Derived z-score</th>
<th>Derived Percentile</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>PCC</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 3;06 years</td>
<td>96.32</td>
<td>93.35</td>
<td>6.47</td>
<td>0.456</td>
</tr>
<tr>
<td>CA: 6;10 years</td>
<td>96.32</td>
<td>95.53</td>
<td>5.58</td>
<td>0.135</td>
</tr>
<tr>
<td><strong>PVC</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 3;06 years</td>
<td>98.85</td>
<td>99.99</td>
<td>0.02</td>
<td>-57</td>
</tr>
<tr>
<td>CA: 6;10 years</td>
<td>98.85</td>
<td>100</td>
<td>0*</td>
<td>-</td>
</tr>
<tr>
<td><strong>PVC without outlier</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 3;06 years</td>
<td>100</td>
<td>99.99</td>
<td>0.02</td>
<td>0.5</td>
</tr>
<tr>
<td>CA: 6;10 years</td>
<td>100</td>
<td>100</td>
<td>0*</td>
<td>-</td>
</tr>
<tr>
<td><strong>Inconsistency Score</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 3;06 years</td>
<td>35.29</td>
<td>10.60</td>
<td>14.96</td>
<td>1.65</td>
</tr>
<tr>
<td>CA: 6;10 years</td>
<td>35.29</td>
<td>3.19</td>
<td>14.96</td>
<td>4.827</td>
</tr>
</tbody>
</table>

* A standard deviation of 0 indicates no variation around the mean score, so that the z-score and percentile could not be computed.

(MEP) in Table 3) were typical between 2:00 and 3:00 years, as documented in Grech’s (1998) study. This further suggested delayed phonological development in comparison to the child’s CA. Child 1’s verbal comprehension and narrative ideas scores are shown in Table 4.

Child 1 understood simple commands and ‘why’ questions. However, comprehension of ‘who’ and ‘what’ was difficult. Therefore, his understanding of wh-questions was still emerging. As documented by Sax and Weston (2007), typically-developing children between 2:00 and 3:00 years comprehend and produce simple wh-questions. However, Friedmann and Szterman (2011) reported that the understanding of wh-questions was still emerging. As documented by Sax and Weston (2007), typically-developing children between 2:00 and 3:00 years comprehend and produce simple wh-questions. However, Friedmann and Szterman (2011) reported that the understanding and production of wh-questions of 11 Hebrew-speaking children, some of whom were fitted with binaural hearing aids and others with a CI, was weaker compared to typically-developing children. Child 1 also understood basic colour terms, but knowledge of numbers and counting was still emerging.

Child 1’s expression was mainly in Maltese. He used proper names and target lexical nouns appropriately with correct singular and plural forms. Various lexical target verbs and the definite and assimilated article were also employed appropriately. There was evidence of the use of the Maltese negative form mhux (not), the English lexeme ‘no’ and spatial and pronominal use of deictic expressions. On 90% of occasions, he produced correct person and number agreement and adequate sentence structures. On the other hand, Child 1 made limited use of adjectives and adverbs, conjunctions, prepositions, direct and indirect pronouns. He did not use possessives and showed gender agreement difficulties. The masculine form was used for 90% of the time. Infrequent and incorrect use of feminine forms possibly indicated their emergence at the time. A similar trend was documented by Coppini (2002), who investigated gender agreement between nouns and adjectives in Maltese children.

3.2 Child 2

Child 2 produced 22 phones, which made up a complete phone repertoire in view of his HA (3:06 years) and CA (7:10 years). Table 5 shows his raw scores on the MESA speech subtests in comparison to his HA- and CA-matched peers. Table 6 lists the phonological error processes.

Child 2 produced 22 phonemes, thus he had also developed a complete phonemic inventory. However, his phonological development was still emerging. On PCC, Child 2 performed moderately and slightly above average compared to his HA- and CA-matched peers respectively. Furthermore, on PVC, the slight difference between the child’s score and that of the normative population was mainly due to the incorrect production of /mer/ (mirror), for which he said /mir/ (woman). Without this outlier, Child 2 obtained a full score, further indicating no vowel difficulty. His inconsistency score indicated a higher possibility of an inconsistent
Child 2 exhibited a chronological mismatch in the phonological processes observed. He produced five systemic error processes and no structural error processes. When compared to his HA-matched group, Child 2 produced developmental phonological processes typical of children younger than his HA. Alternatively, when compared to his CA-matched peers, backing, voicing and stopping should have been eliminated by 3;11 years. Therefore, a phonological delay was observed. Conversely, as expected by his CA, Child 2 did not exhibit structural error processes. Thus, he followed some patterns expected at 6;10 years.

Child 2’s phonological error processes are shown in Table 6.

**Table 6. Child 2’s phonological error processes, frequency of occurrence and age at which they are expected based on data by Grech, Dodd and Franklin (2011)**

<table>
<thead>
<tr>
<th>Error Processes</th>
<th>Frequency</th>
<th>Typically expected</th>
</tr>
</thead>
<tbody>
<tr>
<td>Backing</td>
<td>3</td>
<td>3;00 - 3;05 years</td>
</tr>
<tr>
<td>Devoicing</td>
<td>1</td>
<td>3;00 - 3;05 years</td>
</tr>
<tr>
<td>Stopping</td>
<td>1</td>
<td>3;00 - 3;11 years</td>
</tr>
<tr>
<td>Delteralisation of /l/</td>
<td>1</td>
<td>MEP*</td>
</tr>
<tr>
<td>Friction</td>
<td>1</td>
<td>MEP*</td>
</tr>
</tbody>
</table>

*Minimally used error pattern (MEP): an error pattern used by less than 10% of children between 3;00 and 6;00 years

Child 2 understood simple commands and ‘why’ questions. Comprehension of ‘where’ and ‘what’ questions, as well as knowledge of numbers and counting was emerging. He made appropriate regular lexical choices of nouns and produced the Maltese negative form ma (verb)x. He made regular and adequate use of articles and conjunctions. On 90% of occasions, he also made correct verb choices and used adequate pronouns. The direct object was used appropriately but infrequently, on less than 30% of occasions. Child 2 showed correct gender, number and person agreement. On the other hand, he replaced proper names with nouns and made adequate use of prepositions while omitting others. His use of adjectives and adverbs amounted to less than 30% of their required usage. He rarely employed spatial deictic expressions and sentence structure was occasionally disrupted. The indirect object was incorrectly used due to person and number disagreement and possessive terms did not emerge.

**Table 7. Child 2’s scores on verbal comprehension and narrative ideas (means and standard deviations (SD) quoted from the LAMC manual) with the equivalent z-scores and percentiles derived for HA- and CA-matched peers**

<table>
<thead>
<tr>
<th></th>
<th>Quoted Mean</th>
<th>Quoted SD</th>
<th>Derived z-score</th>
<th>Derived Percentile</th>
</tr>
</thead>
<tbody>
<tr>
<td>Verbal Comprehension</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 3;06 years</td>
<td>10.5</td>
<td>14.87</td>
<td>2.50</td>
<td>-1.748</td>
</tr>
<tr>
<td>CA: 6;10 years</td>
<td>10.5</td>
<td>17.21</td>
<td>2.43</td>
<td>-2.761</td>
</tr>
<tr>
<td>Narrative Ideas</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 3;06 years</td>
<td>16</td>
<td>2.88</td>
<td>3.19</td>
<td>4.113</td>
</tr>
<tr>
<td>CA: 6;10 years</td>
<td>16</td>
<td>16.86</td>
<td>6.30</td>
<td>0.137</td>
</tr>
</tbody>
</table>

**Table 8. Child 3’s PCC, PVC and inconsistency scores (means and standard deviations quoted from the MESA manual) with the equivalent z-scores and percentiles derived for HA- and CA-matched peers**

<table>
<thead>
<tr>
<th></th>
<th>Quoted Mean</th>
<th>Quoted SD</th>
<th>Derived z-score</th>
<th>Derived Percentile</th>
</tr>
</thead>
<tbody>
<tr>
<td>PCC</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 5;03 years</td>
<td>100</td>
<td>99.34</td>
<td>2.21</td>
<td>0.299</td>
</tr>
<tr>
<td>CA: 7;06 years</td>
<td>100</td>
<td>99.18</td>
<td>1.24</td>
<td>0.661</td>
</tr>
<tr>
<td>PVC</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 5;03 years</td>
<td>100</td>
<td>99.94</td>
<td>0.21</td>
<td>0.280</td>
</tr>
<tr>
<td>CA: 7;06 years</td>
<td>100</td>
<td>99.74</td>
<td>0.83</td>
<td>0.313</td>
</tr>
<tr>
<td>Inconsistency Score</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HA: 5;03 years</td>
<td>5.88</td>
<td>0.45</td>
<td>1.63</td>
<td>3.331</td>
</tr>
<tr>
<td>CA: 7;06 years</td>
<td>5.88</td>
<td>1.32</td>
<td>2.62</td>
<td>1.740</td>
</tr>
</tbody>
</table>

http://dx.medra.org/10.14614/COCHIMP.2.60  http://www.um.edu.mt/healthsciences/mjhs
The child’s generation of expressive ideas on narrative was excellent when compared to his HA-matched peers. His performance was above average when compared to his HA-matched peers. Performance was superior to that of his CA-matched peers. This was consistent with Tabone’s (2004) and Xuereb’s (2004) conclusions on the speech development of Maltese children with CI. The child’s expressive ideas were well developed. He was able to produce sentences of two to three information-carrying words. Therefore, rewording of his narratives was not necessary.

4 Discussion

The aim of this study was to describe and evaluate the speech and language skills of three children with bilateral CIs, to derive information on their strengths and weaknesses in relation to their HA- and CA-matched peers. The participants were assessed using three speech and two language subtests of the MESA and LAMC, respectively, all of which were standardized on the Maltese population. Discussion of results follows and acknowledges the potential influence of each child’s internal characteristics, learning environment and speech and language therapy on their speech and language performance.

Child 1 followed typical speech development. Yet, his scores indicated that he was on a par with his HA group whilst delayed when compared to his CA-matched peers. This was consistent with Tabone’s (2004) and Xuereb’s (2004) conclusions on the speech development of Maltese children with CI. On the verbal comprehension subtest, Child 1 performed within the average range compared to his HA-matched peers. Caselli et al. (2012) reported similar trends in lexical comprehension for 17 Italian children who received a CI at 2/0 years of age. The child’s generation of expressive ideas on narrative was excellent when compared to his HA-matched peers. Performance was superior to that of his CA-matched peers. Therefore, at 2;10 years post-simultaneous bilateral implantation, the child’s expressive skills were progressing well. This concurred with Wie’s (2010) findings that the majority of children simultaneously implanted acquire both receptive and expressive language skills at a faster pace than expected in comparison to their HA. As observed from the reviewed files, the family members provided good support to the child. This was well reflected in his expressive language, which was developing at a steady pace. Additionally, it was reported by his speech-language pathologist that co-operation and carryover of therapy in the natural environment contributed to the child’s positive prognosis. Furthermore, the fact that the child was given a shared learning support assistant (LSA) from kindergarten years enhanced and supported his educational needs. The speech-language pathologist liaised with the child’s LSA to consolidate the child’s learning in both the school and clinical contexts.

Child 2’s speech development followed the pattern of typically-developing children. Nevertheless, his scores were similar to his HA-matched peers and sometimes even slightly delayed in comparison. He exhibited typical phonological error processes with a chronological mismatch, since he used error processes from different age groups with inconsistent production of words. The latter might be indicative of incomplete knowledge of two phonological systems. It seems reasonable to associate high variability in speech production with children having the least hearing experience. However, this was still evident at the HA of 3;06 years, reflecting instability in adult-like word production.

Child 2’s performance on verbal comprehension was considerably poor when compared to both age groups. His difficulty understanding wh-questions was possibly due to the delayed auditory stimulation of 3;04 years. In fact, his audiology file reported that his left ear, being the worse one, was implanted at 3;04 years while he was wearing the right hearing aid on a full-time basis. Yet, as documented in his audiology file, Child 2 seldom made use of this hearing aid. He received his second implant at 5;10 years. Scherf et al. (2009) observed improved auditory skills even in children receiving their second CI after the age of 6;00 years. Yet, older children obtained comparable results two years following bilateral use. In the case of Child 2, bilateral use had been in effect for one year. This suggests that further bilateral experience was required. In contrast, Child 2’s narrative ideas were excellent when compared to his HA-matched group and slightly below average when compared to his CA-matched peers. This discrepancy between his receptive and expressive skills is not what is normally experienced by a typically-developing child. The wh-questions were likely to have impeded his performance on the comprehension subtest. From the assessments carried out by his speech-language pathologist, the child showed understanding of commands with two to three information-carrying words. Therefore, rewording of the comprehension assessment questions and substitution of words like ‘briet’ (girls) with simpler ones might have possibly enhanced his performance. However, since both assessments used are standardized, this was not possible.

Additionally, Child 2 had few instances of unintelligibility which

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Table 9. Child 3’s verbal comprehension and narrative ideas scores (means and standard deviations (SD) quoted from the LAMC manual) with the equivalent z-scores and percentiles derived for HA- and CA-matched peers

<table>
<thead>
<tr>
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<th>Raw Score</th>
<th>Quoted Mean</th>
<th>Quoted SD</th>
<th>Derived z-score</th>
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<td><strong>Verbal Comprehension Score</strong></td>
<td></td>
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<tr>
<td>HA: 5;03 years</td>
<td>18.5</td>
<td>19</td>
<td>1</td>
<td>-0.500</td>
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<tr>
<td>CA: 7;06 years</td>
<td>18.5</td>
<td>18</td>
<td>1.41</td>
<td>0.355</td>
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<tr>
<td><strong>Narrative Ideas Score</strong></td>
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<td>HA: 5;03 years</td>
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<td>29.33</td>
<td>4.04</td>
<td>1.403</td>
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<td>CA: 7;06 years</td>
<td>35</td>
<td>23</td>
<td>7.07</td>
<td>1.697</td>
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might have been due to the manifestation of systemic error processes. Allen, Nikolopoulos and O'Donoghue (1998) concluded that children with CI continue to develop intelligibility even after five years of implantation. Also, during the speech and language therapy sessions following implantation, emphasis was placed on the production of Ling sounds (/r/, /l/, /j/, /m/, /f/, /s/) and thus, not all the speech sounds were targeted. This might have also possibly contributed to unintelligibility in the production of words. As in the case of Child 3, speech and language therapy targeted all the speech sounds in all the different word positions and also within sentences.

Child 3 showed typical speech skills. Her performance was similar to her CA-matched peers. Yet, unexpectedly, Child 3 obtained a higher percentile when compared to her CA than when compared to her HA-matched group. However, Grech and Dodd (2007) noted a dip in the phonological developmental profile of 5:00-5:05-year-old typically-developing bilingual children (Child 3’s HA-matched group). Thus, the child’s performance was typical for her HA. Moreover, she showed adequate phonological proficiency in both Maltese and English. This could have been due to her early exposure to both languages.

The language subtests also showed Child 3 to perform better in comparison to her CA- than to her HA-matched group. This led to the reliability of the derived equivalent z-scores and percentiles being queried. As discussed in the LAMC manual, z-score accuracy may be low when used for smaller samples (Grech, Franklin & Dodd, 2011). The English norming group to whom Child 3 was compared was small (N = 30) compared to the Maltese group (N = 205).

Child 3 performed well above average compared to both groups of age-matched peers. She had outgrown her HA peers and performed closely to her CA-matched peers. Her hearing experience of 5:03 years might have influenced her skills, given that she had the longest exposure to language when compared to the other participants in this study. In fact, Schramm, Bohnert and Keilmann (2010) concluded that children with CIs increase their linguistic abilities with their HA, that is, the longer the child is fitted with cochlear implants, the better the linguistic outcome. Furthermore, Child 3 was the only female participant in the study. Schramm et al. (2010) observed that better language competence was developed by females, possibly indicating that this factor might have played a role in her linguistic performance. Interestingly, and unlike the other two participants, it was revealed by Child 3’s file that following the diagnosis of hearing loss, she attended sessions with a teacher of the hearing-impaired, during which she was taught baby signs. It was reported that the child immediately picked up the signs and used them communicatively. However, the means to communicate from an early age might have therefore limited the gap in relation to her CA-matched peers. Also, following the implantations, Child 3 attended speech and language therapy sessions on a weekly basis. At the time of testing, the child did not attend therapy as speech and language difficulties were no longer reported. Furthermore, although Child 3 performed similarly to her CA-matched peers (6:00 years), her actual CA was 7:06 years. Thus, one needs to be cautious when interpreting her results, as a fair and accurate comparison would have ideally involved her actual CA.

It is acknowledged that this study presented with shortcomings which could not be controlled. These called for cautious interpretation of results. Data on Maltese children with bilateral CIs were not available. Thus, reference was made to sparse data on Maltese children with unilateral CI and comparison was then made to results of foreign research studies. However, exact comparisons could not be carried out due to the different languages and methodologies used. Data on Maltese typically-developing children were also limited. Moreover, generalisation of the results was not possible due to the case study design and the individual variability across the participants, which might have contributed to the differences in scores. Therefore, each participant was analysed and discussed separately.

5 Conclusion

The results of the current study shed light on the speech and language performance of three Maltese children with bilateral CIs. These findings are preliminary and cannot be generalised. However, with respect to the three participants, it appears that cochlear implantation provides relevant auditory information that helps develop oral communication. It was concluded that two of the participants presented with delayed but typical speech development, whilst the third child performed on a par with her CA-matched peers. With increased CI experience, phonetic accuracy improved. Additionally, the expressive language score was higher than verbal comprehension for all participants. It emerged that the earlier the cochlear implantation, the shorter was the gap in sequential implants. Moreover, better speech and language outcomes were observed with increased experience with CIs. Results showed that early cochlear implantation enabled the participants to engage in a typical school environment with their HA-matched peers.

6 Acknowledgements

The authors are grateful to Professor Helen Grech for granting them permission to use the standardised assessments and their respective norms, and to the audiologist and speech-language pathologists who helped in identifying the participants. Warmest thanks go to the parents of the participants for consenting to their children’s participation and to the children themselves, who patiently and cheerfully co-operated during assessment.

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8 Conflicts of Interest

The authors report no conflicts of interest.

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