CANADIAN JOURNAL OF SPEECH-LANGUAGE PATHOLOGY AND AUDIOLOGY

CJSLPA | RCOA

REVUE CANADIENNE D'ORTHOPHONIE ET D'AUDIOLOGIE

Fall ▸ automne, 2013

Volume 37, No. 3

CANADIAN JOURNAL OF SPEECH-LANGUAGE PATHOLOGY AND AUDIOLOGY

Vol. 37, No. 3
Fall 2013

Purpose and Scope

The Canadian Association of Speech-Language Pathologists and Audiologists (CASLPA) is the only national body that supports and represents the professional needs of speech-language pathologists, audiologists and supportive personnel inclusively within one organization. Through this support, CASLPA champions the needs of people with communications disorders. The association was founded in 1964 and incorporated under federal charter in 1975. CASLPA's periodical publications program began in 1973.

The purpose of the Canadian Journal of Speech-Language Pathology and Audiology (CJSLPA) is to disseminate contemporary knowledge pertaining to normal human communication and related disorders of communication that influence speech, language, and hearing processes. The scope of the Journal is broadly defined so as to provide the most inclusive venue for work in human communication and its disorders. CJSLPA publishes both applied and basic research, reports of clinical and laboratory inquiry, as well as educational articles related to normal and disordered speech, language, and hearing in all age groups. Classes of manuscripts suitable for publication consideration in CJSLPA include tutorials; traditional research or review articles; clinical, field, and brief reports; research notes; and letters to the editor (see Information to Contributors). CJSLPA seeks to publish articles that reflect the broad range of interests in speech-language pathology and audiology, speech sciences, hearing science, and that of related professions. The Journal also publishes book reviews, as well as independent reviews of commercially available clinical materials and resources.

Copyright

Indexing

CJSLPA is indexed by:

- CINAHL – Cumulative Index to Nursing and Allied Health Literature
- Elsevier Bibliographic Databases (SCOPUS)
- ProQuest – CSA Linguistics and Language Behavior Abstracts (LLBA)
- PsycInfo
- Thomson Gale (Academic Onefile)
- EBSCO Publishing Inc. (CINHAL Plus with full text)
- Directory of Open Access Journals

Online Archive

CJSLPA is now an open-access publication. For full-text articles and archives, visit: www.cjslpa.ca

Advertising

All inquiries concerning the placement of advertisements in CJSLPA should be directed to pubs@caslpa.ca. Acceptance of an ad does not in any way constitute CASLPA's endorsement of the product/service or company. CASLPA reserves the right to reject any ad if the advertisement, organization, product, or service is not compatible with CASLPA's mission or vision. CASLPA does not accept responsibility for the accuracy of statements by advertisers.

CJSLPA Reviewers

Joy Armson, Kathleen Arnos, Venu Balasubramanian, Pauline Beaupré, Renée Beland, François Bergeron, (Barbara) May Bernhardt, Kumiko Boike, Alejandro Brice, Françoise Brosseau-Lapré, Ferenc Bunta, Sonia Cabell, Kate Chase, Margaret Cheesman, Patricia Cleave, Paola Colozzo, Vikram Dayalu, Chantal Desmarais, Louise Duchesne, Carl Dunst, Ollie Eckberg, Caroline Erdos, Irani Farzan, Christian Giguère, Jacqueline Guendouzi, Elaine Hall, Carol Hammond, Ellen Hickey, Irene Hoshko, Anne-Marie Hurteau, Tiffany Hutchins, Merv Hyde, Jean-Pierre Gagné, Sophia Kramer, Marilyn Kertoy, Michael Kiefte, Ariane Laplante-Lévesque, Anne-Lise Leclercq, Pascal Lefebvre, Tony Leroux, Vinaya Manchaiah, Marguerite MacKenzie, Andrea MacLeod, Christelle Maillart, Elina Maniela-Arnold, André Marcoux, Rebecca McCauley, David McFarland, Lu-Anne McFarlane, Shane Moodie, Laura Murray, Glen Nowell, Bruce Oddson, Johanne Paradis, Marianne Paul, Pesco Diane, Laura Plexico, Brigitte Poirier, Karen Pollock, Laya Poost-Foroosh, Yvan Rose, Phyllis Schneider, Melanie Schuele, Alix Seigneuric, Mike Shelton, Gurjit Singh, Jeff Small, Angela South, Kristie Spencer, Elin Thordardottir, Natacha Trudeau, Christine Turgeon, Christine Valiquette, Susan Wagner, Gail Whitelaw.

ISSN 1913-200X

CASLPA Vision and Mission

Vision

The Canadian Association of Speech-Language Pathologists and Audiologists ...the national voice and recognized resource for speech-language pathology and audiology.

Mission

The Canadian Association of Speech-Language Pathologists and Audiologists ...supporting and empowering our members to maximize the communication and hearing potential of the people of Canada.

CJSLPA is published quarterly by the Canadian Association of Speech-Language Pathologists and Audiologists (CASLPA). Publications Agreement Number: # 40036109. Return undeliverable Canadian addresses to: CASLPA, 1 Nicholas Street, Suite 1000, Ottawa, Ontario K1N 7B7. Address changes should be sent to CASLPA by e-mail to pubs@caslpa.ca or to the above-mentioned address.

REVUE CANADIENNE D'ORTHOPHONIE ET D'AUDIOLOGIE

Objet et Portée

L'Association canadienne des orthophonistes et audiologistes (ACOA) est l'association professionnelle nationale reconnue des orthophonistes et des audiologistes du Canada. L'Association a été fondée en 1964 et incorporée en vertu de la charte fédérale en 1975. L'Association s'engage à favoriser la meilleure qualité de services aux personnes atteintes de troubles de la communication et à leurs familles. Dans ce but, l'Association entend, entre autres, contribuer au corpus de connaissances dans le domaine des communications humaines et des troubles qui s'y rapportent. L'Association a mis sur pied son programme de publications en 1973.

L'objet de la Revue canadienne d'orthophonie et d'audiologie (RCOA) est de diffuser des connaissances relatives à la communication humaine et aux troubles de la communication qui influencent la parole, le langage et l'audition. La portée de la Revue est plutôt générale de manière à offrir un véhicule des plus compréhensifs pour la recherche effectuée sur la communication humaine et les troubles qui s'y rapportent. La RCOA publie à la fois les ouvrages de recherche appliquée et fondamentale, les comptes rendus de recherche clinique et en laboratoire, ainsi que des articles éducatifs portant sur la parole, le langage et l'audition normaux ou désordonnés pour tous les groupes d'âge. Les catégories de manuscrits susceptibles d'être publiés dans la RCOA comprennent les tutoriels, les articles de recherche conventionnelle ou de synthèse, les comptes rendus cliniques, pratiques et sommaires, les notes de recherche, et les courriers des lecteurs (voir Renseignements à l'intention des collaborateurs). La RCOA cherche à publier des articles qui reflètent une vaste gamme d'intérêts en orthophonie et en audiologie, en sciences de la parole, en science de l'audition et en diverses professions connexes. La Revue publie également des critiques de livres ainsi que des critiques indépendantes de matériel et de ressources cliniques offerts commercialement.

Droit d'auteur

Inscription au Répertoire

RCOA est répertoriée dans:

- CINAHL – Cumulative Index to Nursing and Allied Health Literature
- Elsevier Bibliographic Databases (SCOPUS)
- ProQuest – CSA Linguistics and Language Behavior Abstracts (LLBA)
- PsycInfo
- Thomson Gale (Academic Onefile)
- EBSCO Publishing Inc. (CINHAL Plus with full text)

Archive en-ligne

Les articles et les archives de la RCOA sont maintenant disponibles au publique à www.cjslpa.ca

Publicité

Toutes les demandes visant à faire paraître de la publicité dans la RCOA doivent être adressées au Bureau national. L'acceptation d'une annonce publicitaire ne signifie absolument pas que l'ACOA fait la promotion du produit, du service ou de la compagnie. L'ACOA se réserve le droit de rejeter une annonce si le message, l'organisation, le produit ou le service n'est pas compatible avec la mission, la vision ou les valeurs de l'ACOA. L'ACOA n'assume pas la responsabilité de l'exactitude des déclarations des annonceurs.

Réviseurs de la RCOA

Joy Armson, Kathleen Arnos, Venu Balasubramanian, Pauline Beaupré, Renée Beland, François Bergeron, (Barbara) May Bernhardt, Kumiko Boike, Alejandro Brice, Françoise Brosseau-Lapré, Ferenc Bunta, Sonia Cabell, Kate Chase, Margaret Cheesman, Patricia Cleave, Paola Colozzo, Vikram Dayalu, Chantal Desmarais, Louise Duchesne, Carl Dunst, Ollie Eckberg, Caroline Erdos, Irani Farzan, Christian Giguère, Jacqueline Guendouzi, Elaine Hall, Carol Hammond, Ellen Hickey, Irene Hoshko, Anne-Marie Hurteau, Tiffany Hutchins, Merv Hyde, Jean-Pierre Gagné, Sophia Kramer, Marilyn Kertoy, Michael Kiefte, Ariane Laplante-Lévesque, Anne-Lise Leclercq, Pascal Lefebvre, Tony Leroux, Vinaya Manchaiah, Marguerite MacKenzie, Andrea MacLeod, Christelle Maillart, Elina Maniela-Arnold, André Marcoux, Rebecca McCauley, David McFarland, Lu-Anne McFarlane, Shane Moodie, Laura Murray, Glen Nowell, Bruce Oddson, Johanne Paradis, Marianne Paul, Pesco Diane, Laura Plexico, Brigitte Poirier, Karen Pollock, Laya Poost-Foroosh, Yvan Rose, Phyllis Schneider, Melanie Schuele, Alix Seigneuric, Mike Shelton, Gurjit Singh, Jeff Small, Angela South, Kristie Spencer, Elin Thordardottir, Natacha Trudeau, Christine Turgeon, Christine Valiquette, Susan Wagner, Gail Whitelaw.

ACOA : Vision et Mission

Vision

L'Association canadienne des orthophonistes et audiologistes : porte-parole national et ressource reconnue dans le domaine de l'orthophonie et de l'audiologie.

Mission

L'Association canadienne des orthophonistes et audiologistes appuie et habilite ses membres en vue de maximiser le potentiel en communication et en audition de la population canadienne.

Vol. 37, No. 3
automne 2013

ISSN 1913-200X

La RCOA est publiée quatre fois l'an par l'Association canadienne des orthophonistes et audiologistes (ACOA). Numéro de publication : #40036109.
Faire parvenir tous les envois avec adresses canadiennes non reçus au 1, rue Nicholas, bureau 1000, Ottawa (Ontario) K1N 7B7.
Faire parvenir tout changement à l'ACOA au courriel pubs@caslpa.ca ou à l'adresse indiquée ci-dessus.

Table of Contents

Table des Matières

From the Editor

FALL ISSUE

The fall issue of CJSLPA presents readers with a variety of papers related to pediatric and adult issues from both audiology and speech-language pathology. In the first article, Stanton and Griffin share new understandings of auditory function from studies in genetics and genomics and offer an interesting commentary on current and potential applications of genetics to the field of hearing loss. They first introduce readers to basic genetic concepts and then describe some of the recent advances in genetics and genomics that are applicable to hearing health.

Lagacé and colleagues contribute to the literature on French assessment of auditory processing disorders. They present the results of a study that aimed to explore the effects of regional differences in New Brunswick when using a French adaptation of the *Staggered Spondaic Word (SSW)* Test. Through their work, the authors underscore the importance of developing specific normative data to account for regional differences when using recorded verbal stimuli.

In the third article, Peladeau-Pigeon and Steele discuss videofluroscopy in an effort to provide speech-language pathologists with a basic understanding of technical aspects of the procedure. The article describes types of available fluoroscopes and discusses factors influencing imaging, as well as various parameters related to imaging. Through their paper, the authors hope to contribute to enhance speech-language pathology – radiology teamwork in videofluroscopy.

In the final article of this issue, Green and Roth report on a pilot study that examined the benefits of a specific 8-week inferential reading comprehension program with a fourth grade student who had a language disorder. The authors describe the specific characteristics of the training program and conclude that their results provide preliminary support for improvement in reading performance.

We continue to enjoy a steady stream of submissions to CJSLPA reflecting the range of research taking place in Canada and elsewhere. We welcome manuscripts from all aspects of Speech-Language Pathology and Audiology and at this time, we are particularly interested in receiving more articles from the Audiology community in both English and French.

I am aware that some authors and reviewers have encountered challenges when submitting through the CJSLPA online system. The CASLPA staff is working to address these issues and is reviewing the current online submission and review system. In the meantime, thank you for your patience and please do not hesitate to contact CJSLPA technical support at support@coverpage.ca. Thank you to the many reviewers who have contributed your time this year and for your patience working through the online system. If you are able to review for the journal, please take a minute to register at www.cjslpa.coverpage.ca or alternatively send us an email to let us know your areas of interest.

Elizabeth Fitzpatrick, PhD

cjslpa.rcoa@caslpa.ca

elizabeth.fitzpatrick@uottawa.ca

Mot de la rédactrice en chef

NUMÉRO D'AUTOMNE

Le numéro d'automne de la RCOA présente à ses lecteurs une variété d'études reliées à des questions touchant les enfants et les adultes, tant en audiologie qu'en orthophonie. Dans le premier article, Stanton et Griffin partagent de nouvelles façons de comprendre la fonction auditive à partir d'études en génétique et en génomique, et proposent un commentaire intéressant sur les applications actuelles et potentielles de la génétique au domaine de la perte auditive. Elles présentent initialement aux lecteurs des concepts génétiques de base, pour ensuite décrire quelques-uns des progrès récents en génétique et en génomique qui sont applicables à la santé de l'ouïe.

Lagacé et ses collègues contribuent à la littérature sur l'évaluation en français des troubles du traitement auditif. Ils présentent les résultats d'une étude réalisée au Nouveau-Brunswick qui se proposait d'explorer les effets des différences régionales lors de l'utilisation d'une adaptation française du test *Staggered Spondaic Word (SSW)*. Par leurs travaux, les auteurs soulignent l'importance du développement de données normatives spécifiques pour tenir compte des différences régionales lors de l'utilisation des stimuli verbaux enregistrés.

Dans le troisième article, Péladeau-Pigeon et Steele discutent de la vidéofluoroscopie pour tenter d'offrir aux orthophonistes une compréhension de base des aspects techniques de la procédure. L'article décrit des types de fluoroscopes disponibles et discute de facteurs qui influencent l'imagerie, ainsi que divers paramètres ayant rapport à ces techniques. Par leur communication, les auteures espèrent contribuer à améliorer le travail d'équipe en orthophonie–radiologie dans la vidéofluoroscopie.

Dans le dernier article, Green and Roth livrent un rapport sur une étude pilote qui examinait les bénéfices d'un programme spécifique de 8 semaines de compréhension inférentielle en lecture avec un élève de 4e année ayant un trouble du langage. Les auteures décrivent les caractéristiques particulières du programme de formation et concluent que leurs résultats offrent un appui préliminaire en vue de l'amélioration du rendement en lecture.

Nous continuons à bénéficier d'un flot ininterrompu de soumissions à la RCOA reflétant l'étendue de la recherche qui se fait au Canada et ailleurs. Nous invitons des manuscrits qui traitent de tous les aspects de l'orthophonie et de l'audiologie et, présentement nous sommes particulièrement intéressés à recevoir en plus grand nombre des articles en anglais et en français dans le domaine de l'audiologie.

Nous savons que plusieurs auteur(e)s et réviseur(e)s ont eu des difficultés à nous faire parvenir des soumissions par le truchement du système en ligne de la RCOA. Le personnel de l'ACOA s'affaire à régler ces problèmes et évalue présentement le système de soumission et de révision en ligne. Entre temps, merci de votre patience et prière de ne pas hésiter à contacter le soutien technique de la RCOA, à support@coverpage.ca. Merci aux nombreux réviseur(e)s qui ont consacré de leur temps cette année et merci de votre patience face aux difficultés rencontrées avec notre système en ligne. Si vous pouvez participer à la révision de manuscrits soumis à notre revue, prenez quelques minutes pour vous inscrire à www.cjslpa.coverpage.ca ou bien faites-nous parvenir un courriel pour nous faire part de vos domaines d'intérêt.

Elizabeth Fitzpatrick, Ph. D.

cjslpa.rcoa@caslpa.ca

elizabeth.fitzpatrick@uottawa.ca

The Genomics of Hearing Loss: A New Era for Clinical Practice

La génomique de la surdité : une nouvelle ère de la pratique clinique

KEY WORDS
GENETICS
INHERITED
NONSYNDROMIC
MUTATION
DEAFNESS
HEARING LOSS

Susan G Stanton
Anne Griffin

Abstract

Many aspects of audiological practice, from the choice of assessment protocols to the selection of intervention options for our hearing impaired patients, are guided by our understanding of the underlying auditory pathology.Biomedical science is undergoing a significant transformation that experts predict will continue at an exponential pace, altering the course of clinical practice for all health care professionals, including audiologists. As a consequence of the progress in genetics and genomics research, a revolution in our understanding of normal auditory function and disease has emerged during the last two decades. Genetic tests provide a window into the temporal bone, and have revolutionized our understanding of human cochlear structure, function and pathobiology. In this paper, we present a brief introduction to basic genetic concepts, and an overview of the recent advances in genetics and genomics relevant to hearing health care.

Abrégé

De nombreux aspects de la pratique de l'audiologie, à partir du choix des protocoles d'évaluation jusqu'à celui des options d'intervention pour nos patients malentendants, sont guidés par la compréhension que nous avons de la pathologie auditive sous-jacente. La science biomédicale est en voie de subir une transformation importante qui, selon les experts, va se poursuivre à une cadence exponentielle et va modifier le cours de la pratique clinique pour tous les professionnels de la santé, y compris les audiologistes. Grâce à la recherche génétique et génomique connaissant une croissance exponentielle, une révolution dans notre compréhension de la fonction auditive normale et de la maladie a émergé pendant les deux dernières décennies. Des tests génétiques ont été axés, entre autres, sur l'os temporal et ils ont révolutionné notre compréhension de la structure cochléaire humaine, de sa fonction et de sa pathobiologie. Dans cet article, nous présentons des concepts génétiques de base et un aperçu des récents progrès en génétique et en génomique qui ont rapport aux soins de santé relatifs à l'audition.

Susan G Stanton, PhD
National Centre for Audiology,
School of Communication
Sciences and Disorders,
Western University,
London, ON
Canada

Anne Griffin, M.Sc. Aud
Griffin Audiology Services
Research Associate, Memorial
University Faculty of Medicine,
Discipline of Genetics
St. John's, NL
Canada

Introduction

Many aspects of audiological practice, from the choice of assessment protocols to the selection of intervention options for our hearing impaired patients, are guided by our understanding of the underlying auditory pathology. We often counsel patients about the nature of hearing loss while explaining strategies for hearing loss prevention, or how treatment options vary depending on the type and location of auditory system damage. Yet, the details of an individual patient's specific etiology are often unresolved, and our elucidation of the underlying pathology is limited to a general categorization by location; sensorineural hearing loss, for example, is a deficit affecting the cochlea and/or auditory nerve. Imagine knowing the precise nature of your patient's lesion, involving a defective gene that causes a cochlear conductive sensorineural hearing loss because the tectorial membrane biomechanics are abnormal (Plantinga, Cremers, Huygen, Kunst & Bosman, 2007).Or, that you are counselling a young patient with a family history of early presbycusis, whose genomic testing reveals a neural receptor gene that confers vulnerability to age-related hearing loss (Friedman et al., 2009). Breakthroughs in genetics and genomics have revolutionized our understanding of human biology and disease; this is particularly true for the cochlea and the pathogenesis of hearing impairment. By studying how individual genes explain a particular phenotype or set of physical traits, scientists have come to realize that for some patients, their disease is due to a defect in a single gene. This is often the case for patients with inherited, early-onset hearing loss. A mutation in a single gene is responsible for their particular hearing loss phenotype. However, even when a single defect in one influential gene can cause disease, this genetic change must be considered in the context of genomics - in other words, an individual's entire genetic makeup, and also their environment. By studying genetics in this context of genomics, scientists and clinicians can tackle not only how interactions between multiple genes and the environment can influence the auditory deficit caused by a single gene of large effect (for example, why two siblings with the same genetic defect have different degrees of congenital loss), but also the etiology of complex, multifactorial impairments such as noise-induced hearing loss and presbycusis. In this review we provide an overview of the progress in genetics and genomics with implications for hearing health care, and introduce some basic concepts for understanding the genetics and genomics of hearing loss.

The New Molecular Era – A Vision for Personalized Health Care

Biomedical science is undergoing a significant transformation that experts predict will continue at an exponential pace, altering the course of clinical practice for all health care professionals, including audiologists. Because of the Human Genome Project (2011), and more recently the HapMap and 1000 Genomes Projects (The 1000 Genes Project Consortium, 2010; The International HapMap 3 Consortium, 2010), it is now recognized that genes contribute in some way to most diseases, a revelation that opens up new avenues for prevention and treatment. A new vision from representatives of the U.S. National Human Genome Research Institute illuminates the evolving science of genomic medicine, including the challenges and implications for human health, as researchers "navigate a course from the base pairs of the human genome sequence to the bedside of patients" (Green, Guyer, & National Human Genome Research Institute, 2011; p. 212). In the future, health care will become personal at a fundamental level, with disease prevention and management tailored to each patient's unique and entire genetic makeup.

However, early predictions for a rapid transition into the molecular era of personalized medicine have yet to be realized. To date, the impact of genetic and genomic science on the everyday clinical practice of most health care professionals has been negligible. Despite this, health, research and educational policy makers are preparing for the "genomics" era (CIHR, 2010; Green et al., 2011; NCHPEG, 2011;OBA, 2011).With public awareness and access to genetic information and services expanding, medical and allied health care providers in Canada (Carrol et al., 2009) and worldwide (Burke et al., 2002; EuroGentest, 2011; GenEd Project, 2011; Greendale & Pyeritz, 2001; HUGO, 2011; Metcalfe, Hurworth, Newstead, Robins, 2002; WHO, 2011) are being encouraged to acquire competencies in genetics and genomics. This is particularly relevant for those providing services to the deaf and hard-of-hearing, a patient population for whom gene discovery and the translation of related biomedical innovations into the clinical realm are outpacing those in most other clinical specialties (Shearer et al., 2010; Van Camp & Smith; 2011).

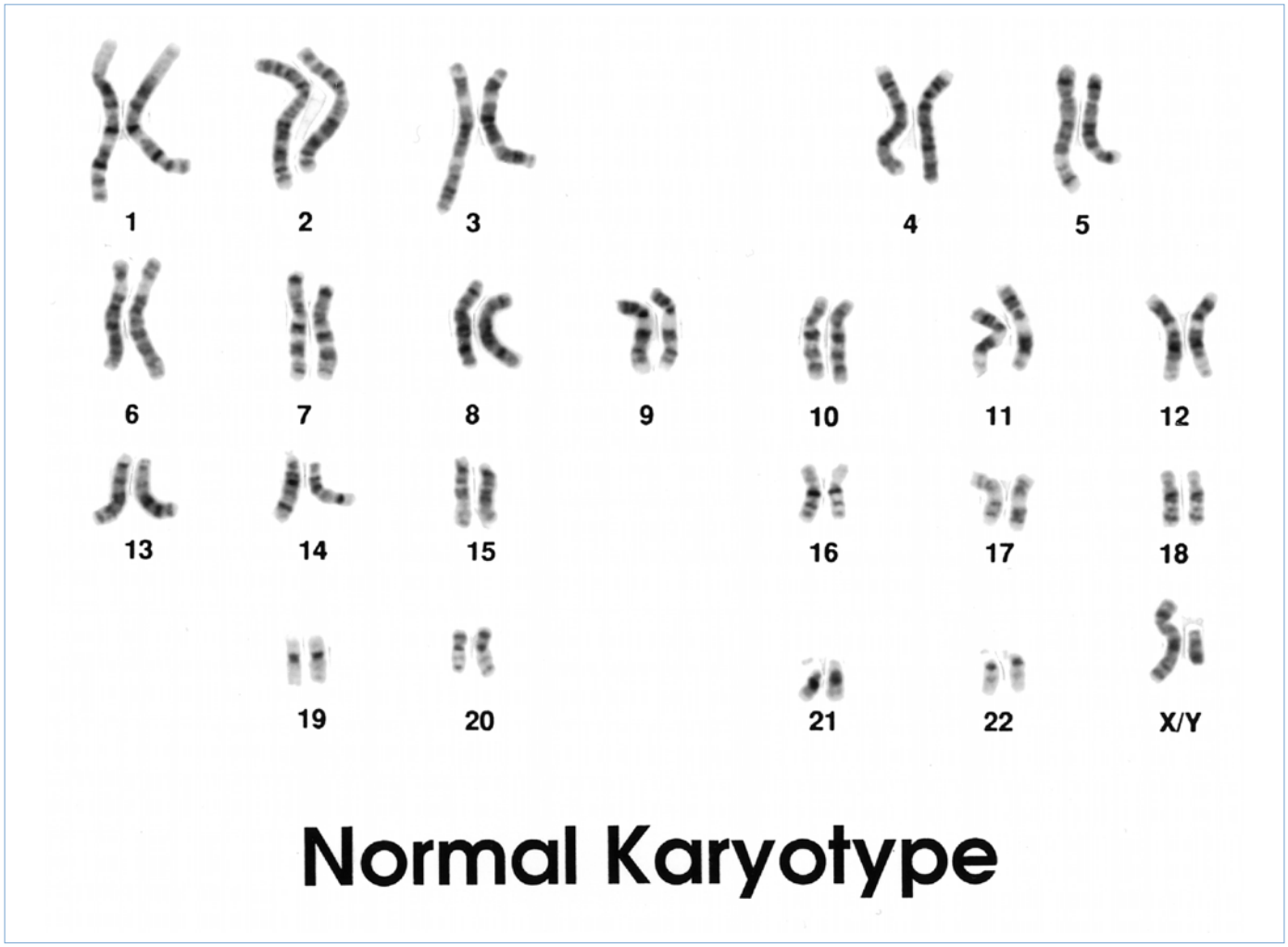
How Genetic Studies Have Evolved in the New "Molecular Era": Implications for Hearing Health Care.

A few decades ago, clinical genetics was a medical specialty dealing with diseases caused by chromosomal or a few rare single gene defects, and genetic testing involved detecting changes in chromosome structure or number, for example Trisomy 21 causing Down Syndrome (see Figure 1). The genes responsible for most diseases were unknown, including those causing hearing loss, and clinical tests for single gene mutations were generally lacking (Korf, 2000). The evaluation of a hearing impaired child involved determining whether the hearing impairment was isolated, or accompanied by other features; in other words, a nonsyndromic or syndromic form of hearing loss. Epidemiological studies indicate that 70-85% of inherited

Figure 1. Human Chromosomes

A. Human karyotype

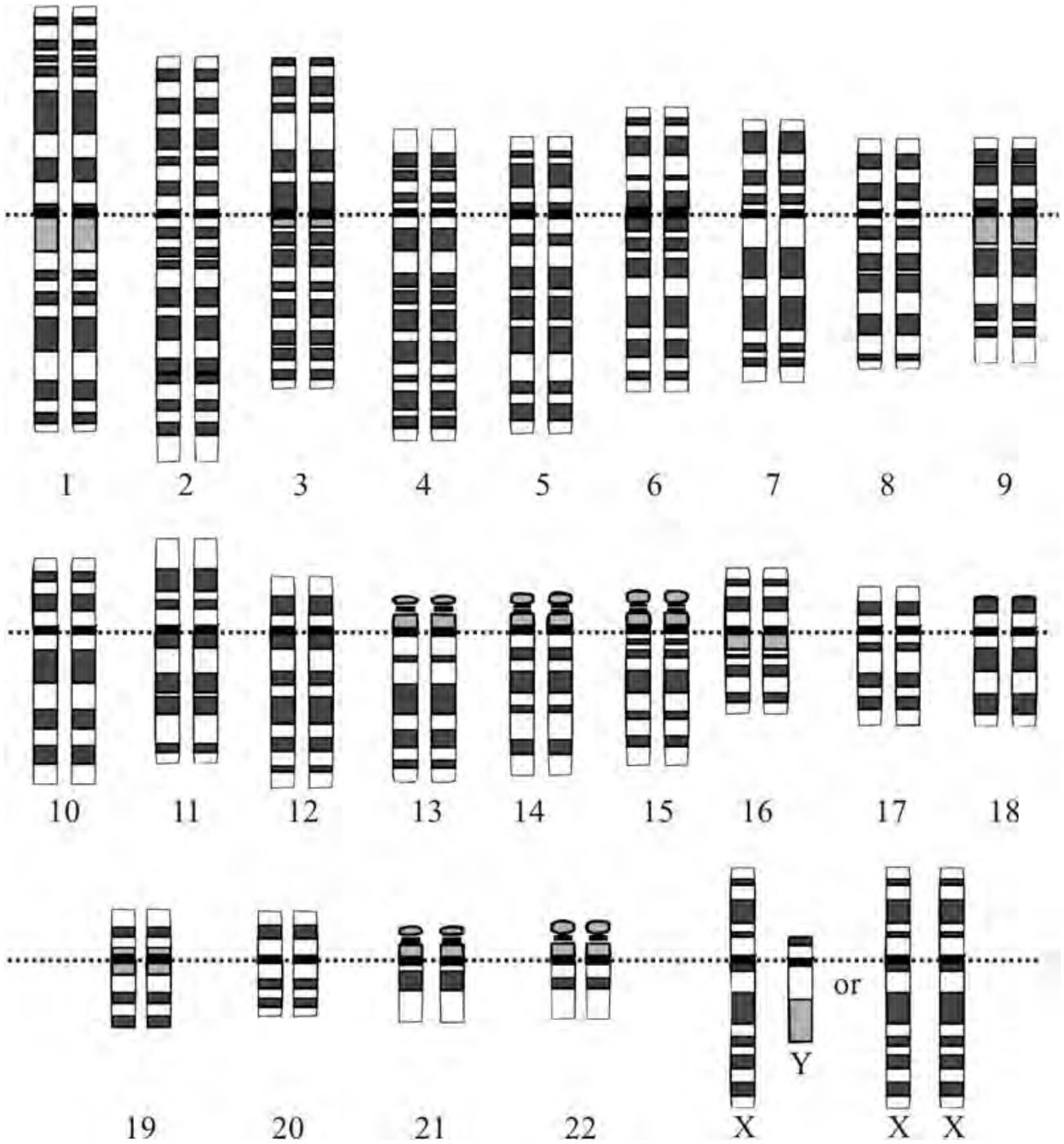
Microsopic images of human chromosomes are arranged in pairs and numbered according to size. Almost all human cells (except gametes and red blood cells) contain 46 chromosomes: 2 sex chromosomes, XX or XY depending on gender, and 22 pairs of matched, non-sex autosomes. For clinical purposes, a karyotype provides information about an individual's chromosome number and structure, their sex chromosomes, and any abnormalities. The karyotype shown is for a normal male: 46, XY.



Source: http://visualsonline.cancer.gov/details.cfm?imageid=2721

B. Ideogram: Normal

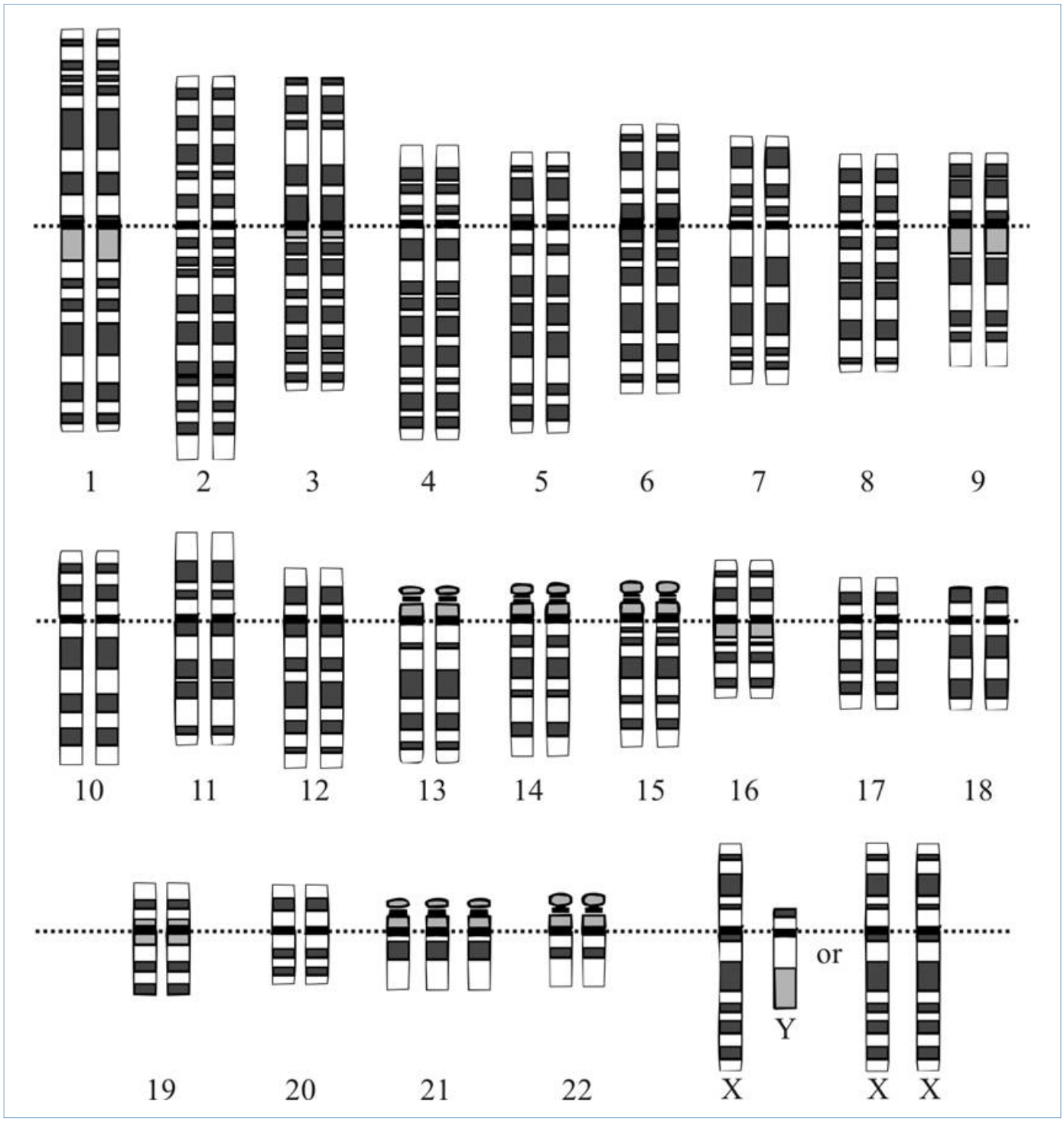
An ideogram is a diagram of the human chromosomes. Ideograms are arranged like the karyotype, with the chromosomes organized according to chromosomal number and relative size. The banding pattern is also shown for each chromosomes. These dark and light bands occur when chromosomes are prepared for microscopy, and are used to describe the location or "locus" of genes on each chromosome. This ideogram describes a normal female: 46, XX.



Source: National Human Genome Research Institute. http://en.wikipedia.org/wiki/File:Down_Syndrome_Karyotype.png#file

C. Ideogram: Trisomy 21

Major abnormalities include changes in the chromosomal number or in the gross structure of chromosomes. Extra or missing chromosomes, as well as breaks or rejoined chromosomes can be detected by microscopic examination and karyotype evaluation. Down syndrome is a chromosomal disorder and is often caused by an error in cell division that results in three copies of chromosome 21, also known as "trisomy 21". The human ideogram shown here represents trisomy 21 in a male: 47, XY, +21.



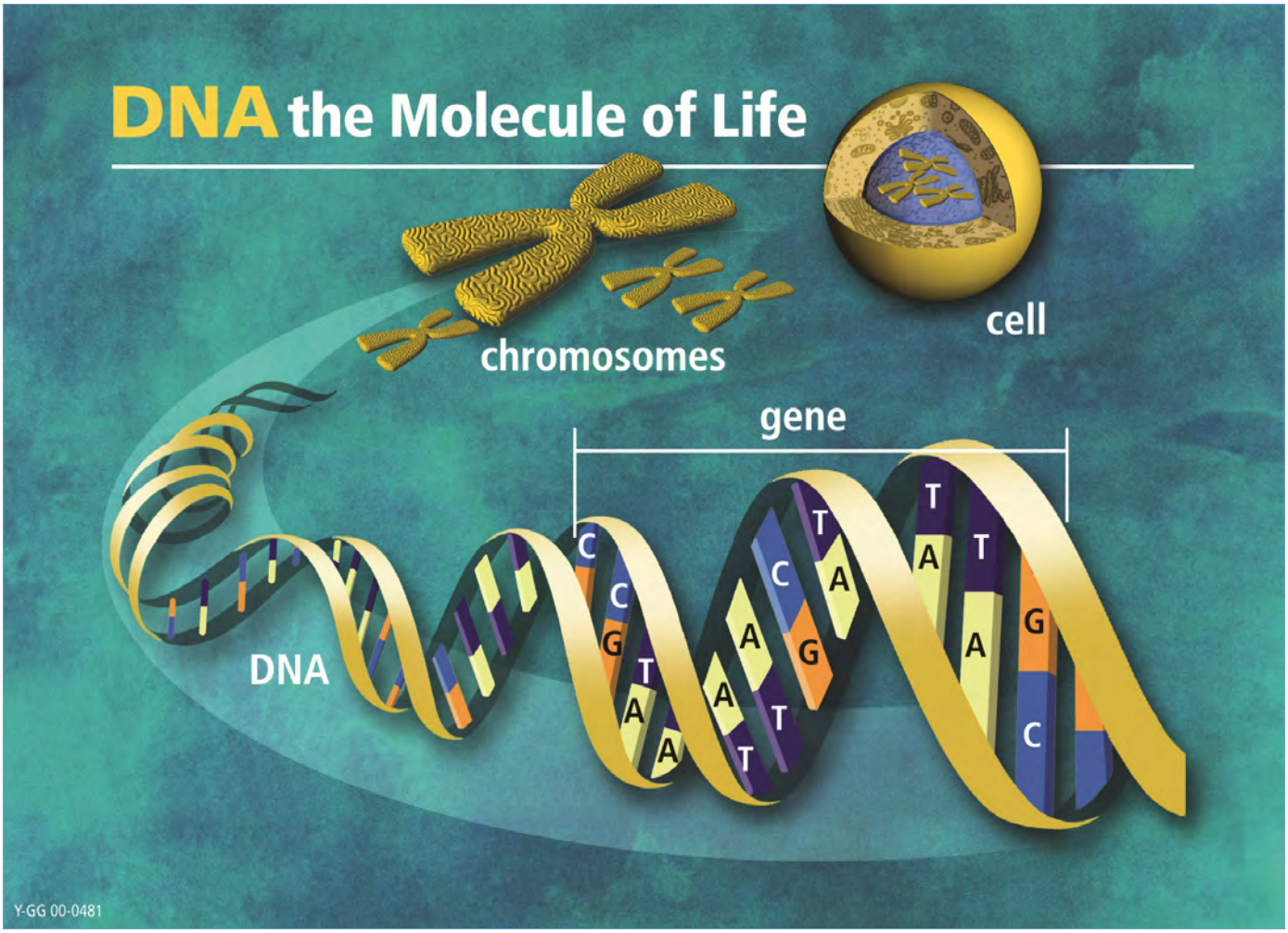
Source: National Human Genome Research Institute. http://en.wikipedia.org/wiki/File:Down_Syndrome_Karyotype.png#file

hearing loss is nonsyndromic (Reardon, Toriello, Downs, 2004). For a child with nonsyndromic hearing loss and a potential genetic etiology, the clinician would estimate recurrence risk - or the likelihood that future offspring in the family would be hearing impaired - based on the family history. Determining etiology was challenging for many nonsyndromic, hearing impaired infants because the parents were often normal hearing with no other significant family history, and prior to universal newborn hearing screening, the identification of hearing loss was usually delayed, with the age of onset based on parent report.

The Human Genome Project, jointly led by the U.S. National Institutes of Health and the Department of Energy between 1990 and 2003 (Human Genome Project Information, 2011) accelerated deafness gene discovery exponentially and launched a new era in the genetic evaluation of hearing loss. The massive work undertaken to sequence the entire human genome immediately prompted the discovery of many disease-related genes, particularly monogenic diseases like hearing loss that are caused by a defect in only one gene (Green, Guyer, & National Human Genome Research Institute, 2011; Jimenez-Sanchez, Childs & Valle, 2001; OMIM, 2011; VanCamp & Smith, 2011). (See Figure 2).

Figure 2. Human Chromosomes: composed of DNA organized into Genes

Human DNA is combined with other molecules and arranged into 46 chromosomes. The DNA strand on each chromosome contains many genes. Each DNA strand is a large molecule containing repeating nucleotide bases: **A**denine, **T**hymine, **C**ytosine, **G**uanine (the four letters of the DNA code A, T, C, G). Each gene has a specific sequence of bases (A,T,C,G) that provide the cell with instructions on how to construct a protein.

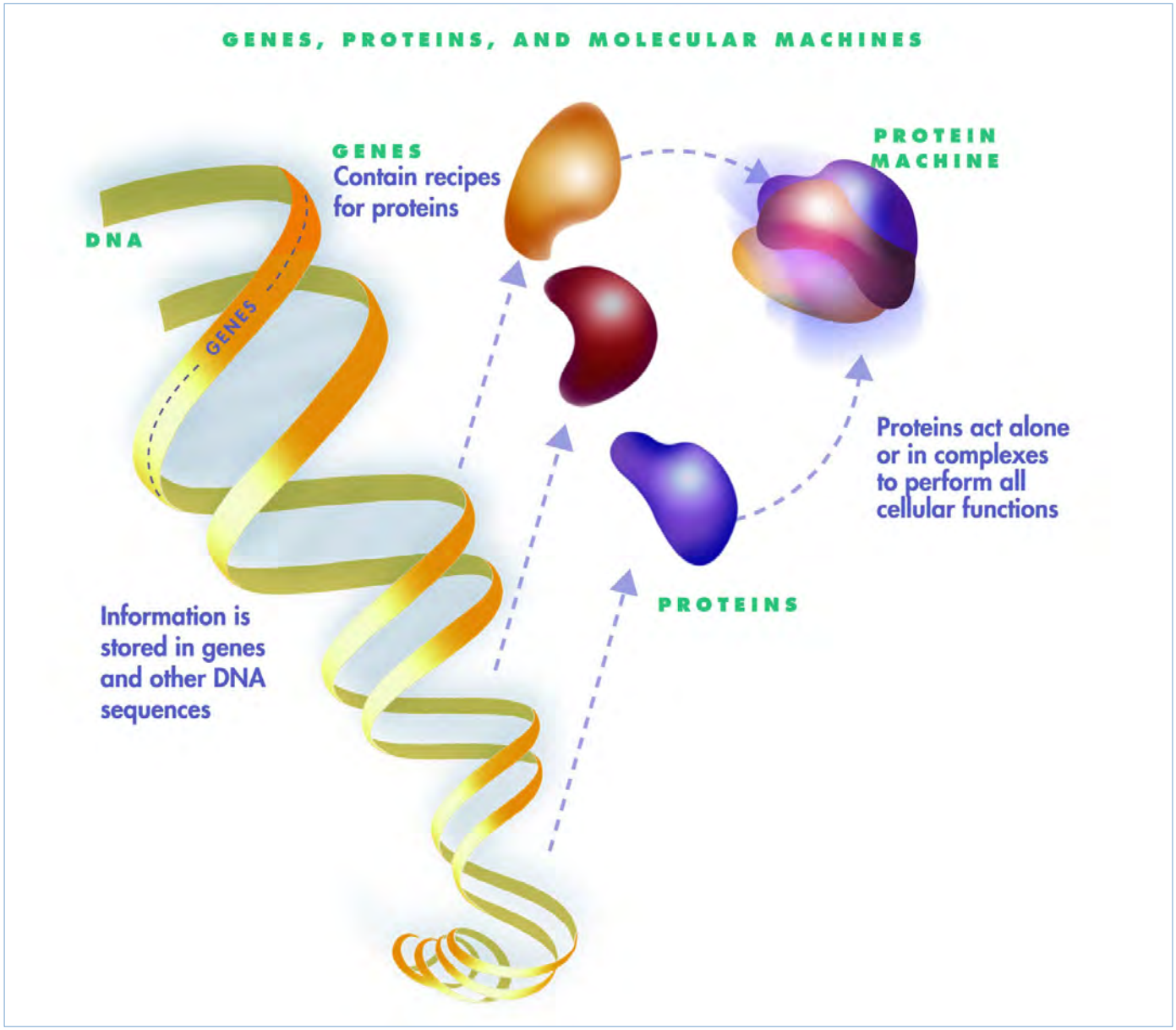


Source:
Office of Biological and Environmental Research of the U.S. Department of Energy Office of Science. http://science.energy.gov/ber/
Prepared by: The Biological and Environmental Research Information System, Oak Ridge National Laboratory. http://genomicscience.energy.gov/ and http://genomics.energy.gov/

Following the sequencing of the human genome, genetic linkage analysis was a key strategy for deciphering which of the 22-25,000 human genes was responsible for hearing loss, a technique still used for deafness gene discovery today. Genetic linkage analysis is accomplished by mapping genetic variance in large families affected by the disease of interest. First, the chromosome which houses the responsible gene is identified by comparing DNA between normal hearing and deaf family members, and the location of the suspect gene is narrowed down to a specific region, or 'locus" on this chromosome. Candidate genes residing within this chromosomal "address" are then explored (see Figures 3 to 6) by searching for DNA nucleotide changes – differences in the DNA nucleotide "letter" code - within the locus. A DNA coding change must then be evaluated to determine if it is actually linked to the presence of disease in the affected family members. Consider the example of a large family with inherited hearing impairment: once

Figure 3. Genes, Proteins and Molecular Machinery

Humans, like other living organisms are composed largely of proteins. Proteins provide the structural elements of cells and tissues, and enzymes for essential cell functions. Proteins are large, complex molecules made up of amino acid subunits organized into a chain. A gene contains the instructions for organizing amino acids into a sequence to form a protein.



Source:
Office of Biological and Environmental Research of the U.S. Department of Energy Office of Science. http://science.energy.gov/ber
Genomes to Life Program Roadmap, April 2001, DOE/SC-0036, U.S. Department of Energy Office of Science. http://genomicscience.energy.gov/
Prepared by: The Biological and Environmental Research Information System, Oak Ridge National Laboratory. http://genomicscience.energy.gov/ and http://genomics.energy.gov

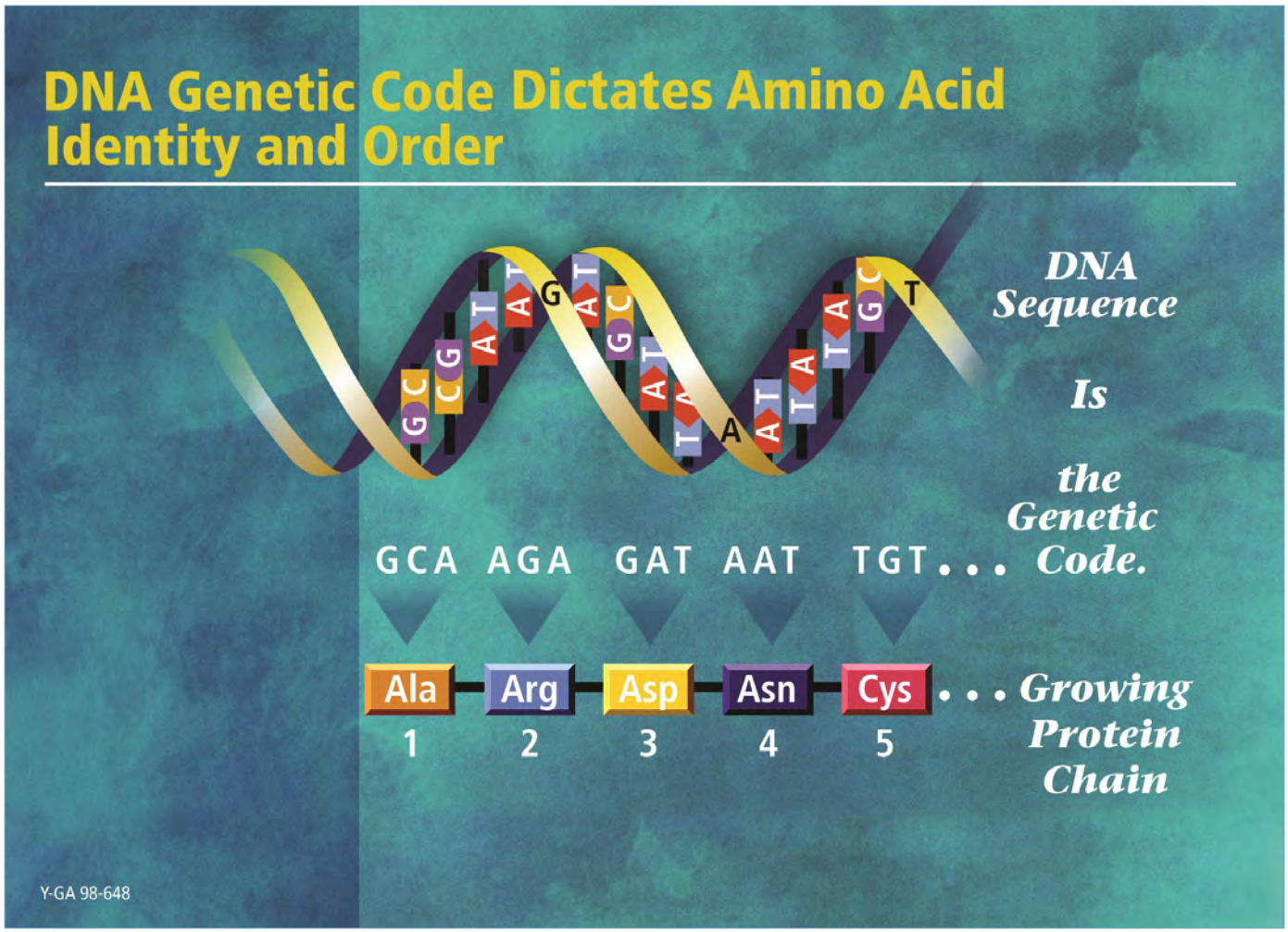
a potential deafness gene locus is identified, the specific DNA sequence of family members with hearing loss is compared to those with normal hearing (Young et al., 2001). Changes in a gene's DNA sequence that are only present in affected individuals may alter the genetic code in a way that changes the protein produced by the gene (see Figures 4 and 5). If this is the case, and the altered protein product is detrimental to human biology, disease may ensue.

This exclusively genetic approach, focusing on individual genes and disease caused by their mutations, was painstakingly slow and insufficient to identify the cause of multifactorial traits, or to account for the interaction of genes. Genomics, the study of an organism's entire set of genes in the context of their environment, opens a new frontier in human biology that will facilitate the exploration of how genes interact with each other and with other factors such as environmental exposure. Now, in addition to the discovery of disease-causing single gene defects, many new techniques such as genome-wide linkage and association studies are used to examine the entire genome in a family or population of interest (Chial, 2008). With this revolution in molecular genetics, the number of diseases suitable for genetic testing continues to grow.

Deafness Gene Discovery and Hearing Health Care – Where Are We Now?

The convergence of universal newborn hearing screening with extensive research following hearing impaired families,

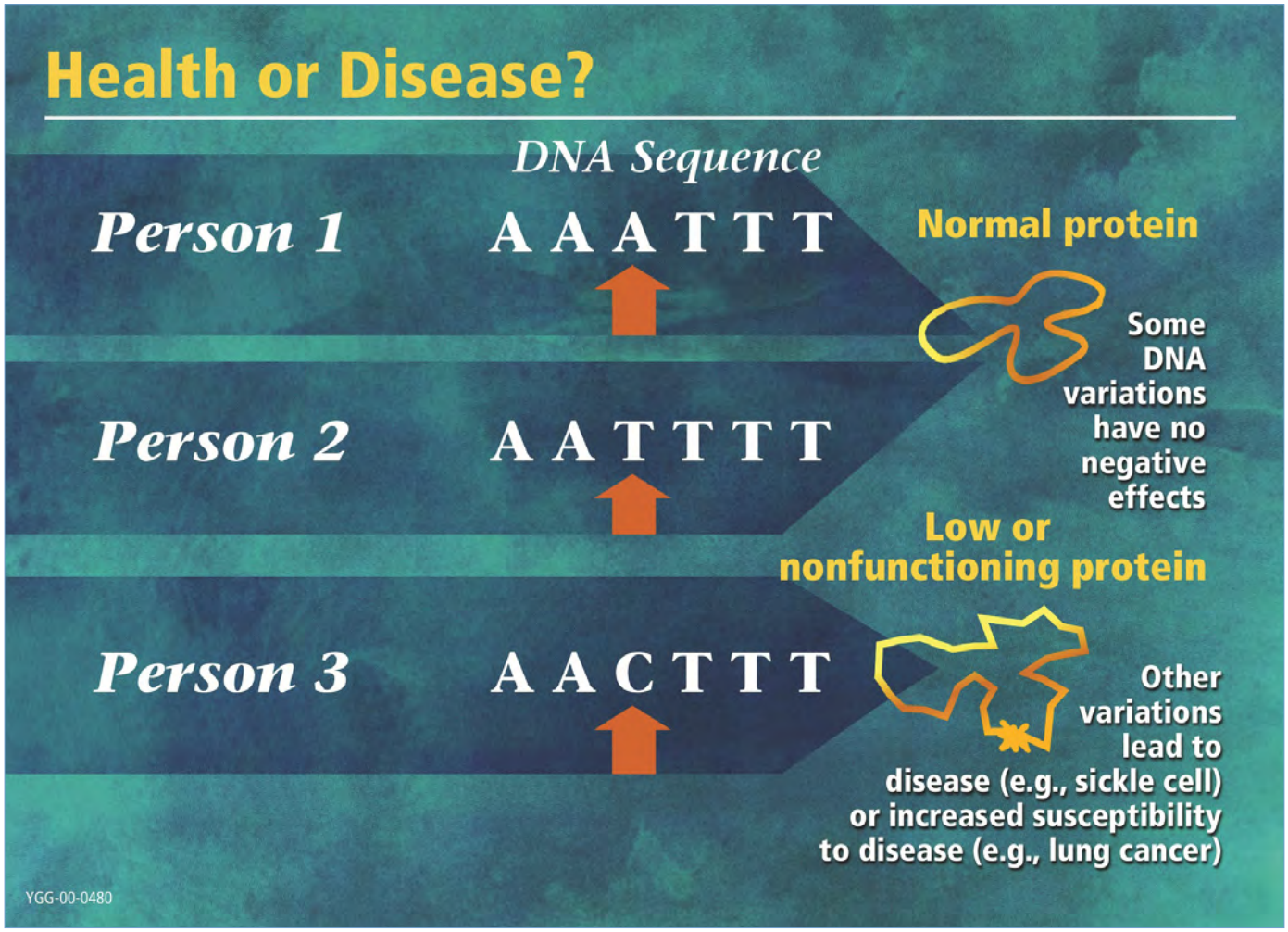
Figure 4. DNA and the Genetic Code
The sequence of nucleotide bases (A,T,C,G) provides the code which a cell uses to produce a protein. Proteins are composed of subunits, called amino acids, which are arranged in a sequence like beads on a string. Within every gene, each specific sequence of three DNA bases (codons) directs the cell to add specific amino acids and manufacture the gene-specific protein product. The DNA base sequence specifies the number and order of amino acid subunits that form the final protein.



Source:
Office of Biological and Environmental Research of the U.S. Department of Energy Office of Science. http://science.energy.gov/ber/
Prepared by the Biological and Environmental Research Information System, Oak Ridge National Laboratory. http://genomicscience.energy.gov/ and http://genomics.energy.gov/

Figure 5. Variations in DNA Sequence Cause Normal Variation and Disease

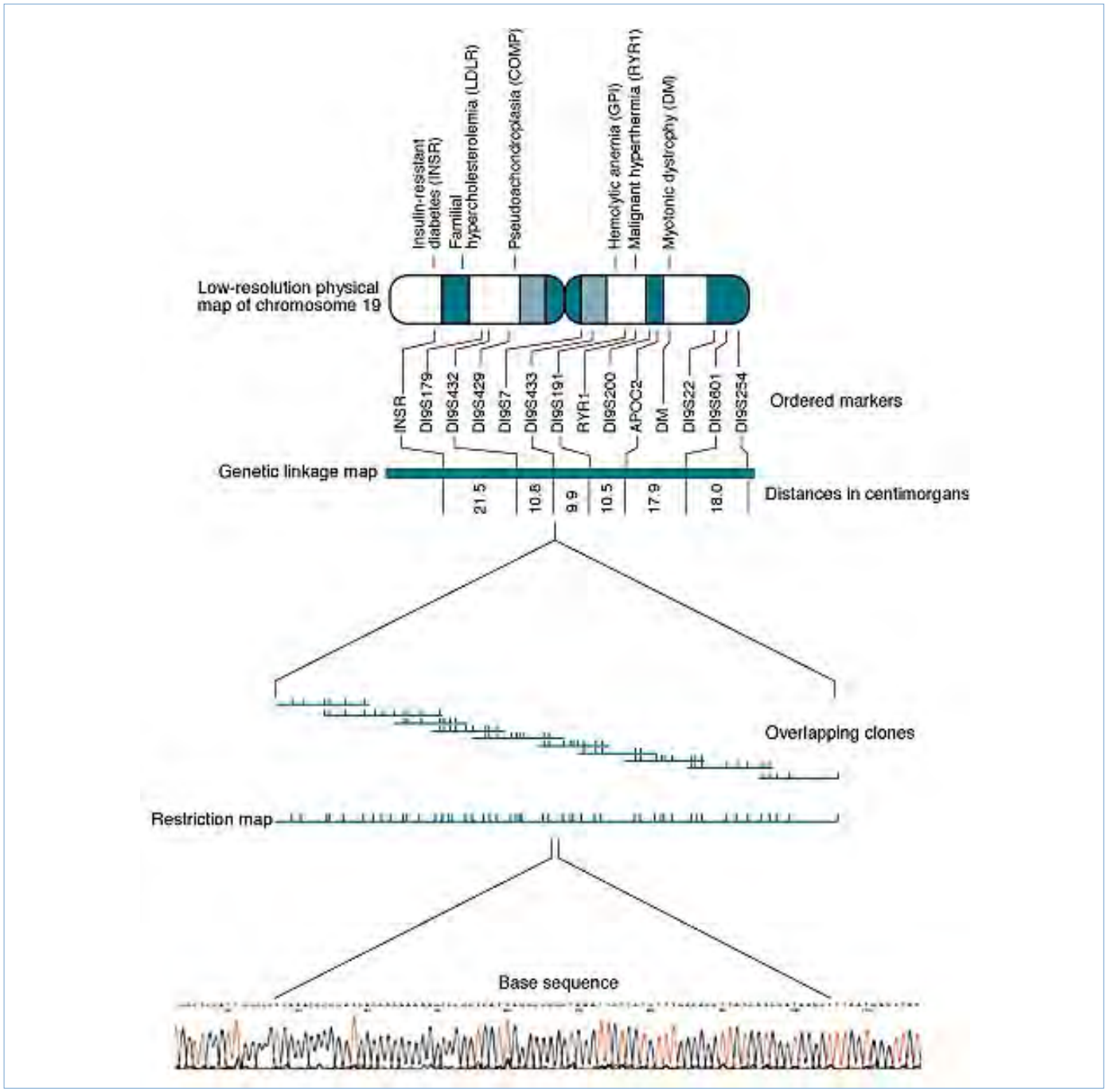
An individual's genome is their complete set of DNA. Most variations in DNA are subtle and require a close analysis of the DNA molecule to find changes in the nucleotide bases and their related codons – the "letters" and "3-letter words". Many of these subtle changes are considered normal variation and have little or no effect on the protein that is produced. Others cause different proteins that account for the normal variation in our physical traits or phenotype (e.g. hair colour). However, some variations in the genetic code cause changes in a protein which then causes a specific disease or leads to an increased susceptibility to a disease. Hereditary hearing loss often results from a subtle change in the genetic code of just one gene. There are many different types of hereditary hearing loss, each with its own specific gene defect, making this a heterogeneous group of monogenic disorders.



Source:
Office of Biological and Environmental Research of the U.S. Department of Energy Office of Science. http://science.energy.gov/ber/
Prepared by: The Biological and Environmental Research Information System, Oak Ridge National Laboratory. http://genomicscience.energy.gov/ and http://genomics.energy.gov/

Figure 6. Gene Discovery and Gene Mapping

Genetic mapping is a method used to identify the chromosome that contains the gene and determine precisely where it lies on that chromosome.Genetic maps have been used successfully to find the single gene responsible for many forms of inherited hearing loss. To produce a genetic map, researchers collect blood or tissue samples from family members where a certain disease or trait is prevalent. Unique patterns of DNA bases, seen only in family members with hearing loss, are used as markers to guide the search. Before researchers can identify the gene responsible for the hearing loss, they must hone in on the approximate location of the suspect gene. DNA markers are used to find the general location of the gene – which chromosome and roughly where the gene is located on the chromosome. If a particular gene is close to a DNA marker, the gene and marker usually stay close together on the chromosome, as the DNA is passed from parent to child. So, if each family member with hearing loss also inherits a particular DNA marker, it is likely that the gene responsible for the hearing loss lies near that marker. In this way the gene is located, and the DNA sequence of the gene is identified.



Source:

Office of Biological and Environmental Research of the U.S. Department of Energy Office of Science. http://science.energy.gov/ber/

Human Genome Program, U.S. Department of Energy, Human Genome Program Report, 1997. http://web.ornl.gov/sci/techresources/Human_Genome/publicat/97pr/index.shtml

Prepared by: The Biological and Environmental Research Information System, Oak Ridge National Laboratory. http://genomicscience.energy.gov/ and http://genomics.energy.gov/

and publication of The Human Genome Project outcomes, has radically changed our understanding of inherited hearing loss. These combined efforts have fueled the golden age of "deafness" gene discovery, and revolutionized hearing science by elucidating the structure and function of the normal and impaired cochlea on a molecular level. Hearing impaired patients and their health care providers are one of the largest groups to directly benefit from these scientific advances in genetics during the past few decades (Hilgert, Smith & Van Camp, 2009a; Korf, 2000; Matsunaga, 2009; Morton & Nance, 2006; Nance, 2003; Petit, 2006).This is because permanent childhood hearing loss is a common sensory disorder, with genetic causes accounting for more than half of the pediatric population, and because only one gene, from a long list of potential causative genes, is largely responsible for an individual's hearing loss. This genetic heterogeneity of hearing loss was discovered via revolutionary genetics and genomics techniques, and now, despite this genetic heterogeneity, clinical tests are available which evaluate 74 "deafness" genes in order to identify the gene responsible for a given patient's hearing impairment (Eppsteiner, Shearer, Hildebrand, Taylor, et al., 2012;Eppsteiner, Shearer, Hildebrand, Deluca et al., 2012). Although such clinical testing is currently expensive, costs are declining rapidly.

In industrialized nations with modern health care, the environmental causes (e.g. infection) of congenital hearing loss are reduced, with genetic causes responsible for more than half (50 - 68%). Prevalence estimates of genetic hearing loss vary with the criteria used to define hearing loss, genetic methods used to define the etiology, the age range considered, and other characteristics of the study population such as country of origin (Cryns & Van Camp, 2004; Kochhar, Hildebrand, Smith, 2007; Morton and Nance, 2006; Reardon et al., 2004; Smith & Taggart, 2004; Van Camp, Willems & Smith, 1997). Furthermore, individuals with a specific form of genetic hearing impairment (autosomal dominant – see below) typically present with delayed onset and/or progression, and are not accounted for in most prevalence estimates derived from prelingually hearing impaired populations. In Canada, the incidence of hearing loss in the newborn population has not been studied, and prevalence estimates of genetic hearing impairment are limited; the only patient population studies evaluating the genetics of nonsyndromic hearing loss have focused on the cochlear implant population (Hochman et al., 2010; Propst, Stockley, Gordon, Harrison, & Papsin, 2006).

Deafness related genes and loci are named according to their Mendelian Inheritance pattern.

Many different deafness genes have been identified, and Mendelian inheritance patterns for nonsyndromic hearing losses have been classified for each gene locus (Van Camp & Smith, 2011). (Note that genetics researchers coined the term "deafness" genes: this convention is observed throughout the paper, but the term refers to all types and degrees of hearing loss associated with genes and gene loci). Although inheritance patterns can be complex, particularly in isolated populations (Young et al., 2001) or in the Deaf community (Blanton et al., 2010), typical patterns of autosomal recessive, autosomal dominant, X-linked, Y-linked and mitochondrial inheritance of hearing loss have been delineated. Autosomal recessive hearing loss is frequently congenital or prelingual, and accounts for the majority (59-77%) of children with nonsyndromic permanent hearing loss. Two copies of the defective gene, one from each parent, must be inherited for hearing loss to be expressed in the individual, meaning that each parent, with only one mutated copy of the culprit gene, is unaffected. A smaller, but significant proportion (22-36%) of children with a genetic hearing loss inherit a single defective gene from one of their parents, who is most often hearing impaired too. In this situation a normal version of the gene, or normal allele, inherited from the other parent, is not sufficient to support normal hearing. This form of hearing loss is therefore considered dominant, and also autosomal, because the aberrant gene is located on one of the non-sex chromosomes. Autosomal dominant hearing losses are often progressive. Postlingual onset is considered typical but it should be noted that documented age of onset and progression of early hearing loss is often based on parental report rather than hearing threshold measurements. Universal newborn hearing screening and early diagnostic programs combined with genetic research will provide invaluable documentation of the natural course of early hearing loss with different genetic etiologies. The remaining <5% of the population affected with permanent hearing loss are accounted for by rare X-chromosome, Y chromosome, and mitochondrial genes with more complex inheritance patterns (Cryns & Van Camp, 2004; Hilgert, Smith & Van Camp, 2009a; Hilgert, Smith & Van Camp, 2009b; Morton & Nance, 2006; Van Camp et al., 1997).

A few deafness genes are relatively common and clinical genetic testing is available in Canada. In the late 1990s, the discovery that mutations in one gene, *GJB2*, are a frequent cause of nonsyndromic recessive hearing loss prompted the development and implementation of cost-efficient clinical testing that was serendipitously feasible because screening for mutations and full sequencing were relatively inexpensive for this small gene with only 2 exons. Diagnostic testing in conjunction with early detection and intervention programs for hearing impairment were proposed by Morton and Nance in 2006, and new predictions suggest that universal newborn genetic screening could be implemented by the next decade (Hochman et al., 2010; Propst, Stockley et al., 2006). Because mutations in GJB2 are fairly common worldwide, affecting

between 30-50% of those with nonsyndromic hearing loss, clinical tests are now available in most clinical genetics facilities (GeneTests., 2011). Canadian audiologists may be quite familiar with genetic testing for hearing loss caused by abnormal connexin 26 proteins that form cochlear gap junction channels, but less so with a related type of hearing loss caused by another gap junction protein, connexin 30. The terms "connexin 26 gene" and "connexin 30 gene" are commonly used, even by geneticists, but are imprecise – the correct terminology for these connexin genes are *GJB2* and *GJB6*, respectively. Genetic testing protocols for both genes are available in Canada. Clinical testing is also available at selected facilities for a small subset of additional deafness genes because they are also relatively common (e.g., *SLC26A4* related to Pendred syndrome and Enlarged Vestibular Aqueduct), or involve a syndrome which is difficult to diagnose and/or progressive (e.g., Usher Syndrome) (GeneTests, 2011; Hilgert, Smith,Van Camp, 2009a). Genetic testing can improve clinical efficiency; for example, the early diagnosis of a genetic etiology can streamline the process, with fewer specialty referrals and laboratory tests, and provide early detection and management for patients with late-onset or progressive syndromes (eg. Usher or Pendred). With respect to genetic counseling, genetic testing can improve recurrence risk estimates (Cryns & Van Camp, 2004; Matsunaga, 2009; Morton, 2002; Morton & Nance, 2006). Knowledge of specific auditory deficits and characteristic phenotypes associated with specific mutations can be translated to improved clinical care, including early detection in at-risk relatives, and greater predictability of onset, progression, and efficacy of various treatment strategies. Furthermore, understanding the genetic etiology caninform prognosis, especially for families with GJB2 nonsyndromic recessive hearing loss or mutations in other genes expressed in the sensory structures of the cochlea. Cochlear implantation has proven to be successful for most children, especially those with a "cochlear" genetic etiology, whereas poorer outcomes may relate to a primarily neural dysfunction induced by a defective gene expressed primarily in the spiral ganglion (Eppsteiner, Hildebrand, Taylor et al., 2012; Fukushima et al.,2002; Propst, Papsin, Stockley, Harrison, & Gordon, 2006).

Inherited hearing loss is a heterogeneous disorder and an important public health issue in Canada. Although mutations in *GJB2* are a relatively common cause of genetic hearing loss worldwide, this gene still accounts for only 20-30% of individuals with nonsyndromic hearing loss (equivalent to 30-50% of those with autosomal recessive hearing loss). Collectively, defects in the many other rare "deafness" genes account for the remaining 70-80% of the population with non-syndromic hearing loss, making this an extremely heterogeneous sensory disorder. Over 65 genes for non-syndromic hearing loss with different inheritance patterns have now been identified, and 50 additional non-syndromic "deafness" gene loci have been mapped, with the specific genes yet to be identified. Together the known deafness genes and loci (>130 total for both syndromic and non-syndromic hearing loss) extend across all 22 human autosomes and both the X and Y sex chromosomes. For many of these genes, different mutation-specific types of change in the DNA sequence can occur within the same gene. Like other monogenic diseases, a mutation in any one of these genes is generally rare, with any single "deafness" gene accounting for a small proportion of the hearing impaired population (Hilgert, Smith & Van Camp, 2009a; Shearer et al., 2010; Van Camp & Smith, 2011).

Scientific advances associated with gene discoveries in hearing and deafness research are now recognized as having major public health significance internationally (NIDCD, 2011) and in Canada (CHR, 2011). Gene mapping, linkage studies and gene mutation studies have led to "deafness" gene discoveries in Canada, with relevance to the clinical management of multiethnic and historically isolated populations in the country (Young et al., 2001; Propst, Stockley et al., 2006). Despite this exponential progress in research, changes in hearing health care service delivery have been modest to date because clinical tests for most of these rare genes are not routinely performed or widely available (GeneTests, 2011; Hilgert, Smith,Van Camp, 2009a). Currently, routine clinical genetic protocols and technology require that genes be analyzed individually and sequentially, requiring significant time and cost. The choice of gene(s) to be tested becomes critical, and is usually guided by the clinical phenotype and ethnicity of the individual. Canada is similar to most industrialized nations, in that only a few clinical genetic tests for the most common deafness genes are routinely available through clinical molecular genetic laboratories. Genetics services are still provided almost exclusively by specialists, medical geneticists and genetic counselors, with very limited involvement by primary care or allied health providers. Thus access togenetic testing depends on the availability of specialized clinical services which involve a medical genetic evaluation, interpretation of the genetic test results, and genetic counselling for patients and their families. Rapid advances in deafness gene discovery have caused an explosion of information about the genetic bases of hearing loss, and it is difficult for clinicians, even geneticists and hearing health care specialists, to keep up (Burton et al., 2006; Korf, 2000).

Deafness Gene Discovery – Implications for the Future of Hearing Health Care in Canada

Genetic tests have revolutionized our understanding of human cochlear structure, function and pathobiology.

The cochlea, buried within the temporal bone, has until recently been inaccessible for detailed analysis in living humans. With genetic tests we now have a non-invasive molecular toolbox which opens a window into the temporal bone, allowing us to probe the cells and molecules of the inner ear and auditory system of our patients. Molecular techniques allow us to define the nature and site of an auditory system lesion at the cellular-molecular level in patients with a positive genetic test result. Permanent hearing loss is currently defined by the clinical features, or phenotype - as conductive, mixed or sensorineural, for example. We now realize that hearing loss, particularly inherited sensorineural loss, is not a homogeneous disorder, but rather a collection of different ear pathologies caused by many rare genes. What does this mean for genetic test outcomes for our patients? The majority of individuals with an inherited nonsyndromic loss harbour a rare gene mutation affecting a single gene which is not identified by clinical tests. This is because routine clinical genetic tests evaluate only the most common deafness genes (eg. *GJB2*).Unfortunately, most patients do not have a common genetic defect; a wide array of rare deafness genes must be considered, and the DNA subjected to multiple gene-specific tests, in order to pinpoint each individual patient's specific gene defect. At present, patients with such rare gene mutations are usually identified through clinical research centres specializing in genetics and deafness (Harvard Medical School Centre for Hereditary Deafness, 2011; University of Iowa, 2011).

When the precise genetic etiology has been identified for a patient, knowing the defective gene and the type of mutation can provide a detailed appreciation of their underlying pathology. Genes that are active in the cochlea express a wide variety of protein products that together form unique molecular structures necessary for normal inner ear function; these include structural molecules that support and connect the hair cell stereocilia, many types of channels for transduction and cell-cell communication, motor molecules in hair cells, to name just a few. Consequently, a person harbouring a mutation in one of these critical genes may produce a defective protein molecule, or no protein at all, which in turn disrupts the structure or function of cells and tissues critical for hearing (cochlear cells and/or auditory neurons) (Friedman and Griffith, 2003; Hilgert, Smith & Van Camp, 2009b; Matsunaga, 2009; Morton, 2002; Morton & Nance, 2006; Petit, 2006; Shearer et al., 2010; Van Camp & Smith, 2011).Table 1 provides some examples of known deafness-causing genes expressed in the cochlea, and the associated cochlear functions that are disrupted by the faulty proteins produced as a consequence of these gene mutations. A comprehensive, regularly updated list of deafness genes and loci, categorized by inheritance pattern, and a diagram of where these genes are expressed in the cochlea, can be found at http://hereditaryhearingloss.org. Several excellent reviews are also available to the reader interested in different genetic etiologies and the underlying pathophysiologic mechanisms (Ealy & Smith, 2010; Hilgert et al., 2009b; Manchaiah, Zhao, Danesh, & Duprey, 2011; Richardson, de Movel, Petit, 2011).

Genetic testing technologies are evolving rapidly, allowing for simultaneous testing of multiple deafness genes.

Experts predict that genome-based approaches will improve the early detection, diagnosis and prevention of many diseases. In this future genomics era, clinical care will be guided by each patient's unique molecular biology in addition to their phenotype, and their diseases classified according to the underlying pathobiology. Genomic testing - analyzing an individual's entire genetic makeup - will not make significant inroads into daily health care practice until at least the next decade. However, the ability to test a subset of multiple genes simultaneously rather than consecutively is now available in specialized clinical research centres. New genetic testing protocols, employing rapidly evolving high-throughput technologies, will make simultaneous testing for multiple deafness genes feasible and cost-effective in the relatively near future, and long before genome-wide testing becomes commonplace (University of Iowa Health Care, 2011). Clinical protocols for testing a wide spectrum of deafness genes in the same patient will revolutionize the diagnostic assessment of hearing impairment. Technological advances can foster radical shifts in the diagnostic protocols used in audiology. Consider the detection of retrocochlear lesions, first with the advent of the Auditory Brainstem Response (ABR), and again as magnetic resonance imaging became widely available. Likewise, the application of otoacoustic emission techniques in the clinic was instrumental in the identification of a new category of hearing loss: auditory neuropathy spectrum disorders. In the future, the genetic etiology of hearing loss and site of lesion classification will be greatly refined as testing for all known "deafness" genes becomes cost-effective and more widely accessible. A revised classification system for patients with inherited hearing loss will be feasible with the implementation of these new methodologies. Apatient's site of lesion may be defined as a defect in the transducer or ion channels of the hair cell stereocilia (Schultz et al.,2005) or a pathology causing cochlear conductive hearing loss due to aberrant biomechanical properties of the tectorial membrane (Plantinga et al., 2007). Re-categorization based on the causative genetic etiology and underlying pathophysiology is now within reach, with inherited hearing loss falling into many different subgroups based on

Table 1. Nonsyndromic Hearing Loss Genes: Protein Products and Function

Gene & Locus Names	Protein Product	Chromosome Location	Protein Function: Cochlear Expression
Electrical & Metabolic Coupling in the Cochlea			
GJB2 DFNB1	connexin 26	13q12.11	Along with other connexins, Cx26 proteins form cochlear gap junction channels that connect adjacent cell membranes. Gap junctions allow transportation and recycling of small molecules and ions through nonsensory epithelial and connective tissue cell networks in the cochlea.
Inner & Outer Hair Cell Structure & Function			
WHRN DFNB31	whirlin	9q32	The α-tectorin protein is an important, non-collagenous component of the tectorial membrane, a structure overlying the hair cell stereocilia.
MYO7A DFNB2	myosin 7A	11q13.5	Myosin VIIA is an unconventional myosin involved in different cell functions including stereociliar development and stability, as well as vesicle trafficking and endocytosis in the hair cell body.
Inner Hair Cell-Auditory Afferent Synapse			
OTOF DFNB9	otoferlin	2p23.3	Together with other molecules, the otoferlin protein facilitates the release of neurotransmitter at the synapse connecting the hair cell and auditory neuron.

For detailed information about individual genes, mutations and related hearing disorders see the Hereditary Hearing Loss Homepage http://hereditaryhearingloss.org.

Gene expression in cochlear structures can also be viewed at http://hereditaryhearingloss.org/main.aspx?c=.HHH&n=86597

For excellent reviews of genetic hearing disorders and "deafness genes" see: Hilgert et al., 2009a; Manchaiah et al., 2011; Smith et al., 2010.

the auditory structures and cells where the defective gene is expressed and exerting a detrimental effect (Hilgert, Smith and Van Camp, 2009a; Hilgert, Smith and Van Camp, 2009b; Shearer et al., 2010; University of Iowa Health Care, 2011).

Common, acquired types of hearing loss, associated with aging, noise, or other types of disease, also have a genetic component. Mapping human genetic variation involves evaluating the entire genetic makeup, or genome, of many healthy individuals across different human populations (The 1000 Genes Project Consortium, 2010; The International HapMap 3 Consortium, 2010). Information about more common gene variants is supplementing the growing database of rare gene mutations associated with human disease. Through these efforts investigators have learned that most diseases have a genetic component. Common diseases are more complex than rare monogenic disorders, often involving one or more genes interacting with environmental triggers. Genome Wide Association Studies (GWAS) are now addressing these multifactorial disorders, including complex systemic conditions like diabetes that increase the risk for developing sensorineural hearing impairment (Green et al., 2011; The 1000 Genes Project Consortium, 2010; The International HapMap 3 Consortium, 2010). Many forms of acquired hearing loss are in fact common, multifactorial disorders that involve predisposing genes. Genetic susceptibility to aging and ototoxins like noise is well established, but for humans, the contributing genes are for the most part unknown (Davis et al.,2001; Davis, 2003; Frisina, 2009; Konings, Van Laer Van Camp 2009; Van Eyken, Van Camp, Van Laer, 2007). Gene identification and investigations of complex gene-gene and gene-environmental interactions are progressing at a rapid pace in humans and animal models (Johnson, Zheng, Noben-Trauth, 2006; McHugh, Friedman 2006; Schultz et al., 2005;

Yan & Liu, 2010). Now on the scene are human genome-wide studies, investigating nuclear genes, and also maternally inherited mitochondrial genes, that confer vulnerability to multifactorial auditory system diseases; some are bearing fruit, such as GMR7 associated with presbycusis (Friedman et al.,2009; Raimundo et al., 2012). Complex communication disorders affecting speech, language and acquired hearing are most often multi-factorial, and genomics approaches will be especially useful to their investigation. Advances in our understanding of acquired hearing loss, and the complex interactions between genetic and environmental contributions to pathobiology, are expected with genome-wide linkage and association studies.

How Will Audiological Practice Evolve in the New Molecular Era?

Many clinicians question whether a new assessment procedure, including genetic test protocols, is justified if the clinical outcomes are not altered by the test results. This is a valid concern. The translation of knowledge from the research laboratory into general clinical practice can involve many stages, and input from clinicians is essential in this process. As new deafness genes and mutations are identified, deep phenotyping – detailed profiling of behavioural, physiological, and imaging measures of auditory structure and function – will be critical to our understanding of how gene mutations affect human audition. Much can be gleaned from animal models, particularly mice, with similar gene defects, but ultimately, the pathophysiological repercussions of the molecular lesion caused by a specific gene mutation must be confirmed in humans. This is a crucial step in the translation of knowledge into the clinical realm, and one in which audiologists can play a key role.

A clear understanding of the causative molecular mechanisms will continue to streamline the diagnostic process and improve the early detection of progressive syndromes and nonsyndromic hearing loss as more genes are discovered and added to routine genetic testing protocols. Although not yet cost-efficient, universal screening and/or diagnostic testing of multiple genes and mutations, and incorporation of such protocols into early hearing detection and intervention programs will most likely be realized during the next decade (Linden Phillips et al., 2013; Wu et al., 2011). Furthermore, as genetic contributions to common diseases (e.g., presbycusis, noise-induced hearing loss, diabetes) are delineated, genetics-based care will become increasingly relevant to hearing health care practitioners, including audiologists. However, biomedical breakthroughs are the most dramatic when they lead to innovative new treatments for disease. Understanding the pathogenesis of hearing disorders will foster the development of molecular-level treatments and specialized biomedical devices, with implications for the treatment of both monogenic and more complex, acquired forms of hearing loss. Most audiologists would agree that patients with the same audiogram vary considerably in other respects, including their response to intervention. Armed with new insights into how genes affect different cochlear and auditory system structures and interact with environmental stressors to modify hearing loss, scientists can now investigate these inter-individual differences in clinical presentation. The links between an individual's genetic defect and specific underlying pathobiology and their responses to medication, amplification or cochlear implantation can now be explored. Pharmacogenomics may identify individual risks for ototoxicity, guide the development of preventative and treatment medications and modify the prescription process, with drug selection and dosage based on an individual patient's genotype (Green et al, 2011; Matsunaga, 2009). New gene-based therapies are now available for treating diseases like blindness, and pharmaceutical treatments influenced by genetic variation are under investigation for auditory disorders in animal models (Bainbridge et al., 2008; Davis et al.,2007). Eventually, this translational research will lead to a more tailored intervention process, and stimulate innovative and complementary modes of therapy in humans (e.g. gene delivery combined with neural prostheses) (Di Domenico et al., 2011; Hildebrand et al. 2008; Jan, Pereira, Turner, & Kotov, 2011).

With these advances on the horizon, the disadvantages of genetic and genomic testing and barriers to knowledge translation must be addressed. Before we can harness the power of genomics and translate this knowledge into meaningful clinical interventions that improve quality of life, genotype-phenotype relationships must be explored and the impact of these findings on the individual, their family, and society at large must be explored. We have at present a superficial knowledge of how hearing-related gene mutations influence human physiology, behaviour, and clinical presentation. The ethical issues of identifying unaffected gene mutation carriers, or those with a causative genotype who are asymptomatic at the time of genetic screening or testing must be confronted (Linden Phillips L, et al., 2013; Schimmenti et al., 2004; Wu et al., 2011). For genomic testing, the management of incidental findings – identifying mutations in genes not related to hearing loss – is critical for the patient and their family members. How will vulnerable groups or an individual's rights be protected with respect to the provision of health insurance? Could the identification of a gene mutation for hearing loss limit future vocational opportunities, for instance? With respect to knowledge translation, scientists, clinicians, and governments face an exciting but immense challenge (Zwart & Nelis, 2009). As genomic technologies continue to evolve,

genotyping is outpacing the comprehensive phenotyping of newly identified genes and mutations, and the social, legal, and ethical implications of these discoveries.

Summary & Concluding Remarks

In this paper, we present a brief introduction to basic genetic concepts, and an overview of the recent advances in genetics and genomics relevant to hearing health care. Personalized medicine, where each patient receives customized health care based on their unique genetic make-up, is the main goal of translational genomics, but more than a decade away (Green et al., 2011). However, as a consequence of the exponential progress in genetics and genomics research, a revolution in our understanding of normal auditory function and disease has emerged during the last two decades. Multiple deafness gene testing protocols are now available at clinical research sites, and rapidly evolving high-throughput technologies will continue to improve their time- and cost-efficiency (Shearer et al., 2010; University of Iowa Health Care, 2011). Noninvasive clinical tests for detecting mutated genes and their defective gene products – the proteins – will enable a precise molecular level definition of auditory system defects in an ever-increasing proportion of our patients.

Over the next few decades, genetics-based care will evolve beyond the domain of medical geneticists and genetic counsellors. As research advances are translated into the clinic, genetic specialists will eventually be overwhelmed by the demand for services and the breadth of disease-specific knowledge encompassed by the "new genomics". As multidisciplinary models become the standard for genetic service delivery, the translation of genomics breakthroughs into the clinical realm will depend on the acquisition of genetic competencies by the health care community (Bottorff et al., 2005; Burke et al. , 2002; Canadian Nursing Association, 2005; Carrol et al., 2009; Gurwitz, Weisman, Rehavi, 2003; Harvey, Stanton, Garrett, Neils-Strunjas & Warren, 2007; OBA, 2011). Recognized by government, research and professional agencies as a crucial issue, joint efforts have led to the development and implementation of health professional and public educational programs worldwide, including Canada, Europe, and the United States (e.g., EuroGentest, Core competencies for health professionals in Europe, 2011; GenEdProject, 2011; Genetics Education and Training, 2011; Genetics in the Practice of Speech Language Pathology and Audiology, 2011; HUGO, 2011; NCHPEG, 2011; OBA, 2011; WHO, 2011).

Supported by two decades of deafness gene discovery and rapidly evolving technology, hearing researchers and clinicians are poised to evaluate these molecular genetic innovations in the clinic. As the promises of personalized medicine are realized, the diagnosis, treatment and counseling of hearing impaired patients and their families will evolve accordingly. Eventually, genetics-related principles will infuse everyday clinical practice, affecting all health care professionals, including audiologists. Those who understand the genetic basis of hearing impairment will be well prepared to embrace these new developments, able to understand new diagnostic protocols and treatments, make appropriate referrals, and provide (re)habilitation and counseling to patients with concerns about the genetic basis of their hearing loss and implications for other family members.

Acknowledgments:

This work was supported by a CIHR grant to SGS (Award: 207714).

References

Bainbridge, J. W. B., Smith, A. J., Barker, S. S., Robbie, S., Henderson, R., Balaggan, K.,...Ali , R.R. (2008). Effect of gene therapy on visual function in Leber's Congenital Amaurosis. *New England Journal of Medicine, 358*, 2231-2239.

Blanton, S. H., Nance, W. E., Norris, V. W., Welch, K. O., Burt, A., Pandya, A., & Arnos, K. S. (2010). Fitness among individuals with early childhood deafness: Studies in alumni families from Gallaudet University. *Annals of Human Genetics, 74*(1), 27-33.

Bottorff, L. J., Blaine, S., Carroll, C. J., Esplen, J. M., Evans, J., Nicolson Klimek, L.M.,...Ritvo, P. (2005). The educational needs and professional roles of Canadian physicians and nurses regarding genetic testing and adult onset hereditary disease. *The Journal of Community Genetics, 8*(2), 80-87.

Burke, W., Acheson, L., Botkin, J., Bridges, K., Davis, A., Evans, J.,...Wilson, M. (2002). Genetics in primary care: A USA faculty development initiative. *The Journal of Community Genetics, 5*(2), 138-146.

Burton, S. K., Blanton, S. H., Culpepper, B., White, K. R., Pandya, A., Nance, W. E.,& Arnos, K.S. (2006). Education in the genetics of hearing loss: A survey of early hearing detection and intervention programs. *Genetics in Medicine, 8*, 510-517.

Canadians for Health Research (CHR). Retrieved from http://www.chrcrm.org/en/rotm/dr-terry-lynn-young

Canadian Nursing Association (2005). Nursing & Genetics: Are you ready? *Nursing Now: Issues and Trends in Canadian Nursing, 20*, 1-5.

Carrol, C. J., Rodeout, L. A., Wilson, J. B., Allanson, J., Blaine, M. S., Esplen, J. M.,...Taylor, S. (2009). Genetic education for primary care providers-Improving attitudes, knowledge, and confidence. *Canadian Family Physician, 55*, e93-99.

Canadian Institutes of Health Research (CIHR). (2010). Government of Canada boosts efforts to find treatments for pediatric cancers and rare genetic diseases. Retrieved from http://www.cihr-irsc.gc.ca/e/43211.html

Chial, H. (2008). Rare genetic disorders: Learning about genetic disease through gene mapping, SNPs, and microarray data. *Nature Education, 1*(1)

Cryns, K., & Van Camp, G. (2004). Deafness genes and their diagnostic applications. *Audiology and Neurotology, 9*, 2-22.

Davis, R. R. (2003). Genetic influences in individual susceptibility to noise: A review. *Noise Health, 5*, 19-28.

Davis, R. R., Kuo, M. W., Stanton, S. G., Canlon, B., Krieg, E., & Alagramam, K. N. (2007). N-acetyl L-cysteine does not protect against premature age-related hearing loss in C57BL/6J mice: A pilot study. *Hearing Research, 226*, 203-208.

Davis, R. R., Newlander, J. K., Ling, X., Cortopassi, G. A., Krieg, E. F., & Erway, L. C. (2001) Genetic basis for susceptibility to noise-induced hearing loss in mice. *Hearing Research, 155*, 82-90.

Di Domenico, M., Ricciardi, C., Martone, T., Mazzarella, N., Cassandro, C., Chiarella, G.,...Cassandro, E. (2011). Towards gene therapy for deafness. *Journal of Cellular Physiology, 226*, 2494–2499.

Ealy, M., & Smith, R. J. (2010). The genetics of otosclerosis. *Hearing research, 266*(1), 70-74.

Eppsteiner, R. W., Shearer, A. E., Hildebrand, M. S., Deluca, A. P., Ji, H., Dunn, C.C.,...Smith, R.J. (2012). Prediction of cochlear implant performance by genetic mutation: The spiral ganglion hypothesis. *Hearing Research. 292*, 51-58.

Eppsteiner, R. W., Shearer, A. E., Hildebrand, M. S., Taylor, K. R., Deluca, A. P., Scherer, S.,...Smith, R.J. (2012). Using the phenome and genome to improve genetic diagnosis for deafness. *Otolaryngology - Head & Neck Surgery. 147*, 975-957.

EuroGentest, Core competencies in genetics for health professionals in Europe Retrieved from http://www.eurogentest.org/professionals/info/public/unit6/core_competences.xhtml

Friedman, R.A., & Griffith, A. J. (2003). Human nonsyndromicsensorineural deafness. *Annual Review of Genomics and Human Genetics, 4*, 341-402.

Friedman, R. A., Van Laer, L., Huentelman, M. J., Sheth, S. S., Van Eyken, E., Corneveaux, J. J.,...Van Camp, G. (2009). GRM7 variants confer susceptibility to age-related hearing impairment. *Human Molecular Genetics, 18*, 789-796.

Frisina, R.D. (2009). Age-related hearing loss: Ear and brain mechanisms. Annals of the New York Academy of Sciences, 1170, 708-717.

Fukushima, K., Sugata, K., Kasai, N., Fukuda, S., Nagayasu, R., Toida, N.,... Nishizaki, K. (2002). Better speech performance in cochlear implant patients with GJB2-related deafness. *International Journal of Pediatric Otorhinolaryngology, 62*, 151-157.

GenEdProject Retrieved from http://www.medicine.manchester.ac.uk/gened/

GeneTests. GTR: Genetic testing registry. Retrieved from http://www.ncbi.nlm.nih.gov/sites/GeneTests/lab/disease/deafness?db=genetests&search_param=contains

Genetics Education and Training (2011, Feb). Report of the Secretary's Advisory Committee on Genetics, Health, and Society. Retrieved from http://oba.od.nih.gov/oba/SACGHS/reports/SACGHS_education_report_2011.pdf

Genetics in the Practice of Speech Language Pathology and Audiology. Retrieved from http://www.nchpeg.org/shla/site.html

Green, E. D., & Guyer, M. S., National Human Genome Research Institute. (2011). Charting a course for genomic medicine from base pairs to bedside. *Nature, 470*, 204-213.

Greendale, K., & Pyeritz, E.R. (2001). Empowering primary care health professionals in medical genetics: How soon? How fast? How far? *American Journal of Medical Genetics, 106*(3), 223-232.

Gurwitz, D., Weizman, A., &Rehavi, M. (2003). Education: Teaching pharmacogenomics to prepare future physicians and researchers for personalized medicine. *Trends in Pharmacological Sciences, 24*,122-125.

Harvard Medical School Centre for Hereditary Deafness. Retrieved from http://hearing.harvard.edu/index.htm

Harvey, E. K., Stanton, S., Garrett, J., Neils-Strunjas, J., & Warren, N. S. (2007). A case for genetics education: Collaborating with speech-language pathologists and audiologists. *American Journal of Medical Genetics A,143A*(14), 1554-1559.

Hildebrand, S., M., Newton, S. S., Gubbels, S. P., Sheffield, A. M., Kochar, A., de Silva, M. G.,...Smith, R. J. (2008). Advances in molecular and cellular therapies for hearing loss. *Molecular Therapy, 16*, 224–236.

Hilgert, N., Smith, J. R., & Van Camp, G. (2009a). Forty-six genes causing nonsyndromic hearing impairment: Which ones should be analyzed in DNA diagnostics? *Mutation Research, 681*, 189–196.

Hilgert, N., Smith, R. J., & Van Camp, G. (2009b). Function and expression pattern of nonsyndromic deafness genes. *Current Molecular Medicine, 9*, 546-564.

Hochman, J. B., Stockley, T. L., Shipp, D., Lin, V. Y., Chen, J. M., & Nedzelski, J. M. (2010). Prevalence of Connexin 26 (GJB2) and Pendred (SLC26A4) mutations in a population of adult cochlear implant candidates. *Otolology and Neurotology. 31*, 919-922.

HUGO (Human Genomic Organization). Committee for Education and Public Awareness in Genomics. Retrieved from http://www.hugo-international.org/comm_hugocommitteeforeducation.php

Human Genome Project. Retrieved from http://www.ornl.gov/sci/techresources/Human_Genome/project/about.shtml

Jan,E., Pereira, F. N., Turner, D. L., & Kotov, N. A. (2011). In situ gene transfection and neuronal programming on electroconductive nanocomposite to reduce inflammatory response. *Journal of Materials Chemistry, 21*, 1109-1114.

Jimenez-Sanchez, G., Childs, B., & Valle, D. (2001). Human disease genes. *Nature, 409*, 853-855.

Johnson, K. R., Zheng, Q. Y., & Noben-Trauth, K. (2006). Strain background effects and genetic modifiers of hearing in mice. *Brain Research, 1091*(1), 79–88.

Kochhar, A., Hildebrand, M. S., & Smith, R. J. (2007). Clinical aspects of hereditary hearing loss. *Genetics in Medicine, 9*, 393-408.

Konings, A., Van Laer, L., & Van Camp, G. (2009). Genetic studies on noise-induced hearing loss: A review. *Ear & Hearing, 30*, 151–159.

Korf, B. R. (2000).New genetics of hearing loss. *Teratology, 61*, 163–164.

Linden Phillips, L., Bitner-Glindzicz, M., Lench, N., Steel, K.P., Langford, C., Dawson, S.J., ...Packer, C. (2013). The future role of genetic screening to detect newborns at risk of childhood-onset hearing loss. *International Journal of Audiology, 52*(2), 124-133.

Manchaiah, V. K., Zhao, F., Danesh, A. A., & Duprey, R. (2011). The genetic basis of Auditory Neuropathy Spectrum Disorder (ANSD). *International Journal of Pediatric Otolaryngology, 75*, 151-158.

Matsunaga, T. (2009). Value of genetic testing in the otological approach for sensorineural hearing loss. *Keio Journal of Medicine, 58*, 216-222.

McHugh, R. K., & Friedman, R. A. (2006). Genetics of hearing loss: Allelism and modifier genes produce a phenotypic continuum. *The Anatomical Record, 288A*, 370-381.

Metcalfe, S., Hurworth, R., Newstead, J., & Robins, R. (2002). Needs assessment study of genetics education for general practitioners in Australia. *Genetics in Medicine, 4*(2), 71-77.

Morton, C. C. (2002). Genetics, genomics and gene discovery in the auditory system. *Human Molecular Genetics, 11*, 1229–1240.

Morton, C. C., & Nance, E. W. (2006). Newborn hearing screening—A silent revolution. *The New England Journal of Medicine, 354*, 2151-2164.

Nance, W.E. (2003). The Genetics of Deafness. *Mental Retardation and Developmental Disabilities Research Reviews, 9*, 109–119.

NCHPEG (National Coalition for Health Professional Education in Genetics. (2011). About NCHPEG). Retrieved from http://www.nchpeg.org/index.php?option=com_content&view=article&id=83&Itemid=57

NIDCD (National Institute of Deafness and Communication Disorders). (2010). Healthy hearing 2010: Where are we now? Retrieved from http://www.nidcd.nih.gov/health/inside/spr05/pg1.asp

OBA (Office of Biotechnology Activities). (2011). Genetics education and training: Report of the secretary's advisory committee on genetics, health, and society. Retrieved from http://oba.od.nih.gov/oba/SACGHS/reports/SACGHS_education_report_2011.pdf

OMIM (Online Mendelian Inheritance in Man). Retrieved from http://www.ncbi.nlm.nih.gov/omim

Petit, C. (2006). From deafness genes to hearing mechanisms: Harmony and counterpoint. *Trends in Molecular Medicine, 12*, 57–64.

Plantinga, R. F., Cremers, C. W. R. J., Huygen, P. L. M., Kunst, H. P. M., & Bosman, A. J. (2007). Audiological evaluation of affected members from a Dutch DFNA8/12 (TECTA) family. *Journal of the Association of Research in Otolaryngology, 8*, 1–7.

Propst, E. J., Stockley, T. L., Gordon, K. A., Harrison, R. V., Papsin, B. C. (2006). Ethnicity and mutations in GJB2 (connexin 26) and GJB6 (connexin 30) in a multi-cultural Canadian paediatric Cochlear Implant Program. *International Journal of Pediatric Otorhinolaryngology, 70*, 435-444.

Propst, E. J., Papsin, B. C., Stockley, T. L., Harrison, R. V., & Gordon, K. A. (2006). Auditory responses in cochlear implant users with and without GJB2 deafness. *Laryngoscope, 116*, 317-327.

Raimundo, N., Song, L., Shutt, T. E., McKay, S. E., Cotney, J., Guan, M. X.,... Shadel, G.S. (2012). Mitochondrial stress engages E2F1 apoptotic signaling to cause deafness. *Cell, 148*(4), 716-726.

Reardon, W., Toriello, V.H., & Downs, C. A. (2004). Epidemiology, etiology, genetic patterns and genetic counseling. In Toriello, V. H., Reardon, W., & Gorlin, J. R. (Eds.), *Hereditary Hearing Loss and its Syndromes* (pp. 8-16). New York, NY: Oxford University Press, Inc.

Richardson, G. P., de Monvel, J. B., & Petit, C. (2011). How the genetics of deafness illuminates auditory physiology. *Annual review of physiology, 73*, 311-334.

Schimmenti, L. A., Martinez, A., Fox, M., Crandall, B., Shapiro, N., Telatar M.,...Palmer, C. G. (2004). Genetic testing as part of the early hearing detection and intervention (EHDI) process. *Genetics in Medicine, 6*, 521–525.

Schultz,J.M., Yang, Y., Caride, A. J., Filoteo, A. G., Penheiter, A. R., Lagziel, A.,... Griffith, A. J. (2005). Modification of human hearing loss by Plasma-Membrane Calcium Pump PMCA2. *New England Journal of Medicine, 352*, 1557-1564.

Shearer, A. E., DeLuca, A. P., Hildebrand, M. S., Taylor, K. R., Gurrola, I. J., Sherer, S.,...Smith, R. J. H. (2010). Comprehensive genetic testing for hereditary hearing loss using massively parallel sequencing. *Proceedings of the National Academy of Sciences, 107*, 21104-21109.

Smith, S. D., & Taggart, R. T. (2004). Genetic hearing loss with no associated abnormalities. In Toriello, V. H., Reardon, W. & Gorlin, J. R. (Eds.), *Hereditary hearing loss and its syndromes* (pp. 37-82). New York, NY: Oxford University Press, Inc.

The 1000 Genes Project Consortium. (2010). A map of human genome variation from population-scale sequencing. *Nature, 467*, 1061-1073.

The International HapMap 3 Consortium. (2010). Integrating common and rare genetic variation in diverse human populations. *Nature, 467*, 52-58.

University of Iowa Health Care. (2011) UI consult. New test screens all deafness genes. Retrieved from http://medcom.uiowa.edu/uiconsult/new-test-can-screen-all-deafness-genes-at-once/

University of Iowa. (n.d.)Molecular Otolaryngology and Renal Research Laboratory. Retrieved from http://www.healthcare.uiowa.edu/labs/morl/index_CDS.htm

Van Camp, G., & Smith, J. R. H. Hereditary hearing loss homepage. Retrieved from http://www.hereditaryhearingloss.org

Van Camp, G., Willems, J. P., & Smith, J. R. (1997). Nonsyndromic hearing impairment: Unparalleled heterogeneity. *The American Journal of Human Genetics, 60*, 758-764.

Van Eyken, E., Van Camp, G., & Van Laer, L. (2007). The complexity of age-related hearing impairment: Contributing environmental and genetic factors. *Audiology and Neurootology, 12*, 345–358.

WHO (World Health Organization) Genomic Resource Centre. (?) Continuing education for health professionals. Retrieved from http://www.who.int/genomics/professionals/education/en/index.html

Wu, C. C., Hung, C. C., Lin, S. Y., Hsieh, W. S., Tsao, P. N., Lee, C. N.,...Hsu, C.J. (2011). Newborn genetic screening for hearing impairment: a preliminary study at a tertiary center. *PLoS One. 6*, e22314.

Yan, D., & Liu, X. L. (2010). Modifiers of hearing impairment in humans and mice. *Current Genomics, 11,* (4) 269–278.

Young, T. L., Ives, E., Lynch, E., Person, R., Snook, S., Maclaren, L.,...King, M. C. (2001). Non-syndromic progressive hearing loss DFNA38 is caused by heterozygous missense mutation in the Wolfram syndrome gene WFS1. *Human Molecular Genetics, 10*, 2509-2514.

Zwart, H., & Nelis, A. (2009). What is ELSA genomics?. *EMBO reports, 10*(6), 540-544.

Authors' Note

Correspondence concerning this article should be addressed to Susan G. Stanton, PhD, National Centre for Audiology, School of Communication Sciences and Disorders, Rm 2262L Elborn College, 1201 Western Rd., Western University, London, ON N6G 1H1 Canada. Email: stanton@nca.uwo.ca

Received date: November 7, 2011

Accepted date: March 29, 2013

Exploration de l'effet des variantes linguistiques sur les performances à une épreuve d'écoute dichotique chez deux populations francophones du Canada

Exploration of the effect of linguistic variations on a dichotic listening test in two Canadian francophone populations.

MOTS-CLÉS

TEST D'ÉCOUTE DICHOTIQUE

ENFANTS

DIFFÉRENCES LINGUISTIQUES RÉGIONALES

TROUBLE DE TRAITEMENT AUDITIF

DONNÉES NORMATIVES

Josée Lagacé
Véronique Doiron
Stéphanie Breau Godwin
Benoît Jutras

Josée Lagacé, Ph.D.
Professeure adjointe
et audiologiste
Université d'Ottawa
Ottawa, ON
Canada

Véronique Doiron, M.Sc.S.
Université d'Ottawa
Ottawa, ON
Canada

Stéphanie Breau Godwin, M.Sc.S.
Université d'Ottawa
Ottawa, ON
Canada

Benoît Jutras, Ph.D.
Professeur agrégé
Université de Montréal
Chercheur
Centre de recherche,
CHU Sainte-Justine
Montréal, QC
Canada

Abrégé

L'objectif principal de cette étude rétrospective était d'explorer l'effet des variantes linguistiques régionales sur les performances mesurées avec la version franco-canadienne du test d'écoute dichotique de mots - le *Staggered Spondaic Word (SSW)*. Pour ce faire, une comparaison des données normatives du test SSW établies au Centre hospitalier universitaire Dr-Georges-L.-Dumont de Moncton au Nouveau-Brunswick et au Centre hospitalier Rivière-des-Prairies de Montréal au Québec a été effectuée pour des enfants de six à dix ans ainsi que pour des adultes. L'analyse des résultats a révélé une différence significative entre les données normatives de ces deux régions canadiennes. Ces résultats appuient les recommandations quant à l'importance de développer des données normatives propres aux populations auprès desquelles les épreuves de perception auditive pré-enregistrées comptant des stimuli verbaux sont administrées.

Abstract

The main objective of this retrospective study was to explore the effect of the regional linguistic differences on the performance measured with the French adaptation of the *Staggered Spondaic Word (SSW)* Test. The normative data obtained with children from six to ten years old, as well as in adults, at Dr.-Georges-L.-Dumont University Hospital Centre, Moncton, New-Brunswick were compared to the data from the Rivière-des-Prairies Hospital, Montréal, Quebec. Data analysis revealed a significant difference between the norms from these two Canadian regions. Results from this study support recommendations regarding the importance of developing population specific normative data for evaluation of auditory processing using pre-recorded verbal stimuli.

Introduction

Dans la société francophone canadienne, les différences linguistiques régionales sont une réalité à laquelle les audiologistes doivent faire face. Par exemple, lorsqu'ils administrent une épreuve de perception auditive enregistrée comportant des stimuli verbaux, le caractère familier du vocabulaire employé et la prononciation des mots peuvent influencer la perception et par le fait même, la réponse de la personne évaluée. Aucune étude connue n'a examiné l'effet des différences linguistiques régionales sur les performances à des épreuves cliniques employées en audiologie. Les études publiées portent plutôt sur l'effet des différences culturelles entre nationalités et essentiellement pour des épreuves en anglais (Cameron, Barker, et Newall, 2003a; Dawes, 2011; Keith, Katbamna, Tawfik et Smolak, 1987; Marriage, King, Briggs et Lutman, 2001; Sockalingam et coll., 2004). L'ensemble des études révèle des différences dans les performances à diverses épreuves d'écoute de stimuli verbaux entre les populations de divers pays. Jusqu'à maintenant, on ignore si les variantes linguistiques régionales sont suffisamment importantes pour influencer les performances aux différentes épreuves verbales utilisées en audiologie.

L'étude de Marriage et coll. (2001) porte sur les données normatives du *Screening Test for Auditory Processing Disorders (SCAN*; Keith, 1986) développées auprès d'enfants d'âge scolaire de langue anglaise des États-Unis. L'objectif de l'étude visait à vérifier si ces données normatives pouvaient s'appliquer aux enfants du même âge dont l'anglais était celui parlé au Royaume-Uni. La batterie de tests *SCAN* est utilisée pour le dépistage du trouble de traitement auditif. La performance des enfants du Royaume-Uni était significativement inférieure à celle obtenue auprès des enfants des États-Unis (n= 133, âgés entre six et 11 ans). L'analyse des erreurs a révélé que l'accent du locuteur ainsi que la fréquence d'exposition à ces mots pouvaient expliquer cette différence dans les performances. Par exemple, les items *hot* (chaud) et *end* (fin) prononcés avec l'accent américain peuvent être perçus chez des locuteurs du Royaume-Uni comme étant *hut* (hutte) et *and* (et) en raison des différences dans la prononciation des mots entre ces deux populations (Marriage et coll., 2001). Les auteurs recommandent alors de refaire des normes spécifiques à la population visée, suivant l'enregistrement du test avec un locuteur du Royaume-Uni afin de contrôler le facteur lié à l'accent, et en remplaçant les mots moins connus par les enfants de ce pays.

Sockalingam et coll. (2004) ont mené une étude similaire en comparant les données normatives américaines du *Test for Auditory Processing Disorders in Adolescents and Adults (SCAN-A*; Keith, 1994) à celles développées auprès de la population anglophone d'Australie. Les résultats ont indiqué que la performance d'australiens (n= 32) âgés entre 18 et 47 ans était significativement inférieure ($p < .05$) à celle d'adultes américains au sous-test des mots filtrés (*Filtered words*), alors qu'elle était meilleure au sous-test de phrases compétitives (*Competing sentences*). Aucune différence significative n'a été notée entre les deux groupes pour les sous-tests d'identification de monosyllabes dans le bruit (*Auditory figure ground*) et de mots différents présentés à chaque oreille en même temps (*Competing words*). Les auteurs invitent à interpréter prudemment les résultats du test *SCAN-A* lorsque le test est utilisé auprès de personnes ne provenant pas des États-Unis, étant donné que les données normatives ont été établies auprès d'une population américaine. Par ailleurs, la cohorte de participants de l'étude de Sockalingam et coll. (2004) était composée que d'étudiants universitaires australiens, ayant probablement une meilleure connaissance de l'anglais américain que la population générale. Étant donné que la connaissance des mots d'une langue augmente avec l'exposition à cette même langue ou dialecte (Shi & Sanchez, 2011), il est possible que ces participants aient obtenu de meilleures performances aux différents sous-tests du test *SCAN-A* que des australiens provenant d'autres milieux.

Dawes (2011) s'est interrogé sur la fiabilité des données normatives américaines du test *SCAN-A* lorsqu'elles sont utilisées auprès d'adultes du Royaume-Uni. Trente et un adultes âgés entre 19 et 64 ans, de niveau universitaire ou collégial et présentant une acuité auditive normale, ont participé à l'étude. Les participants ont eu des performances significativement plus faibles que les données normatives américaines à trois des quatre sous-tests : la tâche des mots filtrés ($p < .001$), la tâche des phrases compétitives ($p < .05$) et la tâche des mots compétitifs ($p < .01$). D'après Dawes (2011), la raison principale de cette différence serait l'accent linguistique propre à chacune de ces deux populations. L'auteur suggère que le diagnostic de trouble de traitement auditif ne devrait pas être effectué à partir de résultats obtenus avec des tests auditifs incluant des stimuli verbaux, comme le test SCAN. Il invoque que les épreuves devraient porter sur les processus pré-cognitifs et perceptifs, et que par conséquent, elles ne devraient pas être composées de stimuli verbaux. Les différences relevées entre les données normatives des deux populations s'ajoutent aux raisons invoquées par l'auteur, pour expliquer les limites d'utiliser des épreuves composées de stimuli verbaux pour l'évaluation des habiletés de traitement auditif.

Keith et coll. (1987) ont exploré l'effet de l'expérience linguistique d'une langue seconde sur les résultats à deux tests d'écoute dichotique, soit le *Staggered Spondaic Word (SSW*; Katz, 1978) et le *Dichotic Consonant Vowel Scores (CV*; Berlin, Lowe-Bell, Janetta, et Kline, 1972). Pour ce faire, les performances de dix participants adultes de 19 à 35 ans,

originaires des États-Unis et ayant l'anglais comme langue maternelle, ont été comparées à celles de 30 participants âgés entre 24 et 30 ans dont l'anglais était la langue seconde. La langue maternelle de ce dernier groupe de participants était l'hindi ou l'arabe. Les performances obtenues au test *SSW*, auprès du groupe dont l'anglais était la langue seconde, montraient un taux d'erreurs moyen de 6.2% en condition droite non-compétitif, 17.6% en condition droite compétitif, 18.9% en condition gauche compétitif et 5.6% en condition gauche non-compétitif. Ces performances se sont révélées significativement plus faibles ($p < .05$) que celles mesurées auprès du groupe dont l'anglais était la langue maternelle, ayant obtenu un taux d'erreur moyen de 0% aux quatre conditions (Keith et coll., 1987). Quant aux performances mesurées avec le test *CCV*, les participants dont la langue maternelle était l'anglais et ceux dont la langue maternelle était l'hindi ont obtenu des résultats similaires. Les participants dont la langue maternelle était l'arabe ont obtenu des performances plus faibles que les deux autres groupes. Les participants pour qui l'anglais n'était pas la langue maternelle, avaient pourtant indiqué être très à l'aise avec l'utilisation de cette langue dans leurs activités de la vie quotidienne. Les auteurs soulignent que l'expérience linguistique d'une langue seconde peut influencer les performances aux épreuves de perception auditive composées de stimuli verbaux même lorsque la personne rapporte être très à l'aise à l'utiliser.

Cameron et coll. (2003a) ont vérifié si l'expérience linguistique d'une langue seconde pouvait influencer la performance au test *Macquarie Pediatric Speech Intelligibility (MPSI*; Cameron, Barker, et Newall, 2003b) en comparant les performances mesurées auprès de quatre enfants âgés entre huit et neuf ans présentant une acuité auditive normale et ayant l'anglais comme langue seconde, aux données normatives. Le *MPSI* fait partie d'une batterie de tests servant à identifier un trouble de traitement auditif. Cette épreuve consiste à présenter des phrases en même temps qu'un message compétitif (d'autres phrases) en condition ipsilatérale et controlatérale et à différents rapports signal-sur-bruit (s/b). Les enfants à l'étude, dont l'anglais était la langue seconde, ont obtenu des performances à l'intérieur des limites normales pour leur âge en condition ipsilatérale aux rapports s/b de 0 et -20 dB, de même qu'en condition controlatérale au rapport s/b de 0 dB. Cependant, pour la condition controlatérale au rapport s/b de -20 dB, leurs performances étaient plus faibles (à 2 écarts-types) que celles attendues pour leur âge, d'après les données normatives. Les auteurs suggèrent que le test *MPSI* permet de mettre en lumière les difficultés d'écoute subtiles d'enfants pour qui l'anglais n'est pas la langue maternelle. Ces difficultés seraient reliées à leur compétence linguistique et non à leurs habiletés auditives. Les auteurs reconnaissent les limites de leur étude en raison du petit échantillon, mais ils avancent que ces résultats ne font que souligner l'importance d'interpréter avec prudence les performances mesurées auprès de personnes dont la langue maternelle est différente de la langue employée dans un test de perception auditive de la parole.

L'ensemble des études effectuées auprès des populations anglophones démontre qu'il est important de considérer les connaissances et les aspects culturels d'une langue lors de l'administration d'épreuves de perception auditive formées de stimuli verbaux. L'importance d'utiliser des données normatives spécifiques aux populations servies lors de l'interprétation clinique des résultats est bien connue. Or, en clinique, on s'interroge à savoir si ce principe s'applique aux différences régionales parmi les locuteurs partageant la même langue maternelle. Au Canada par exemple, le français parlé présente des variantes régionales bien connues. Entre autres, le français parlé dans les Maritimes diffère de celui parlé au Québec, en Ontario ou dans les provinces de l'ouest. Par ailleurs, des variantes sont documentées à l'intérieur d'une même province (Chevalier, 2008).

La version franco-canadienne du test *SSW* (Katz, 1978), adaptée par Rudmin et Normandin (1983), fait partie des épreuves les plus utilisées par les audiologistes travaillant auprès de populations francophones au Canada, pour l'évaluation des habiletés de traitement auditif (Garcia, Paradis, Sénécal, et Laroche, 2006). Le test *SSW* permet d'évaluer la capacité d'un individu à combiner l'information provenant des deux oreilles (Katz et Smith, 1991), soit l'intégration binaurale. Ce test consiste à présenter, à l'aide d'écouteurs, 40 séquences de quatre monosyllabes différentes, par exemple: « grand, mère, pôle, nord ». La tâche consiste à répéter les mots entendus dans l'ordre qu'ils ont été présentés. Ainsi, les premier et deuxième mots sont émis à une oreille, alors que les troisième et quatrième mots le sont à l'autre oreille. Les deuxième et troisième mots arrivent presque simultanément, chacun à une oreille, ce qui représente la condition d'écoute dichotique du test. Les deux conditions d'écoute où les stimuli sont entendus simultanément se nomment oreille *droite compétitive* (DC) et oreille *gauche compétitive* (GC) tandis que les deux conditions où le stimulus est présenté seulement à une oreille à la fois s'appellent oreille *droite non compétitive* (DNC) et oreille *gauche non compétitive* (GNC) (Jutras, Gagné, Morin, Dénommée, et Meilleur, 1997). L'ordre de présentation des séquences alterne d'une oreille à l'autre, c'est-à-dire que si une séquence de quatre mots commence à l'oreille droite, la séquence suivante débutera à l'oreille gauche.

Entre 1990 et 1993, Bérard a développé des données normatives au Centre hospitalier Rivière-des-Prairies (CHRP) de Montréal, au Québec, pour la version franco-

canadienne du test *SSW*. Des données sont disponibles pour les enfants âgés entre six et dix ans, et pour les adultes. Ces normes sont utilisées dans les cliniques audiologiques francophones du Québec, de même que dans certaines cliniques hors-Québec (Ordre des orthophonistes et audiologistes du Québec – OOAQ, 2007).

À Moncton au Nouveau-Brunswick, des données normatives pour cette même épreuve ont été établies par les audiologistes du Centre hospitalier universitaire Dr-Georges-L.-Dumont (CHUDGLD) en 1995. Elles comprennent des données recueillies auprès d'enfants de six à douze ans et d'adultes. Ces données normatives du CHUDGLD sont utilisées dans la plupart des services d'audiologie francophones du Nouveau-Brunswick.

L'objectif de cette étude rétrospective était d'explorer l'effet des variantes linguistiques régionales qui existent entre deux populations francophones à une tâche d'écoute dichotique. Il s'agissait donc d'examiner si les données normatives pour la version franco-canadienne (Rudmin et Normandin, 1983) du test d'écoute dichotique de mots *SSW* (Katz, 1978), recueillies auprès de deux populations francophones ayant une réalité linguistique différente, notamment la population de Moncton (N.-B.) et celle de Montréal (QC), étaient influencées par les variantes linguistiques propres à chaque milieu. Étant donné que la collecte de données a été effectuée au cours de la même décennie (entre 1993 et 1995) dans ces deux centres hospitaliers, l'évolution naturelle de la langue n'a probablement pas influencé les résultats obtenus.

Même si le français est la langue d'utilisation commune pour les deux populations de la présente étude, on note des différences entre autres, au plan de la prononciation de certains phonèmes (Mougeon et Béniak, 1989; Péronnet, 1995; Ryan, 2003). Par exemple, l'affrication des occlusives /t/ et /d/ si typique en français québécois, n'est presque pas observée dans le français parlé acadien. Par ailleurs, des différences au plan du vocabulaire sont aussi notées (Chevalier et Rodrigue, 2009; Kadlec, 2005). Par exemple, on retrouve beaucoup plus d'expressions maritimes dans le français parlé en Acadie qu'en franco-québécois, tels *abrier :* «couvrir», *amarrer :* «attacher», *caler :* «s'enfoncer», etc.

Les participants ayant collaboré à la collecte de données normatives du CHRP vivent dans un milieu majoritairement francophone où le français québécois prédomine (grande région de Montréal) (Mougeon et Beniak, 1989), plus particulièrement pendant les années auxquelles cette collecte de données a eu lieu. Le français parlé dans cette région est similaire à celui retrouvé sur l'enregistrement de l'épreuve du *SSW* en français (Rudmin & Normandin, 1983). Il en est autrement pour les participants du CHUDGLD qui vivent dans un milieu à caractère essentiellement bilingue – anglais et français, soit la grande région de Moncton. D'après Boudreau et Perrot (2010), les francophones représentent environ 40% de la population de la région du grand Moncton et la majorité d'entre eux, soit 88%, est bilingue. D'autre part, seulement 24% des anglophones de cette région sont bilingues, faisant en sortent que la plupart du temps, les francophones parlent anglais dans les situations de communication quotidiennes (Boudreau et Perrot, 2010). Par ailleurs, le français couramment parlé dans la région du grand Moncton est le «chiac». Cette langue vernaculaire du sud-est du Nouveau-Brunswick, est surtout reconnue pour son mélange de français traditionnel et d'anglais (Boudreau, communication personnelle, janvier 2012). En fait, il est attendu que les participants du CHUDGLD aient plus d'erreurs au test *SSW* que ceux du CHRP en raison de l'exposition limitée à la langue franco-québécoise. En effet, la version française du test *SSW* a été développée à l'Université de Montréal et le vocabulaire choisi a probablement été influencé par l'environnement culturel des auteures. D'autre part, étant donné que l'enregistrement du *SSW* a été produit avec une locutrice parlant le franco-québécois, il est possible que cet aspect ait ajouté une difficulté supplémentaire auprès des participants du CHUDGLD, particulièrement chez les plus jeunes. En effet, les habiletés cognitives et linguistiques permettant de comprendre la langue maternelle prononcée avec un accent différent, s'acquièrent avec l'âge et l'expérience linguistique (Marriage et coll., 2001).

Méthodologie

Il s'agit d'une étude rétrospective comparant les données normatives pour la version francophone du test *SSW* (Rudmin et Normandin, 1983) colligées auprès d'enfants et d'adultes au CHRP de Montréal (QC) et au CHUDGLD de Moncton (N.-B.). Dans les deux cas, ce sont des audiologistes ayant une bonne expérience dans l'évaluation des habiletés de traitement auditif auprès des enfants qui ont participé à la collecte de données.

Participants

Selon les informations disponibles au sujet de la procédure suivie pour le développement des données normatives, le recrutement des participants au CHUDGLD a été effectué à l'aide d'affiches ainsi que par l'envoi d'une invitation par courriel, à tous les employés. Le recrutement des participants du CHRP a été fait auprès d'une garderie de la région, de même qu'auprès des employés de cet hôpital. Le nombre de participants recrutés pour chaque groupe d'âge est indiqué dans les Tableaux 1 et 2.

Pour les deux groupes, le français était la langue maternelle des participants. Les enfants recrutés au

Tableau 1. Moyenne de la valeur du score corrigé (en pourcentage) obtenu aux quatre conditions du test *SSW* (DNC, DC, GC, GNC) pour les six groupes de participants du centre hospitalier universitaire Dr-Georges-L.-Dumont. L'écart-type apparaît entre parenthèses.

Groupe	DNC	DC	GC	GNC
6 ans	11,70	32,60	41,30	10,40
n = 10	(±7,82)	(±11,35)	(±17,40)	(±7,04)
7 ans	9,40	25,20	28,50	9,20
n = 10	(±8,93)	(±9,35)	(±18,64)	(±7,51)
8 ans	14,50	27,80	24,50	10,10
n = 10	(±6,31)	(±5,77)	(±5,99)	(±5,84)
9 ans	6,40	19,20	21,00	9,70
n = 10	(±5,17)	(±6,96)	(±6,24)	(±4,18)
10 ans	7,00	14,90	17,10	8,70
n = 10	(±6,25)	(±10,19)	(±10,37)	(±4,79)
Adultes	3,00	8,30	5,50	1,80
n = 10	(±3,89)	(±5,29)	(±3,95)	(±1,98)

DNC=Droite non compétitive; DC=Droite compétitive; GC=Gauche compétitive; GNC=Gauche non compétitive

Tableau 2. Moyenne de la valeur du score corrigé (en pourcentage) obtenu aux quatre conditions du test *SSW* (DNC, DC, GC, GNC) pour les six groupes de participants du centre hospitalier Rivière-des-Prairies. L'écart-type apparaît entre parenthèses.

Groupe	DNC	DC	GC	GNC
6 ans	7,63	25,21	32,79	8,11
n = 19	(±5,13)	(±10,17)	(±8,48)	(±6,78)
7 ans	4,79	17,41	25,76	5,94
n = 17	(±3,33)	(±8,10)	(±11,49)	(±6,05)
8 ans	2,68	18,64	23,29	2,32
n = 14	(±5,74)	(±10,59)	(±10,16)	(±3,59)
9 ans	4,85	11,62	16,38	3,38
n = 13	(±2,82)	(±5,71)	(±8,05)	(±4,81)
10 ans	2,44	11,67	15,00	3,28
n = 9	(±5,04)	(±3,94)	(±7,52)	(±3,43)
Adultes	3,25	8,75	7,13	1,25
n = 8	(±3,01)	(±6,25)	(±5,87)	(±2,31)

DNC=Droite non compétitive; DC=Droite compétitive; GC=Gauche compétitive; GNC=Gauche non compétitive

CHRP étaient âgés entre six et dix ans inclusivement, alors que ceux du CHUDGLD incluaient aussi des enfants de 11 et 12 ans. Pour les fins de la présente étude, seules les données recueillies auprès des enfants âgés entre six et dix ans inclusivement ont été retenues. Pour les données des adultes, l'échantillon des deux centres hospitaliers était composé de participants âgés entre 18 et 45 ans.

Selon les réponses données par les parents à l'entrevue initiale pour les participants du CHUDGLD, ou au questionnaire pour ceux du CHRP et par les adultes, aucun participant ne présentait un problème diagnostiqué ou connu aux plans de l'audition, des apprentissages ou du développement du langage, incluant les problèmes d'articulation. Le développement des habiletés cognitives et langagières était donc considéré normal pour tous les participants, même si aucune évaluation formelle n'avait été effectuée.

Procédure

Pour la cohorte du CHRP, les parents devaient remplir un questionnaire comptant dix questions relatives aux comportements auditifs, aux capacités d'attention et à la compréhension du langage de leur enfant. Pour les adultes, les questions concernant la possibilité de problème aux plans langagier, cognitif et de l'attention étaient posées verbalement étant donné que le questionnaire était formulé à l'attention des parents.

Pour la cohorte du CHUDGLD, des questions portant sur les mêmes aspects étaient posées en entrevue aux parents ou aux adultes, le cas échéant. Dans tous les cas, si les réponses suggéraient la présence d'un problème de langage ou d'attention, les participants n'étaient pas retenus pour les données normatives.

Selon l'information disponible, une évaluation audiologique périphérique a été effectuée incluant un audiogramme aux deux oreilles, une mesure du seuil de réception de la parole, un test d'identification de monosyllabes au niveau normal de conversation (50 dB HL) dans le silence, de même qu'un tympanogramme et la mesure du réflexe stapédien. Dans le cas d'un indice de problème auditif (seuil à plus de 20 dB HL à une fréquence ou plus) ou des résultats anormaux à l'immitancemétrie, les participants ou les parents, le cas échéant, étaient avisés avec explications et recommandations, et le participant n'était pas retenu pour la collecte de données.

Ensuite, le test *SSW* était administré selon les procédures habituelles, soit à 50 dB SL de la moyenne des seuils auditifs à 500, 1000 et 2000 Hz à l'oreille concernée. Pour quelques participants, l'évaluation s'est échelonnée sur deux rencontres afin de minimiser l'effet de fatigue, tel que recommandé par les lignes directrices publiées dans le domaine (AAA, 2010; ASHA, 2005; OOAQ, 2007). Tant au CHRP qu'au CHUDGLD, la collecte de données a eu lieu dans une cabine insonorisée. Le test *SSW* était présenté à l'aide d'écouteurs supra-auriculaires reliés à un audiomètre de marque Madsen (OB822) et auquel était branché un lecteur de cassette. Dans les deux cas, ce fut la version 2, de la cassette francophone du *SSW* distribuée par le Centre de Consultation Ressource et Recherche en Audiologie (Montréal, Québec) qui a été employée. L'étalonnage était effectuée en ajustant le vu-mètre à 0 à l'aide du son de référence présent au début de la cassette.

Résultats

Pour chaque groupe d'âge, la moyenne des «scores d'erreurs corrigés» (*C-SSW*) obtenus aux quatre conditions du *SSW* pour les participants du CHRP et du CHUDGLD est illustrée à la Figure 1. En résumé, le score *C-SSW* se calcule en prenant le nombre d'erreurs commises dans chaque condition du *SSW* pour les 40 séquences du test. Ce nombre est multiplié par 2.5 afin d'obtenir un pourcentage d'erreurs pour chaque condition d'écoute. De ce pourcentage, il faut soustraire le pourcentage d'erreurs obtenu au test d'identification des monosyllabes dans le silence à chaque oreille. Le détail de ce calcul se trouve dans le manuel d'instructions du test *SSW* (version originale; Katz, 1978).

L'interprétation clinique des résultats se fait avec le score *C-SSW*. En se référant aux résultats de la Figure 1, on peut noter que le pourcentage d'erreurs *C-SSW*, pour chaque condition du test, diminue en fonction de l'âge pour les deux populations à l'étude. De façon générale, le pourcentage d'erreurs *C-SSW* moyen de chaque condition du test est plus élevé pour le groupe du CHUDGLD que pour celui du CHRP.

Afin de vérifier ces observations, une analyse de variance (ANOVA) mixte à trois facteurs a été effectuée, soit le facteur Condition (4 niveaux), le facteur Âge (6 niveaux) et le facteur Région (2 niveaux), à l'aide du logiciel SPSS (*IBM SPSS Statistics 19*). Le facteur Condition était le seul dont les mesures étaient répétées. Les résultats de l'analyse ont révélé un effet significatif pour le facteur Condition [$F_{(3,384)}$=194.71, p<.001, η^2= 0.60], le facteur Région [$F_{(1,28)}$= 22.52 p<.001, η^2= 0.15] et le facteur Âge [$F_{(5, 128)}$=25.06, p<.001, η^2= 0.50]. L'interaction double Condition X Âge [$F_{(15,384)}$= 8.65, p<.001, 2= 0.25] était significative alors que les interactions doubles Condition X Région [$F_{(3,384)}$= 1.05, p=.372] et Région X Âge [$F_{(5,128)}$= 1.25, p=.292] n'étaient pas significatives, de même que l'interaction triple Condition X Région X Âge [$F_{(15,384)}$=0.95, p=.504].

Pour explorer l'interaction double Condition X Âge, une ANOVA à un facteur, soit le facteur Âge, a été effectuée pour chaque condition du *SSW*, en jumelant les résultats obtenus aux deux centres hospitaliers. Ainsi, pour la condition DNC, une différence statistiquement significative a été observée

Tableau 3. Résultat des comparaisons post hoc pour explorer l'effet d'âge à la condition droite non-compétitive du test *SSW* en utilisant le test de Tukey. Les comparaisons ayant atteint un niveau de signification statistique sont identifiées par un astérisque (*).

	Condition droite non compétitive				
	7 ans	**8 ans**	**9 ans**	**10 ans**	**Adultes**
6 ans	0,622	0,969	0,313	0,213	0,019*
7 ans		0,980	0,994	0,959	0,459
8 ans			0,822	0,681	0,157
9 ans				1	0,808
10 ans					0,942

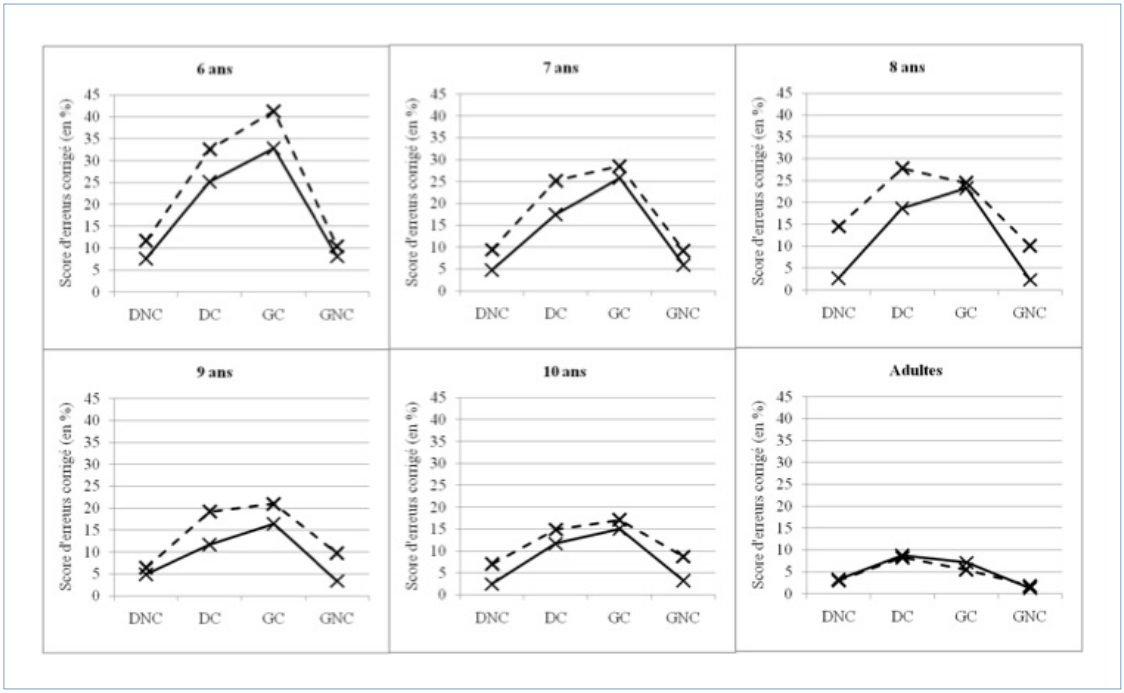
Tableau 4. Résultat des comparaisons post hoc pour explorer l'effet d'âge à la condition droite compétitive du test *SSW* en utilisant le test de Tukey. Les comparaisons ayant atteint un niveau de signification statistique sont identifiées par un astérisque (*).

	Condition droite non compétitive				
	7 ans	**8 ans**	**9 ans**	**10 ans**	**Adultes**
6 ans	0,025	0,265	0,000*	0,000*	0,000*
7 ans		0,954	0,278	0,105	0,000*
8 ans			0,048*	0,014*	0,000*
9 ans				0,993	0,206
10 ans					0,559

Tableau 5. Résultat des comparaisons post hoc pour explorer l'effet d'âge à la condition gauche compétitive du test *SSW* en utilisant le test de Tukey. Les comparaisons ayant atteint un niveau de signification statistique sont identifiées par un astérisque (*).

	Condition droite non compétitive				
	7 ans	**8 ans**	**9 ans**	**10 ans**	**Adultes**
6 ans	0,020*	0,001*	0,000*	0,000*	0,000*
7 ans		0,910	0,058	0,010*	0,000*
8 ans			0,484	0,162	0,000*
9 ans				0,981	0,004*
10 ans					0,051

Figure 1. Taux d'erreurs moyens pour les quatre conditions (DNC = Droite non compétitive; DC = Droite compétitive; GC = Gauche compétitive; GNC = Gauche non compétitive) du test *SSW* en fonction du groupe d'âge et de la région où les données ont été prises. Les symboles reliés par une ligne continue correspondent aux résultats obtenus auprès des participants du Centre hospitalier Rivière-des-Prairies, alors que ceux reliés par une ligne en pointillé correspondent aux résultats obtenus auprès des participants du Centre hospitalier universitaire Dr-Georges-L.-Dumont.



[$F_{(5,139)}$= 2.66, p=.025] pour le facteur Âge, mais la taille de l'effet notée était faible, soit η^2= 0.09. Les comparaisons post hoc ont été faites en utilisant le test de Tukey et les résultats sont rapportés au Tableau 3. La seule différence significative a été notée entre le groupe des six ans et le groupe d'adultes pour la condition DNC.

Une tendance similaire a été notée lors de l'analyse des résultats obtenus en condition GNC. Une différence statistiquement significative a été observée [F(5,139)= 3.78, p=.003]. La taille de l'effet notée est faible, i.e.: η^2= 0.12. Tel qu'indiqué au Tableau 6, les comparaisons post hoc ont révélé une différence significative entre le groupe des six ans et les adultes, ainsi qu'entre le groupe des sept ans et celui des adultes.

Pour les deux conditions d'écoute compétitives, soit DC [$F_{(5,139)}$= 13.93, p<.001] et GC [$_{F(5,139)}$= 21.28, p<.001], une différence statistiquement significative est notée. Telles qu'illustrées dans les Tableaux 4 et 5, des différences statistiquement significatives existent entre plusieurs groupes d'âge. En condition DC (Tableau 4), la moyenne du score *C-SSW* obtenue auprès des participants de six ans est statistiquement différente de celle obtenue auprès des enfants de sept ans, neuf ans, dix ans et des adultes. De même, il existe une différence significative entre la moyenne du score *C-SSW* obtenue auprès des participants de sept ans et des adultes, ainsi qu'entre les participants de huit ans et les participants de neuf ans, dix ans et les adultes.

En condition GC (Tableau 5), la moyenne du score *C-SSW* obtenue auprès des participants de six ans est statistiquement différente de celle obtenue auprès des enfants de sept ans, huit ans, neuf ans, dix ans et des adultes. De même, il existe une différence significative entre la moyenne du score *C-SSW* obtenue auprès des participants de sept ans et celle du groupe de dix ans, et entre les participants de sept ans et les adultes. Pour les participants de huit ans, il y a une différence significative avec les résultats des adultes. Enfin, il y a une différence significative entre les participants de neuf ans et les adultes.

Discussion

L'objectif de cette étude était d'explorer l'effet des variantes linguistiques régionales à la version franco-canadienne (Rudmin et Normandin, 1983) du test d'écoute dichotique de mots *SSW* (Katz, 1978). Les données normatives développées auprès de deux populations francophones du Canada, soit celles de Moncton au Nouveau-Brunswick et celles de Montréal au Québec, ont été comparées. Ces données ont été recueillies il y a plus de 20 ans mais elles sont encore utilisées dans les cliniques d'audiologie des différentes communautés francophones du Canada. Le pourcentage d'erreurs moyen obtenu auprès des participants de Moncton est significativement plus élevé que celui des participants de Montréal. Étant donné que les deux populations à l'étude se distinguent essentiellement par leur français parlé, ces résultats suggèrent que les

Tableau 6. Résultat des comparaisons post hoc pour explorer l'effet d'âge à la condition gauche non compétitive du test *SSW* en utilisant le test de Tukey. Les comparaisons ayant atteint un niveau de signification statistique sont identifiées par un astérisque (*).

	Condition gauche non compétitive				
	7 ans	8 ans	9 ans	10 ans	Adultes
6 ans	0,867	0,332	0,523	0,595	0,001*
7 ans		0,942	0,989	0,993	0,022*
8 ans			1	1	0,208
9 ans				1	0,126
10 ans					0,155

variantes linguistiques régionales peuvent influencer les performances mesurées à l'épreuve dichotique de mots *SSW*. En plus des particularités notées au plan du vocabulaire et de la prononciation de certains phonèmes, le caractère bilingue du français parlé dans la région de Moncton, comparativement à celui unilingue francophone des participants de Montréal, peuvent avoir influencé les résultats. Par exemple, Tabri, Smith, Abou Chacra et Pring (2011) ont démontré que des adultes bilingues (depuis l'âge de cinq ou moins) présentaient des scores inférieurs à ceux d'adultes monolingues à la reconnaissance de mots en finale des phrases du test R-SPIN (Bilger, Nuetzel, Rabinowitz, et Rzeczkowski, 1984) lorsque présentés avec un bruit de verbiage compétitif et ce, malgré des performances similaires pour la même tâche d'écoute sans le bruit compétitif. Par exemple, au rapport s/b de + 10 dB, la performance moyenne des adultes monolingues était de 88% alors qu'elle était de de 83% pour les adultes bilingues (Tabri et coll., 2011). La version franco-canadienne du test *SSW* a été développée à l'Université de Montréal et l'enregistrement de l'épreuve a été produit par une locutrice franco-québécoise, ce qui a pu entraîner des difficultés subtiles chez les participants de Moncton, surtout dans les conditions d'écoute difficiles du test, que les participants de Montréal n'auraient pas eues.

L'ambition de développer des données normatives spécifiques à chaque communauté linguistique, pour chaque épreuve composée de stimuli verbaux, peut paraître utopique. Certains suggèrent que l'évaluation des habiletés de traitement auditif ne devrait être effectuée qu'avec des épreuves comprenant des stimuli non verbaux entre autre pour éviter cette problématique (Dawes, 2011). D'autres invoquent cependant l'importance d'inclure des épreuves composées de stimuli verbaux en raison de la primauté de ce type de signal dans les activités quotidiennes (Musiek et Chermak, 2007). De plus, la comparaison des données obtenues à partir des deux types de tests (composés de stimuli verbaux et non verbaux) permet parfois de préciser la nature des difficultés d'écoute (Musiek et Chermak, 2007). Par exemple, si les performances mesurées auprès d'une personne sont sous les limites de la normale aux épreuves composées de stimuli verbaux, mais qu'elles sont normales aux épreuves composées de stimuli non verbaux, il est probable que les problèmes d'écoute soient reliés à un déficit des fonctions langagières plutôt qu'auditives. La combinaison des deux types d'épreuves apporte une information utile à l'audiologiste, notamment pour la planification des activités de réadaptation.

Les données de cette étude ont été recueillies dans un but clinique et non de recherche. Ainsi, plusieurs facteurs tels que la latéralité, le sexe, le niveau de développement du langage, les capacités d'attention, etc., n'ont pas été contrôlés de façon aussi stricte qu'il aurait été dans le cas pour une étude prospective multicentrique. Par ailleurs, le petit échantillon et le fait que les données ne proviennent que deux communautés francophones font en sorte que d'autres études sont requises pour mieux comprendre l'effet des variantes linguistiques aux épreuves pré-enregistrées d'écoute dichotique de mots.

Les résultats de la présente étude concordent avec ceux des investigations effectuées auprès de populations anglophones (Cameron et coll., 2003a; Dawes, 2011; Keith et coll., 1987; Marriage et coll., 2001; Sockalingam et coll., 2004) et soulignent l'importance de développer des données normatives propres aux populations auprès desquelles les épreuves pré-enregistrées comptant des stimuli verbaux sont administrées. Les audiologistes doivent être attentifs à l'impact possible des variations et biais culturels sur les performances à ce type d'épreuve. Le fondement, l'efficacité

et la validité de leurs programmes de traitement en dépendent.

Remerciements

Les auteurs aimeraient remercier les audiologistes du centre hospitalier universitaire Dr-Georges-L.-Dumont (Moncton, NB) ainsi que Christiane Bérard, audiologiste au centre hospitalier Rivière-des-Prairies (Montréal, QC) pour leur collaboration à ce projet.

Références

American Academy of Audiology (2010). Clinical practice guidelines. Diagnosis, treatment and management of children and adults with central auditory processing disorder [Technical Report]. Disponible au http://www.audiology.org/resources/documentlibrary/Pages/CentralAuditoryProcessingDisorder.aspx

American Speech-Language-Hearing Association (2005). (Central) Auditory processing disorders. [Technical Report]. Disponible au http://www.asha.org/members/deskref-journals/deskref/default.

Bérard, C. (1990-1993). Données normatives du test SSW: Hôpital Rivière-des-Prairies. Montréal, Québec.

Bilger, R. C., Nuetzel, J. M., Rabinowitz, W. M. et Rzeczkowski, C. (1984). Standardization of a test of speech perception in noise. *Journal of Speech and Hearing Research, 27*, 32–38.

Berlin, C. I., Lowe-Bell, S. S., Janetta, P. J. et Kline, D.G. (1972). Central auditory deficits after temporal lobectomy. *Archives of Otolaryngology, 96*, 4-10.

Boudreau, A. (2012). (Communication personnelle). La définition du «chiac». Université de Moncton, Moncton, Nouveau-Brunswick.

Boudreau, A. et Perrot, M.E., (2010). Le chiac c'est du français. Représentation du mélange français/anglais en situation de contact inégalitaire. Dans H. Boyer (dir.), *Hybrides linguistiques. Genèse, statuts, fonctionnement*. Paris: L'Harmattant.

Cameron, S., Barker, R. et Newall, P. (2003a). The effect of linguistic background on the Macquarie Pediatric Speech Intelligibility Test. *The Australian and New Zealand Journal of Audiology, 25*, 95-98.

Cameron, S., Barker, R. et Newall, P. (2003b). Development and evaluation of an Australian version of the Pediatric Speech Intelligibility Test for Auditory Processing Disorder. *The Australian and New Zealand Journal of Audiology, 25*, 16-27.

Chevalier, G. (2008). Les français du Canada : Faits linguistiques, faits de langue. *Alternative Francophone, 1*, 80-97.

Chevalier, G. et Rodrigue, L. (2009). *Les mots pour parler des maux: Lexique différentiel des termes acadiens liés à la santé*. Consortium national de la formation en santé: Moncton.

Dawes, P. (2011). The SCAN-A in testing for auditory processing disorder in a sample of British adults. *International Journal of Audiology, 50*, 107-111.

Garcia, L. J., Paradis, J., Sénécal, I., et Laroche, C. (2006). Utilisation et satisfaction à l'égard des outils en français évaluant les troubles de la communication. *Revue canadienne d'orthophonie et d'audiologie, 30*, 239-249.

Jutras, B., Gagné, J., Morin, L., Dénommée, C. et Meilleur, G. (1997). Troubles auditifs centraux chez des enfants et adolescents cérébrolésés: Évaluations audiologiques effectuées à partir d'épreuves non verbales et de l'adaptation française d'épreuves verbales. *Revue d'orthophonie et d'audiologie, 21*, 294-300.

Kadlec, J. (2005). Le monde acadien et son reflet dans les particularités du français acadien. Dans M. Paluszkiewicz-Misiaczeck, A. Reczyńska, et A. Śpiewak (dir.), *Lieu et mémoire au Canada: Perspectives globales* (pp. 209- 214). Polska Akademia Umiejętności (open library).

Katz, J. (1978). *S.S.W. workshop manual*. Author: New York.

Katz, J. et Smith, S. P. (1991). The Staggered Spondaic Word Test: A ten-minute look at the central nervous system through the ears. *Annals New York Academy of Sciences, 51,* 233-251.

Keith, R. W. (1986). SCAN- *A Screening Test for Auditory Processing Disorders*. San Diego: The Psychological Corporation.

Keith, R. W. (1994). SCAN-A: *Test for Auditory Processing Disorders in Adolescents and Adults.* San Antonio: The Psychological Corporation Harcourt Brace Jovanovich.

Keith, R. W., Katbamna, B., Tawfik, S. et Smolak, L. H. (1987). The effect of linguistic background on staggered spondaic word and dichotic consonant vowel scores. *British Journal of Audiology, 21*, 21-26.

Marriage, J., King, J., Briggs, J. et Lutman, M. E. (2001). The reliability of the SCAN test: Results from a primary school population in the UK. *British Journal of Audiology, 35*, 199-208.

Mougeon, R. et Beniak, E. (1989). Le français parlé hors Québec: Aperçu sociolinguistique. Québec : Presses de l'Université Laval.

Musiek, F. et Chermak, G. (2007). Future directions in the identification and diagnosis of (Central) Auditory Processing Disorder. Dans F. Musiek et G. Chermak, (dir.), *Handbook of (central) auditory processing disorder. Auditory neuroscience and diagnosis.* San Diego: Plural Publishing Inc., (pp. 455-468).

Ordre des orthophonistes et audiologistes du Québec (OOAQ). (2007). Révision des pratiques courantes en audiologie entourant le trouble de traitement auditif (TTA) chez l'enfant. Montréal: Rapport du comité ad hoc.

Péronnet, L. (1995). L'apport de la tradition orale à la description linguistique. *Érudit, 5,* 37-44.

Rudmin, F. et Normandin, N. (1983). Experimental dichotic test in French modeled on SSW design. *Journal of Human Communication Canada, 7*, 348-360.

Ryan, R. (2003). Particularités phonologiques et phonétiques d'un parler acadien de l'Île-du-Prince-Édouard (Canada). Actes du Congrès International d'Études Françaises. La Rioja, Espagne, 123-129.

Shi, L.-F. & Sánchez, D. (2011). The role of word familiarity in Spanish/English bilingual word recognition. *International Journal of Audiology, 50*, 66–76.

Sockalingam, R., Morrison, R., Cole, D., James, S., Morin, S. et Potter, S. (2004). Comparing SCAN-A scores between speakers of standard Australian English and American English: A preliminary study. *The Australian and New Zealand Journal of Audiology, 26*, 110-115.

Tabri, D., Smith Abou Chacra, K. et Pring, T. (2011). Speech perception in noise by monolingual, bilingual and trilingual listeners. *International Journal of Language and Communication Disorders, 46*, 1-12

Note des auteurs

Adresse pour correspondance : Josée Lagacé, Ph. D., Professeure adjointe et audiologiste, Université d'Ottawa Pavillon Roger Guindon Pièce 3053 451 Smyth, Ottawa, ON K1H 8M5 Canada. Courriel : josee.lagace@uottawa.ca

Date soumis : Le 17 avril, 2012

Date accepté : Le 29 avril, 2013

Technical Aspects of a Videofluoroscopic Swallowing Study

Aspects techniques de l'étude vidéofluoroscopique de la déglutition

Melanie Peladeau-Pigeon
Catriona M. Steele

KEY WORDS

DYSPHAGIA
DEGLUTITION
DEGLUTITION DISORDER
SWALLOWING
VIDEOFLUOROSCOPY
FLUOROSCOPY

Abstract

Dysphagia management has become a core area of practice for Speech-Language Pathologists (S-LPs). Videofluoroscopy is a readily available tool used to determine swallowing safety and efficiency as characterized by airway protection, penetration or aspiration and excess oral or pharyngeal residue. Performing a swallowing study successfully requires proper balance of many features. While many of the technical aspects are controlled by the radiology staff, it is important for the conducting S-LP to have a basic understanding of the technical aspects that can impact the quality and integrity of the video.

This article describes the types of fluoroscope available, factors influencing the image contrast, the creation of a contrast impregnated fluid, imaging techniques (pulsed versus continuous), imaging resolution (spatial and temporal) and safety considerations. This article hopes to clarify concepts to avoid future misuse of fluoroscopic imaging terminology as applied to a swallowing study. In addition, this article hopes to provide the foundations for S-LPs to be able to communicate effectively with the radiology staff, as the optimal videofluoroscopic exam can only be successfully obtained when both parties work together as a team.

Abrégé

La gestion de la dysphagie est devenue l'un des domaines centraux de la pratique en orthophonie. La vidéofluoroscopie est un outil facilement disponible utilisé pour déterminer la sécurité et l'efficience de la déglutition, caractérisée par la protection, la pénétration ou l'aspiration des voies respiratoires, et un excédent de résidu buccal ou pharyngé. Pour réussir une étude de la déglutition, il faut un juste équilibre entre divers éléments. Si une bonne part des aspects techniques sont contrôlés par le personnel de radiologie, il est important pour l'orthophoniste en charge d'avoir une compréhension de base des aspects techniques pouvant avoir un impact sur la qualité et l'intégrité de la vidéo.

Cet article décrit les types de fluoroscopes disponibles, les facteurs qui influencent le contraste de l'image, la création d'un fluide imprégné de substances à contrastes, les techniques d'imagerie (pulsée par opposition à continue), la résolution de l'imagerie (spatiale et temporelle) et les considérations de sûreté. Cet article vise à clarifier les concepts de façon à éviter une mauvaise utilisation de la terminologie de l'imagerie fluoroscopique telle qu'appliquée à l'étude de la déglutition. En plus, il couvre les notions qui permettront aux orthophonistes de communiquer efficacement avec le personnel de radiologie, parce qu'un examen vidéofluoroscopique optimal ne peut être réussi que lorsque les deux parties travaillent en équipe.

Melanie Peladeau-Pigeon, M.H.Sc.
Toronto Rehabilitation Institute
University Health Network
Toronto, ON
Canada

Catriona M. Steele, Ph.D.
Toronto Rehabilitation Institute
University Health Network
University of Toronto
Toronto, ON
Canada

Introduction

Swallowing is not only a basic function essential for maintaining proper nutrition and hydration but also a function important for quality of life given the central role that eating and drinking play in all varieties of human social activity. The term dysphagia refers to swallowing impairment. Impairments or abnormalities in swallowing physiology have both functional and social participation consequences. Dysphagia management has become a core area of practice for Speech-Language Pathologists (S-LPs) in Canada as demonstrated by the inclusion of dysphagia in the Canadian Association of Speech-Language Pathologists and Audiologists (CASLPA) national certification examination in 1999 (CASLPA, 2007) and re-iterated in the *Position Paper on Dysphagia in Adults* by CASLPA (2007). The importance of swallowing management in the scope of practice for S-LPs is also reflected in policies and guidelines provided by the provincial colleges/associations such as the College of Audiologists and S-LPs of Ontario (CASLPO) (2007), the Alberta College of S-LPs and Audiologists (ACSLPA) (2009) and the Manitoba Speech and Hearing Association (MSHA) (2009).

In clinical practice, the evaluation of swallowing focuses primarily on determining whether two areas of dysfunction exist: a) impaired airway protection leading to penetration or aspiration of material into the respiratory system; and b) impaired swallowing efficiency resulting in prolonged transit times and/or oral or pharyngeal residue (Clavé et al., 2008). A key component of dysphagia competency is the ability to perform and interpret videofluoroscopic swallowing examinations (VFSS), which may be used to confirm the presence/absence and severity of these areas of dysfunction, to identify abnormalities in swallowing physiology and to probe candidacy for specific forms of intervention (CASLPO, 2007). In an early and seminal article on videofluoroscopy practice, Drs. Bronwyn Jones and Martin Donner wrote: "examination of the patient with dysphagia depends on two major factors: a) meticulous attention to the examination itself; and b) an in-depth knowledge of normal and abnormal anatomy and physiology of swallowing" (Jones and Donner, 1989, p.162). The purpose of this tutorial article is to address the first of these two factors by reviewing technical aspects of fluoroscopy and the VFSS procedure. We believe that it is critical that S-LPs understand how technical considerations can influence the quality of data and information acquired during the VFSS assessment.

The VFSS Assessment

Fluoroscopy is a medical imaging technique that enables the visualization of the motion of internal fluids and anatomical structures. When used to examine oropharyngeal swallowing physiology and bolus flow through the upper aerodigestive tract, this procedure is commonly referred to as a VFSS or Modified Barium Swallow (MBS). Other names for the procedure may reflect particular protocol decisions (e.g., the "cookie swallow", which is a term used by Logemann (1993), reflecting the inclusion of a Lorna Doone cookie in their protocol) or health insurance billing codes (e.g., "palatopharyngeal analysis"). The assessment typically begins in the lateral (sagittal) plane; however an anterior-posterior (A-P) view may also be included at the end of the procedure (CASLPO, 2007; Martin-Harris et al., 2008). The lateral plane is ideal for detecting invasion of material into the airway (penetration-aspiration) as the view clearly differentiates the airway from the esophagus, and allows visualization of the entry of material into the supraglottic space and larynx (Martin-Harris & Jones, 2008). The A-P view provides information regarding symmetry of structures, function and bolus flow, and is particularly useful when further exploration of bolus flow through the cervical esophagus is desired (Martin-Harris & Jones, 2008).

The following is a list of the different types of information that can be gathered from a VFSS to inform and justify clinical management decisions:

- Assessment:
    - Determine the presence, nature and severity of swallowing impairment;
    - Assess the various components of swallowing physiology and detect abnormalities;
    - Evaluate swallowing efficiency and safety (Martin-Harris & Jones, 2008; Martin-Harris, Logemann, McMahon, Schleicher & Sandidge, 2000):
        - Efficiency of bolus preparation in the oral cavity;
        - Efficiency of transport from the oral cavity, to the pharyngeal cavity and into the esophagus;
        - Safety of airway protection;
    - Determine the presence of and response to penetration or aspiration;
    - Analyze the timing of swallowing events;
    - Determine the impact of fatigue on swallowing physiology.
- Management Plan:
    - Evaluate changes in swallowing efficiency and safety as a function of food/fluid consistency;

- Evaluate the effects of rehabilitation techniques such as postural changes, sensory enhancement and behavioural manoeuvres on swallowing function (Logemann, 1997).

As part of the VFSS exam, a dynamic movie of swallowing is recorded; in addition to providing the opportunity for careful review by the clinician, this movie can be used for patient education or for the communication of findings to other health care practitioners (Kelchner, 2004).

In order to competently perform and interpret a VFSS, health practitioners should have a clear understanding of physiological features and abnormalities (Jones & Donner, 1989). The optimal VFSS examination aims to capture an accurate representation of swallowing physiology; this is best achieved through interdisciplinary collaboration between radiology staff and S-LPs. In turn, successful interpretation of the VFSS depends on the number and quality of the images obtained, the observer's skills (education, experience, and confidence), and on human visual perception and the ability to recognize patterns (Rauch, 2008). In the sections that follow, we review a number of technical features of fluoroscopy as applied in the VFSS. Topics covered include: equipment type, image contrast, contrast agents, imaging modes, imaging resolution and safety considerations. We believe that an understanding of all of these issues by S-LPs is important for ensuring successful performance of quality VFSS examinations, as illustrated schematically in Figure 1.

Figure 1: Technical aspects influencing the successful acquisition of a videofluoroscopic swallowing exam

Fluoroscopy Equipment Type: Flat panel detectors and Image Intensifier Systems

Fluoroscopy is an imaging modality used to acquire a continuous series of x-ray images, which can be viewed in real time, allowing an appreciation of dynamic physiology. There are two major types of fluoroscopy system: flat-panel detectors (FPD) and image intensifier systems. The type of system used impacts the resulting image and its resolution, which will be discussed in a subsequent section. The easiest method of differentiating the two systems is to look at the image display. For an image intensifier, the corners of the display are typically cut off and straight lines such as that of a mesh are slightly curved, as in a 2-dimensional depiction of the world as a globe. Unfortunately, this system produces image distortions which can impact quantitative analyses performed on the series of images (Cerveri, Forlani, Borghese, & Ferrigno, 2002). In an FPD system, the grid shows up with no distortion; all lines in the mesh are perpendicular and the image produced uses the full screen size. Fluoroscopy images of a mesh grid produced by each type of system can be found in an article by Nickoloff (2011). The type of fluoroscopy system used can also determine the type of video capture system that is suitable to use for post VFSS analysis: an image intensifier typically yields an analog signal while an FPD generates a digital signal. Fluoroscopy systems used in swallowing studies are typically image intensifiers.

Image Contrast and Brightness

The fluoroscopy image sequence has a characteristic contrast or range of grayscale values. The contrast is obtained due to the various tissue compositions present, each holding their own characteristic densities. Overall contrast can be altered by the fluoroscope operator by changing the energy properties of the x-ray photons that are emitted. Automatic brightness control (ABC) is a feature that is often used by the technologist to help maintain the overall image brightness at a constant level and ensure adequate contrast of anatomical features on the image.

Contrast Agents: Barium Radio-opacity, Concentration, Density and Recipes

The contrast or opacity of the bolus that is being observed can be further enhanced using contrast agents. Barium is the most common active ingredient for oral or gastrointestinal contrast due to its high density, which shows up as a radiopaque substance on the fluoroscopic image. Radio-opacity refers to the ability of a substance or object to obstruct the passage of energy such as x-rays. On a traditional x-ray image, radiopaque material such as bone or barium, has greater attenuation and is represented by the lighter end of the grayscale spectrum (i.e., white). However, in fluoroscopy, the resulting representation is a reverse

pattern: barium impregnated fluids, bones and attenuating materials such as metal will typically appear as darker objects.

Barium impregnated fluids or foods are used so that the clinician can track the movement of stimuli from the oral cavity to the upper esophagus. Caution must be used when choosing both the test fluid and the barium product. The addition of barium, liquid or powder, will affect the density and viscosity of the test fluid. In the United States, Varibar™ is a commercially available line of low concentration (40% w/v) barium products that are produced in different consistencies (e.g., thin, nectar-thick, honey-thick) and were developed specifically for use in oropharyngeal swallowing examinations. Given that Varibar™ products are not currently available for clinical use in Canada, it is common practice to mix or dilute other gastrointestinal imaging preparations for use in the examination of the oropharynx. Clinicians should be aware that mixing barium preparations in ways that differ from the manufacturer's labeled instructions constitutes an "off-label" use of the product. Essentially, this means that the product is being used for reasons that have not yet received approval from Health Canada. Off-label uses include varying dosage and using different routes of administration than those indicated on the product label. The manufacturer and supplier are not allowed to advertise the product for off-label use. In addition, federal authorities such as Health Canada do not regulate off-label use (CMPA, 2012). Regulation of off-label use may be addressed under either provincial jurisdictions or by professional regulatory bodies or colleges. Regardless of whether product use falls under intended or off-label use, attention should always be paid to manufacturer instructions regarding the shelf-life and expiry dates of products once opened. It is recommended that S-LPs consult with colleagues in the radiology department to understand shelf-life restrictions of barium, once opened, and that a log be maintained for bottle open dates. Alternatively, the open date or the expiry date can be labeled on the bottle directly.

Recipes can help to ensure that standard preparations of the stimuli are used across examinations. Ideally, standardized recipes would be used across institutions; however, none exist at the current time. When mixing barium into concentrations other than those described on the manufacturer's label, two pieces of information are required to determine the appropriate amounts of powder (or solution) to mix with given volumes of water (or other test stimuli): a) the concentration; and b) the density of the original product. There is often confusion between the terms "concentration" and "density" when describing barium mixtures. The term "concentration" refers to the amount of one compound in reference to another compound, expressed in a weight to volume ratio (w/v), volume to volume ratio (v/v) or weight to weight ratio (w/w). For example, the Polibar Liquid Plus label says that it is a 105% w/v solution (E-Z-EM Canada Inc., 2012). This means that there are 105g of barium sulfate in 100mL of the Polibar Liquid Plus solution. The concentration is also listed as 58% w/w meaning that there are 58g of barium sulfate in 100g of the Polibar Liquid Plus solution. The concentration of a barium preparation relates directly to its opacity, or visibility on an x-ray image; higher concentrations will appear more radio-opaque (darker) on the image. Higher concentrations of barium are also intended to coat the mucosal walls of the gastrointestinal tract, to allow double-contrast examinations in which the outline of an anatomical space or cavity can be appreciated as well as the outline of a fluid flowing through that space. Clinicians need to be aware that higher concentrations of barium may leave a coating in the oropharynx that could be mistakenly interpreted as being residue (Steele, Molfenter, Péladeau-Pigeon and Stokely, 2013).

By contrast, the term "density" refers to the mass per unit volume of a material. For example, water has a density of approximately 1g/mL at 20 C. This simply means that a measured volume of 1mL weighs 1g. The temperature of the material is typically noted when reporting density; given that density is a physical property that varies with temperature. Density is also pressure dependent (a fact that is sometimes omitted). The densities of barium solutions are often not listed on the product label. However, density is a material property that can be easily measured in a lab setting.

When preparing barium for use in VFSS, the goal is to produce a product with a concentration that is adequately opaque to be visible on the radiographic image, but not so concentrated that significant mucosal coating occurs. The Varibar™ product line in the USA is manufactured to have a 40% w/v concentration. However, a recent article by Fink and Ross (2009) argued that even this low-concentration solution is not like a "true thin liquid" and proposed further dilution of thin liquid Varibar™ in 50% ratio with water to yield a concentration of approximately 20-22% w/v. Sample calculations for preparing a 22% w/v barium solution using a commercially available barium preparation (Liquid Polibar) are shown in the Appendix of this article.

Imaging Modes: Continuous versus Pulsed Fluoroscopy

There are three modes of operation in fluoroscopy: continuous, high dose, and pulsed. In VFSS, continuous and pulsed modes are commonly used. Continuous fluoroscopy generates a steady current. Images are generated at a rate of 30 images per second. Therefore, each image is exposed or acquired over a timeframe of 33 milliseconds. Pulsed fluoroscopy delivers short or pulsed bursts of current. The image exposure time can vary between 3 and 10

milliseconds and the pulse rate can be set at 30, 15 or 7.5 pulses per second. Pulsed fluoroscopy offers the advantage of eliminating motion blur caused by long acquisition or exposure times (Schueler, 2000). In addition, the pulsed mode can theoretically help to reduce radiation exposure. For example, an equivalent total examination time of 5 seconds would involve 5000 milliseconds of exposure under continuous fluoroscopy conditions, but this could be reduced to 1500 milliseconds of exposure using a pulse rate of 30 pulses per second and 10 millisecond duration bursts.

Continuous and pulsed fluoroscopy yield different image quality. If the same tube current is used in videos pulsed at 30 and 15 pulses per second, there will be a noticeable deterioration in image quality at the lower pulse rate due to human visual field perception. When viewing a video, which is essentially a series of images, the "eye integrates the noise content of all images presented within a period of approximately 0.2 seconds" (Van Lysel, 2000). Therefore, when fewer images are displayed, as in the example of 15 pulses per second, the observer perceives an increase in noise even if the image quality and resolution has remained constant. The reader is directed to a presentation by Rauch (2008) for examples of videofluoroscopic images obtained using pulsed versus continuous fluoroscopy modes. It should be noted that fluoroscopy pulse rate is not the same thing as video frame rate. Controversies regarding pulse and video frame rate will be discussed below.

Image Resolution: Spatial and Temporal Resolution

While image quality is partly an intrinsic property of the imaging system, it is also dependent on the visual perception of the observer (Rauch, 2008). Spatial and temporal resolutions are two key features that contribute to intrinsic image quality. Spatial resolution describes the level of detail that is captured in an image and temporal resolution refers to the number of images displayed over a given time period.

Spatial Resolution

Spatial resolution refers to "the ability to see small detail" (Bushberg, Seibert, Leidholdt, & Boone, 2012). A system with higher spatial resolution is able to detect the presence of smaller objects or details. The lower limit of spatial resolution is "the size of the smallest possible object that the system can resolve" (Bushberg et al., 2012). There are two spatial dimensions and thus two resolutions of interest; vertical and horizontal.

Vertical spatial resolution is determined by the number of horizontal lines contained in the image. This is also called the number of raster lines or scan lines. The CASLPO Practice Standards and Guidelines for Dysphagia indicate that the VFSS video should contain a minimum of 400 raster lines (CASLPO, 2007). It should be noted that limits to image resolution may be influenced both by monitor used to display the live fluoroscopic images and/or by the equipment used to capture these images in a recording. Spatial resolution can also be influenced by the Field of View (FOV) or magnification for an image intensifier fluoroscopy system. For a given number of raster lines, a smaller FOV would have a higher vertical resolution than that of a larger FOV. However, in an FPD system, the maximum spatial resolution is inherent to the system. Horizontal spatial resolution refers to the number of vertical lines contained in an image and is proportional to bandwidth. While vertical and horizontal resolutions can differ, they are typically designed to be equal (Van Lysel, 2000). The number of raster lines and the bandwidth of the system can be found in the user manual or by contacting the manufacturer. The radiology staff may also be able to provide this information.

Temporal Resolution

Temporal resolution is a factor of fluoroscopy pulse rate during the VFSS exam (image registration rate), but will also vary depending on frame rate of the system used to capture or record the video (video recording frame rate). These are two very different and distinct features; one related to the fluoroscopy equipment and the other to the video recording device. Historically, the terminology of 30 frames per second has been confusing and prone to misinterpretation because it has been used to describe both the fluoroscopy image registration rate and video recording frame rates. Image registration rates for pulsed fluoroscopy are typically described by reporting the pulse rate. For continuous fluoroscopy, however, there is no pulse rate information that can be reported; in the dysphagia literature the image registration rate of continuous fluoroscopy has often been reported 30 frames per second. This is not to be confused with the frame rate of the video recording.

The standard video recording system in North America has a video frame rate of *30 frames per second*; in Europe, Australia, Japan and South America, standard video frame rates are slightly lower at 25 frames per second. These frame rates correspond to the upper temporal resolution achievable for a video recording and are independent of the fluoroscopy system. What is important to realize is the fact that temporal resolution of any given VFSS recording will be determined by the lowest resolution of either the fluoroscopy equipment or the recording settings. For example, if a VFSS is performed using a pulse rate of *15 pulses per second* (yielding 15 images per second) and a recorded at a video frame rate of *30 frames per second*, the temporal resolution will in fact be *15 frames per second*. In this situation, each image from the fluoroscopy will be displayed twice (or over 2 successive video frames) in the video recording.

While the CASLPO Guidelines (2007) recommend recording VFSS at 30 frames per second, no recommendations are in terms of the imaging mode (i.e., continuous or pulsed fluoroscopy) or image registration rates (i.e., minimum pulse rate). Clinicians need to be aware that trade-offs in temporal resolution occur when reducing radiation exposure through pulsed fluoroscopy at rates below 30 pulses per second. Using 30 images per second, Cohen (2009) demonstrated that the depth and severity of transient penetration-aspiration was apparent on only a single frame in 70% of children (age range of 1 month to 3 years 9 months); consequently, if image registration rates lower than 30 pulses per second had been used in this study, the severity of penetration-aspiration would have been under-estimated. Similar conclusions are reported by Bonilha, Blair and colleagues (2013), who contrived to present 30-image-per-second VFSS recordings in full resolution and half resolution (i.e., 15 images per second) for interpretation by trained S-LPs. They demonstrated that the lower resolution was insufficient to capture swallowing events: differences were observed in numerous MBSImp™© measurements as well as in Penetration-Aspiration Scale (PAS) Scores. In a related study (Bonilha, Humphries et al., 2013), the authors also showed that the availability of 30 images per second leads to a more efficient VFSS examination, thereby limiting radiation exposure; procedures yielding only 15 images per second may require a larger number of swallowing tasks in order to adequately capture swallowing impairment.

Video Capture Considerations

As previously mentioned, CASLPO Practice Standard and Guidelines require S-LPs to record the videofluoroscopic exam for post-VFSS analysis (CASLPO, 2007). A wide array of commercially available hardware and software combinations exist for video capture. This section does not seek to provide an extensive review of available products but rather to explain the basic principles behind these video capture methods. In determining the best video capture method, both the radiology staff and information management systems should be consulted. Digital video capture systems will specify minimum requirements for computer processor speed, RAM, graphics cards and the computer monitor, due to the fact that digital video conversion requires a lot of processing power. When high quality video recordings are viewed on computers with inadequate processing power, this can result in poor video viewing conditions, such as periodic freezing and an inability to view all recorded frames.

If the VFSS is recorded to a computer, there is a one major component present: a digital video converter, which is used to transfer the video signal from the fluoroscope to a computer system. Converters typically come with preferred software, which can be used to capture the video. Depending on the software, a number of different digital file formats can be selected. The choice of format and settings will impact the quality of the video recording. Compression software may also be available to restrict file size to reduce burden to the hospital servers, data storage, and transmission rates (Hirshfeld, et al., 2004). While the details of video settings and compression codes are not covered in this article, it is important for S-LPs to be aware that choices in compression may influence video quality for post VFSS analysis. Each video compression tool differs in the method or algorithm it uses to reduce the file size; additional details regarding these methods can be found with a simple internet search. If compression is required in a hospital in order to limit file size for storage, S-LPs may want to explore optimal choices by preparing different versions of a set of VFSS and comparing observations and feedback regarding image quality across clinicians.

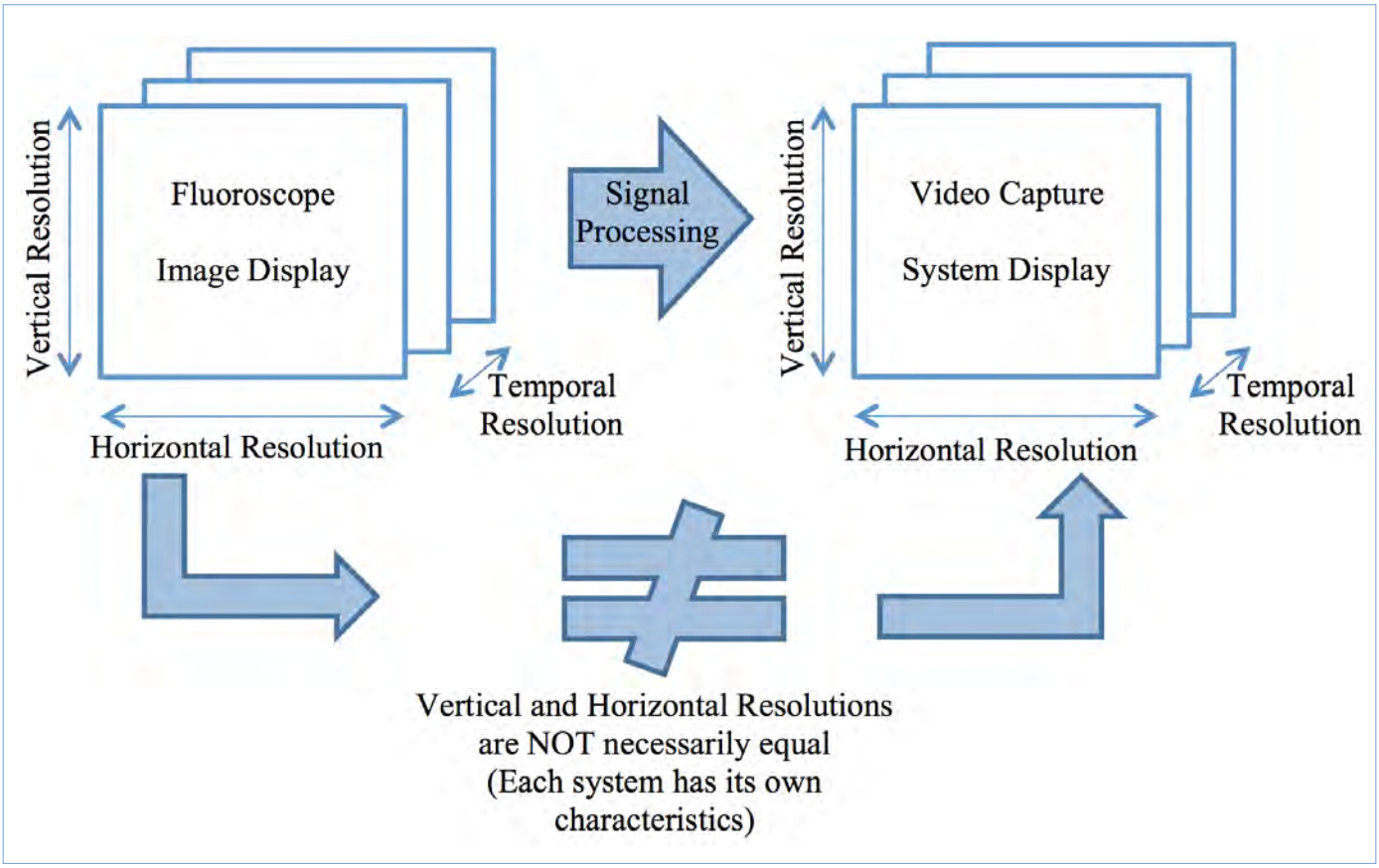
A downscanner (scan converter) may be required to reduce the data rate of the video stream between the fluoroscope and the recording system. Scan conversion is a video processing tool that changes the horizontal frequency or bandwidth (refer to Spatial Resolution Section for definition) to reduce the video data rate. This conversion creates compatibility with conventional video equipment and recording devices. In North America, the National Television System Committee (NTSC) defines the standard. However, video streaming rate and data format standards vary across countries (e.g., PAL, SECAM, NTSC). Depending on whether the fluoroscopy system used is analog or digital, and whether the video capture method is digital (e.g., computer) or analog (e.g., VHS tape), analog-to-digital or digital-to-analog converters may be required. Some fluoroscopic units already integrate this type of conversion prior to video display on the monitor (Schueler, 2000).

Figure 2 provides a schematic summary of all of the previously discussed issues with respect to fluoroscope and image resolution considerations for VFSS.

Safety and Radiation Exposure

Optimal methods should be used to ensure that the patient's exposure to radiation is kept to a minimum. Radiation dose is measured in millisieverts (mSv) and is present in everyday activities such as flying in a plane (0.005 mSv per hour) or smoking cigarettes (0.18 mSv per half pack) as well as during medical tests such as a chest x-ray (0.02 mSv) or a CT scan (10 mSv) (Green, 2011). On average, Canadians are exposed to approximately 2-4 mSv of background radiation per year (Health Canada, 2011). In a workplace, including work with x-ray equipment, the radiation exposure limit is 50 mSv in a single year and 100 mSv over 5 years, according to the Canadian Nuclear Safety Commission (Health Canada, 2011).

Figure 2: Illustration of fluoroscope and video capture image features



In a VFSS, radiation dose is typically controlled by the x-ray technician or radiologist. However, it is important for S-LPs to have some basic understanding of the factors influencing the radiation dose as it can affect the health of both themselves and their patients. The radiation dose is a factor of:

- Equipment used (design)
- Equipment set-up (equipment parameters)
- Equipment maintenance
- Proper utilization of the equipment
- Knowledge and skill of the radiologist or radiology technician
- Use of personal protective equipment
- Position or proximity to the equipment or radiation source

The voltage chosen by the radiology staff impacts both the dose and the image contrast. Increasing the voltage, in kV, reduces skin exposure to the beam because higher values have increased penetration. However, increased voltage can compromise the image contrast.

For patient radiation exposure, two factors that can in part be controlled by the S-LPs are the magnification and the imaging time. The magnification mode or Field of View (FOV) impacts the radiation exposure of the patient. While the energy released remains the same, the absorbed dose to the region of tissue does change. For a 2 fold magnification, the dose increases fourfold (IAEA, 2012). The second factor is the imaging time, which has a large impact on the overall radiation exposure. The shortest videofluoroscopic times should be used while ensuring adequate information is obtained for the clinical analysis. This is consistent with the radiological principle ALARA, "as low as reasonably achievable".

The major source of occupational radiation exposure during this procedure is due to scattered radiation (McLean, Smart, Collins, & Varas, 2006). S-LPs can help to reduce their own radiation exposure by using personal protective equipment (PPE), increasing their distance from the radiation source, and limiting the exposure time. A position 6 feet away from the patient in any direction is considered to be a zero exposure location.

The type of PPE used in a typical VFSS includes a lead apron and a thyroid guard. Eye protection and lead gloves may also be available. It is not recommended to use lead gloves when administering stimuli to a patient as a radiopaque substance in the FOV will cause fluctuations in image contrast and radiation due to the use of the automatic brightness control (ABC) (briefly mentioned in the image contrast section) (Kelchner, 2004). There exists a delicate balance between radiation dose and image quality. It is imperative that the S-LP works with the radiology staff to minimize radiation exposure while ensuring that an adequate video is recorded for the swallowing assessment.

Radiation exposure follows the inverse square law, therefore, increasing the distance by a factor of two results in a fourfold decrease in radiation exposure. It is recommended that the S-LP stand as far from the patient and the source of radiation as feasible. Exposure time is directly related to the radiation exposure. Therefore, a reduction in time by half results in half the radiation to both the patient and the attending S-LP. Once again, the shortest video fluoroscopic times should be used while ensuring adequate information is obtained for the clinical analysis.

Ionizing radiation exposure can be monitored using dosimeters. One dosimeter should be placed outside the lead apron in the neck area. A second dosimeter is recommended and should be placed under the apron (ASHA, 2004). Safety and monitoring device requirements are specific to a hospital and policies should be reviewed prior to the VFSS. The approximate radiation dose to an S-LP during a typical VFSS exam is summarized in Table 1.The median levels of radiation exposure associated with a VFSS in patient populations have been quantified by researchers and the exposures summarized in Table 2.

Table 1: Staff (S-LP) ionizing radiation exposure during a VFSS

Study	Length of Time Measured	System Used	Location of Measurement	Exposure
McLean et al (2006)	Average per procedure with an average procedure time varying from 3.0 to 3.6 min	Unknown for all sites	Outside thyroid shield	Site 1- 17 µGy Site 2 - 3.2 µGy Site 3- Below detection limit of 6µGy
			Under S-LP lead apron at waist level	All 3 sites - Below detection limit of 6µGy
Crawley et al (2004)	Total for 21 exams over 6 months with a mean screening time of 3.7 min (range: 2.5-4.3 min)	Siemens Uroskop C2	Under S-LP lead apron	Below detection limit of 0.3mSv
			Finger on right hand of right handed operators	0.9mSv
			Forehead of operators	0.5mSv

Table 2: Patient ionizing radiation exposure during a VFSS

Study	System Used	Sample Size (# of Patients)	Mean Equipment Parameters	Mean DAP (Gy cm²)	Mean Effective Radiation Dose (mSv)	Mean Length of Exam (s)
Zammit-Maempel, Chapple, & Leslie (2007)	Siemens Sireskop 5-45	230	77kVp	1.6 (0.05-10)	0.20 (0.01-1.4)	181 (18-564)
Moro & Cazzani (2006)	Prestige VH by GE	22	78.4kV 0.7mA	2.3 (1-5.4)	0.4 (0.17-0.92)	155 (84-306)
Crawley, Savage, & Oakley (2004)	Siemens Uroskop C2	21	N/A	3.5 (3.1-5.2)	(Median 0.85) (0.76-1.3)	220 (150-258)
Wright, Boyd, & Workman (1998)	Siemens Siregraph 2	23	65.4kV	4 (0.28-9.74)	0.4 (0.027-1.1)	286 (32-497)

Radiology staff members have a wealth of knowledge about radiation and mitigation strategies for both staff and patients. Dialogue between the S-LPs and radiology staff is highly encouraged in order to minimize the radiation risk to everyone present and to obtain the highest quality VFSS data acquisition.

Conclusion

This article described the types of fluoroscope available, factors influencing the image contrast, the creation of a contrast impregnated fluid, imaging techniques (pulsed versus continuous), imaging resolution (spatial and temporal), and safety considerations. The optimal videofluoroscopy features can only be successfully obtained when both S-LPs and radiology staff work together as a team. This article aimed to describe fundamental fluoroscopy features as applied to the VFSS for S-LPs to effectively communicate with the radiology staff. In addition, clarification of fluoroscopic imaging terminology was provided with the hopes of avoiding future terminology misuse in publications relating to videofluoroscopic studies.

References

ACSLPA. (2009). *Guideline: Swallowing (dysphagia) and feeding.* Retrieved from http://www.acslpa.ab.ca/public/data/documents/Dyshagia_-_single-sided_-_Final.pdf?0E6CA8F4-6866-4150-8A7D17E228FF660D

ASHA. (2004). *Guidelines for speech-language pathologists performing videofluoroscopic swallowing studies.* Retrieved from http://www.asha.org/policy/GL2004-00050.htm

Bonilha, H. S., Blair, J., Carnes, B., Huda, W., Humphries, K., McGrattan, K., . . . Martin-Harris, B. (2013). Preliminary investigation of the effect of pulse rate on judgments of swallowing impairment and treatment recommendations. *Dysphagia.* Online first. doi: 10.1007/s00455-013-9463-z

Bonilha, H. S., Humphries, K., Blair, J., Hill, E. G., McGrattan, K., Carnes, B., . . . Martin-Harris, B. (2013). Radiation exposure time during MBSS: Influence of swallowing impairment severity, medical diagnosis, clinician experience, and standardized protocol use. *Dysphagia, 28*(1), 77-85.

Bushberg, J. T., Seibert, J. A., Leidholdt, E. M., & Boone, J. M. (2012). *The Essential Physics of Medical Imaging.* Philadelphia: Wolters Kluwer Health/Lippincott Williams & Wilkins.

CASLPA. (2007) . CASLPA Position paper on dysphagia in adults. Retrieved from http://www.caslpa.ca/PDF/position%20papers/English_Dysphagia_June%202007.pdf

CASLPO. (2007). Practice Standards and Guidelines for Dysphagia Intervention by Speech-Language Pathologist. Retrieved Oct 26, 2012, from http://www.caslpo.com/portals/0/ppg/dysphagia_psg.pdf

Cerveri, P., Forlani, C., Borghese, N.A., & Ferrigno, G. (2002). Distortion correction for x-ray image intensifiers: Local unwarping polynomials and RBF neural networks. *Medical Physics, 29*(8), 1759-1771.

Clave, P., Arreola, V., Romea, M., Medina, L., Palomera, E., & Serra-Prat, M. (2008). Accuracy of the volume-viscosity swallow test for clinical screening of oropharyngeal dysphagia and aspiration. *Clinical Nutrition, 27*(6), 806-815.

CMPA. (2012). *Risk management when using drugs or medical devices off-label*. Retrieved from http://www.cmpa-acpm.ca/cmpapd04/docs/resource_files/perspective/2012/03/pdf/com_p1203_15-e.pdf

Cohen, M. (2009). Can we use pulsed fluoroscopy to decrease the radiation dose during video fluoroscopy feeding studies in children. *Clinical Radiology , 64*, 70-73.

Crawley, M., Savage, P., & Oakley, F. (2004). Patient and operator dose during fluoroscopic examination of swallow mechanism. *British Journal of Radiology , 77*, 654-656.

E-Z-EM Canada Inc. (2012). Liquid Polibar Plus Barium Sulfate [Product Label]. Canada

Fink, T. A., & Ross, J. B. (2009). Are we testing a true thin liquid? *Dysphagia, 24*(3), 285-289.

Green, H. (2011, Jun 23). *Radiation exposure and air travel – Should we worry?* Retrieved from http://www.farecompare.com/travel-advice/radiation-exposure-and-air-travel-should-we-worry/

Health Canada. (2011, Nov 2). *Occupational exposure to radiation.* Retrieved from http://www.hc-sc.gc.ca/hl-vs/iyh-vsv/environ/expos-eng.php

Hirshfeld, J.W. Jr., Balter, S., Brinker, J. A. , Kern, M. J., Klein, L. W., Lindsay, B. D., Tommaso, C. L., Tracy, C. M., Wagner, L. K., Creager, M. A., Elnicki, M., Hirshfeld, J.W. Jr., Lorell, B. H., Rodgers, G. P., Tracy, C. M., Weitz, H. H. (2004). ACCF/AHA/HRS/SCAI clinical competence statement on physicianknowledge to optimize patient safety and image quality in fluoroscopicallyguided invasive cardiovascular procedures. A report of the American College of Cardiology Foundation/American Heart Association/American College of Physicians Task Force on Clinical Competence and Training. *Journal of the American College of Cardiology, 44*(11), 2259-2282.

IAEA. (2012). *Fluoroscopy.* Retrieved from https://rpop.iaea.org/rpop/rpop/content/informationfor/healthprofessionals/1_radiology/fluoroscopy.htm

Jones, B., & Donner, M. W. (1989). How I do it: Examination of the patient with dysphagia. *Dysphagia, 4*(3), 162-172.

Kelchner, L.N. (2004). Radiation safety during the videofluoroscopic swallowing study: The adult exam. *Perspectives on Swallowing and Swallowing Disorders (Dysphagia)*, 13(3), 24-28.

Logemann, J.A. (1993). *Manual for the videofluorographic study of swallowing: Second edition.* Austin, TX: Pro-Ed, Inc.

Logemann, J. (1997). Role of the modified barium swallow in management of patients with dysphagia. *Otolaryngology- Head and Neck Surgery* , 116(3), 335-338.

Martin-Harris, B., Brodsky, M. B., Michel, Y., Castell, D. O., Schleicher, M., Sandidge, J., . . . Blair, J. (2008). MBS measurement tool for swallow impairment--MBSImp: Establishing a standard. *Dysphagia, 23*(4), 392-405.

Martin-Harris, B., & Jones, B. (2008). The videofluorographic swallowing study. *Physical Medicine and Rehabilitation Clinics of North America, 19*(4), 769-785.

McLean, D.M., Smart, R., Collins, L., & Varas, J. (2006). Thyroid dose measurements for staff involved in modified barium swallow exams. *Health Physics., 90*(1), 38-41.

MSHA. (2009). *Guidelines for provision of dysphagia services (for adults).* Retrieved from http://www.msha.ca/documents/1-GuidelineforProvisionofDysphagiaServicesforAdults.pdf

Moro, L., & Cazzani, C. (2006). Dynamic swallowing study and radiation dose to patients. *La Radiologia Medica, 111*, 123-129.

Nickoloff, E. L. (2011). AAPM/RSNA physics tutorial for residents: Physics of flat-panel fluoroscopy systems. *Radiographics, 31*, 591-602.

Rauch, P. (2008). *Fluoroscopic imaging equipment considerations for optimization of performance and dose* [PowerPoint slides]. Retrieved from http://www.aapm.org/meetings/amos2/pdf/35-9917-69462-287.pdf

Royal College of Speech and Language Therapists (2012) *Videofluoroscopic evaluation of oropharyngeal swallowing disorders (VFS): The role of speech and language therapists.* Retrieved from http://www.rcslt.org/news/docs/rcslt_vfs_position_paper_consult (Not referenced in the text)

Schueler, B. (2000). The AAPM/RSNA physics tutorial for residents: General overview of fluoroscopic imaging. *Radiographics, 20*(4), 1115-1126.

Steele, C. M., Molfenter, S. M., Péladeau-Pigeon, M., & Stokely, S. (2013). Letter to the Editor: Challenges in preparing contrast media for videofluoroscopy. *Dysphagia.* [Epub ahead of print]

Van Lysel, M. (2000). The AAPM/RSNA physics tutorial for residents: Fluoroscopy: Optical coupling and the video system. *Radiographics*, *20*(6), 1769-1786.

Wright, R., Boyd, C., & Workman, A. (1998). Radiation doses to patients during pharyngeal videofluoroscopy. *Dysphagia, 13*, 113-115.

Zammit-Maempel, I., Chapple, C., & Leslies, P. (2007). Radiation dose in videofluoroscopic swallow studies. *Dysphagia, 22*(1), 13-15.

Authors' Note

Correspondence concerning this article should be addressed to Melanie Peladeau-Pigeon, M.H.Sc., Toronto Rehabilitation Institute, University Health Network, 550 University Avenue, Toronto, ON, M5G 2A2. Canada. Email: Melanie.Peladeau-Pigeon@uhn.ca

Received date: November 6, 2012

Accepted date: July 15, 2013

Appendix

Fluid barium mixture concentration and density calculations

The following are calculations for a total volume of 250mL with a desired barium concentration of 22% w/v (weight of barium to volume of total solution).

Given that Liquid Polibar contains 100% w/v or 56% w/w (E-Z-EM, 2012), as shown on the product label then the following calculations can be used:

Inputs:

(1) Liquid Polibar Concentration = 100% w/v (100g Barium / 100mL Liquid Polibar Solution)

(2) Liquid Polibar Concentration = 56% w/w (56g Barium / 100 g Liquid Polibar Solution)

(3) Total Mixed Solution Volume = 250mL

(4) Mixed Solution Concentration = 22% w/v (22g Barium / 100mL Mixed Solution)

Calculations:

(1) *Mass of Barium Desired (g)*

$$= Total\ Mixed\ Solution\ Volume\ (mL) * Mixed\ Solution\ Concentration\left(\frac{g\ Barium}{mL\ Mixed\ Solution}\right)$$

$$= 250mL\ Mixed\ Solution\left(\frac{22g\ Barium}{100mL\ Mixed\ Solition}\right)$$

$$= 55g\ Barium$$

(2) Mass of Liquid Polibar Desired (g)

$$= \frac{Mass\ of\ Barium\ Desired\ (g)}{Liquid\ Polibar\ Concentration\left(\frac{g\ Barium}{gLiquid\ Polibar\ Solution}\right)}$$

$$= \frac{55g\ Barium}{56gBarium/100g\ Liquid\ Polibar\ Solution}$$

$$= \sim 98g\ Liquid\ Polibar\ Solution$$

NOTE: If a scale is not readily available, the mass of barium required can be converted to volume of Liquid Polibar solution:

Volume of Liquid Polibar Desired (mL)

$$= \frac{Mass\ of\ Barium\ Desired\ (g)}{Liquid\ Polibar\ Concentration\left(\frac{g\ Barium}{mL\ Liquid\ Polibar\ Solution}\right)}$$

$$= \frac{55g\ Barium}{100gBarium/100mL\ Liquid\ Polibar\ Solution}$$

$$= 55mL\ Liquid\ Polibar\ Solution$$

(3) Add the desired mass or volume of Liquid Polibar and fill the remainder of the desired 250mL with the fluid of interest (e.g., water)

Increasing Inferential Reading Comprehension Skills: A Single Case Treatment Study

Augmenter les habiletés de compréhension en lecture à l'aide d'inférences : une étude de traitement

Laura B. Green
Karen L. Roth

KEY WORDS

INFERENCES

READING COMPREHENSION

LANGUAGE/
LEARNING DISABILITIES

Abstract

This pilot study investigated the effects of an inferential reading comprehension intervention program implemented in a public school setting with a fourth grader with a language disorder. The 8-week treatment period involved teaching a systematic approach for differentiating literal and inferential question types and specific strategies for answering the latter. Improvement was noted in the ability to answer inferential comprehension questions after reading a passage and in standardized reading comprehension test performance. No change was noted on a receptive vocabulary control measure. These results lend preliminary support to the effectiveness of this intervention approach.

Abrégé

Cette étude pilote explorait les effets d'un programme d'intervention sur la compréhension en lecture à l'aide d'inférences mis en œuvre dans une école publique avec un élève de quatrième année atteint d'un trouble du langage. La période de traitement de huit semaines comprenait l'enseignement d'une approche systématique de différenciation entre les questions littérales et celles nécessitant des inférences ainsi que des stratégies particulières de réponses à ce type de questions. On a noté une amélioration dans la capacité de répondre aux questions de compréhension après la lecture d'un passage requérant des inférences et dans les performances à un test standardisé de compréhension en lecture. Aucun changement n'a été noté sur une mesure de contrôle du vocabulaire réceptif. Ces résultats offre un appui préliminaire à l'efficacité de cette approche d'intervention.

Laura B. Green, Ph.D. CCC/SLP
Department of Communication Sciences and Disorders
Texas Woman's University
P. O. Box 425737
Denton, TX 76204
U.S.A

Karen L. Roth, M.S. CCC/SLP
Arizona State University
411 N Central Ave, Phoenix, AZ 85004
U.S.A

When children move into the upper primary grades, there is a shift in instructional emphasis from learning to read, or decoding, to reading to learn, or comprehension. There are very few areas of the school curriculum that don't require the ability to read and understand (McGee & Johnson, 2003). Reading requires active participation in the search for meaning. Text comprehension is critical to academic success and must be approached in a purposeful way.

Comprehending a passage is a complicated process in that the text's meaning is a combination of the explicit, literal meanings of the words and sentences, as well as the inferred meanings that can be uniquely generated by the reader. Given that meaning is not given solely in the text, but is mentally constructed by readers during the reading process (Maria, 1990), the total message within the written discourse is dependent upon the reader applying additional knowledge and "reading between the lines." An author does not explicitly state all of the information necessary for comprehension, as this would be both laborious and redundant (Gillam, 2007). The key to the generation of the implicit meanings involves the reader's ability to make inferences. The ability to generate inferences is an essential skill that greatly determines the degree to which a passage will be understood (Cain, Oakhill, & Elbro, 2003; Casteel, 1993; Omanson, Warren, & Trabasso, 1978; Zabrusky, 1986). Without explicit training, it is more difficult for children to answer inferential questions about a text than literal ones (Hansen & Pearson, 1983)

Given the impact of inferential ability on successful comprehension, students with language/ learning disabilities, who often struggle to understand what they hear and read, may exhibit specific difficulties with this skill. Research supports this conclusion (Adams, Clarke & Haynes, 2009; Dodwell & Bavin, 2008; Laing & Kamhi, 2002; Scannell-Miller, 1982). While some studies have focused on children with specific language difficulties and others on children with learning disabilities (e.g., reading decoding and/or comprehension difficulties), several common characteristics have emerged with regard to their listening and reading comprehension skills. Regardless of whether a deficit is inherent in basic language skills, reading, or both, there is evidence that these students engage in inferential processing less often and less effectively than their normally-achieving peers during both listening and reading comprehension tasks. Specifically, in the context of generating inferences in listening comprehension tasks, participants with language/learning disabilities tend to perform like younger children (Adams, Clark & Haynes, 2009; Scannell-Miller, 1982), to make more errors on inferential questions than their typically developing peers (Dodwell & Bavin, 2008), and to generate fewer inferences than their age mates (Laing & Kamhi, 2002; Snyder, 1984). In the context of reading comprehension activities, students with language/learning disabilities have been found to demonstrate the ability to infer but need more direction about the nature and appropriateness of inferential strategies (Wong, 1988). Additionally, they produce similar proportions of inferences during free recalls of a target passage but of a different quality (Tierney, Bridge & Cera, 1978), answer fewer inferential questions correctly (Oakhill, 1984) and provide more illogical, intuitive answers to inferential comprehension than their typically developing peers (Wilson, 1979). Thus, students with language/learning disabilities struggle with inferential processing and, as a result, struggle with comprehension.

These students' difficulties with inferential tasks may occur for a variety of reasons. One possibility is that they are not given enough practice with this type of reasoning. Activities within basal readers tend to include and classroom teachers tend to ask more literal than inferential questions (Guszak, 1967; Hansen, 1981). In addition, studies of classroom interaction suggest that the lower-achieving students or poorer readers are asked fewer inferential questions than are the better readers (Sadker & Sadker, 1982). Another source of difficulty may be that poor readers do not consistently or effectively use their prior knowledge to answer inferential questions (Gillam, 2007) and, even with accurate prerequisite information, answer them less effectively (Holmes, 1984). There is also evidence that poor readers produce fewer elaborations from prior knowledge during reading (Reder, 1980; Tierney et al., 1978). Inferential difficulty could also result from an overemphasis on background knowledge and subsequent formation of "intuitive" or tangential answers when prior knowledge overshadows text information (Williams, 1993). Erroneous conclusions may not be discarded and negatively influence future comprehension. Lastly, children with language/ learning disabilities might be "inactive learners" who do not activate selective attention and/or do not choose and employ appropriate cognitive strategies (Carr & Thompson, 1996). This lack of self-regulated learning is common in a large percentage of students with language/learning disabilities (Bashir & Singer, 2006; Graham & Harris, 2012; Wong, 1994) such that many have limited awareness of domain-specific knowledge, skills and strategies, how to apply them, and when to deploy them for effective and efficient task performance (Garner, 1990; Troia, 2002).

Given the importance of inferential thinking to successful reading comprehension and academic success, there is a need for an intervention program that efficiently addresses all of these potential difficulties. Several intervention programs have been created to improve inferential comprehension skills for students with language/learning disabilities. McGee and Johnson (2003) taught less skilled

"comprehenders" between the ages of 6 and 9 how to make inferences through question generation and prediction, which resulted in improved reading comprehension performance after 6 training sessions. Hansen and Pearson (1993) made good and poor fourth grade readers aware of the importance of drawing inferences, activated background knowledge prior to reading, encouraged predictions based on this knowledge, and provided opportunities for practice answering inferential questions. Results showed that poor readers benefited significantly from the instruction. Holmes (1984) taught disabled readers a structured inferential comprehension strategy involving the use of key words in the text and self-questioning in the context of reading materials arranged from easy to more difficult. The group that had both strategies and graded materials scored significantly better than the other experimental groups on inference question tasks. Carr, Dewitz, & Patberg (1989) developed the Inferential Training Technique (ITT) for expository text, which includes a modified cloze procedure to introduce and model the strategy and a self-monitoring checklist to transfer the strategy to new situations. The exercises focus attention on text clues and relate text information to prior knowledge to fill in the cloze blanks. They found that ITT was successful in improving comprehension and comprehension monitoring (Carr, Dewitz & Patberg, 1989). Brown, Palinscar, and Armbruster (1984) assessed the effects of explicit training in comprehension for children who had poor comprehension by using a gradual transfer of responsibility for asking inferential questions from the teacher to the child. Children were first taught directly and then gradually regulated their own activity in summarizing, questioning, and predicting text content and they improved performance on comprehension exercises which also generalized to other text-based tasks. In sum, teacher modeling, active student engagement, and strategy use appear to be important elements in comprehension intervention.

Based on the inferential difficulties faced by students with language/learning disabilities and treatment studies that have been conducted, improved inferential comprehension requires the following: (1) awareness of, exposure to, and practice with inferential reading comprehension questions; (2) activation of prior knowledge prior to and appropriate application of while answering inferential questions; (3) appropriate interpretation of background information provided in the text; and (4) self-regulated learning via active use of inferential comprehension strategies. Self-regulated learners establish and maintain motivation, use supports when help is needed, mediate performance with language, and understand how and when to use specific strategies (Schunk & Ertmer, 2000).

While the treatment studies described earlier each include several of these four elements, Milosky and Ford (1996) developed a multi-faceted inferential comprehension intervention that includes all four. Students first learn to identify and distinguish between literal (e.g., the answer can be found in the passage) and inferential questions, specifically recognizing the nature of an inferential question (e.g., the answer is not in the passage). They are then taught to activate their background knowledge and to make appropriate predictions during reading, shown how individual words add meaning to a sentence and can be used to activate their knowledge efficiently, and taught an inference-specific strategy that involves a "puzzle metaphor" to guide self-regulated thinking/learning. Specifically, they identify an inferential ("puzzle") question and then "put the pieces together" through self-talk (e.g., *Is there a sentence in the book that gives the answer? If "No" then this is a puzzle question. What information in the book will help me? What else do I know about _____ that will help me? How does this information fit together to create an answer that makes sense?*). These skills are modeled by the teacher, written out on a help sheet, and practiced initially in controlled and scaffolded activities. Students then progress to applying their knowledge and to using this process in the context of functional academic material. To date, no data are currently available as to the success of this program, nor have any inferential comprehension intervention studies been conducted in a typical, public school therapy setting.

The last issue that needs to be considered is the concept of evidence-based practice (EBP), or "the conscientious, explicit and judicious use of current best practice in making decisions about the care of individual patients… by integrating individual clinical expertise with the best available external clinical evidence from systematic research" (Sackett, Rosenberg, Gray, Haynes, & Richardson, 1996, p. 71). The potential benefits of EBP include bridging the divide between research and practice, improving clinical services, making clinicians more accountable, and reducing variation in service provision (Zipoli & Kennedy, 2005). The American Speech-Language-Hearing Association (ASHA) has issued an official policy stating that speech-language pathologists integrate the principles of evidence-based practice into the clinical decision-making process (ASHA, 2005). Yet, in the area of language disorders in children, there is not a large body of clinical research evidence available (Cirrin & Gillam, 2008). In their review of the last two decades of language intervention studies in search of those that met Level 1 (randomized clinical trials) and Level 2 (non-randomized comparison studies or multiple baseline single participant designs) evidence requirements, they only found 21 studies that met these criteria. And, many of those studies that did qualify were conducted with preschool (i.e., 3-4-year-old) children.

Therefore, the goals of the current study were to document improved inferential comprehension skills after implementing a multi-faceted treatment program in a public school setting and to add to the evidence base in child language intervention using a Level 3b (Phillips et al., 2001; http://www.cemb.net/levels_of_evidence.asp) quasi-experimental single participant treatment design. Specifically, the purpose of the present investigation was to examine improvement in the inferential reading comprehension skills of a fourth grader with a language disorder following implementation of the Milosky and Ford (1996) treatment paradigm.

Method

Design

Treatment was administered in the context of an ABABABA design with an initial triple baseline (i.e., the first A, or no treatment, phase) to ensure stability of the target behavior. Interspersed within the B (i.e., intervention) phases, treatment was withdrawn for a week creating the latter A (i.e., no treatment) phases to examine performance in the absence of intervention. Standardized pre- and post-test measures were also utilized to further examine behavior change. The pretest, treatment and posttest schedule is documented in Appendix A.

Participant

One fourth grade Caucasian male (age 10;6), Z, was selected for treatment based on the following characteristics: He demonstrated a language disorder and qualified as a student with a speech/language impairment based on Washington State criteria (i.e., "2(l) Speech or language impairment means a communication disorder, such as stuttering, impaired articulation, a language impairment, or a voice impairment, that adversely affects a student's educational performance."), was served by his public school's speech/language pathologist, received no other special education services (e.g., content mastery), received language therapy for expressive and receptive language delays, spoke English as a first language, and had current treatment goals addressing improved comprehension, categorization, and oral narrative production. In addition, the student's teacher reported that he was struggling with reading comprehension because he was "concrete and didn't draw conclusions." Prior to the study, a standardized measure of reading comprehension and of receptive vocabulary were administered to gain additional information about vocabulary and reading abilities relevant to the investigation. These instruments then served as pre- and post-test measures. The second author received informed parental consent to share study-related assessment and treatment data.

Pre- and Post-Test Assessment Measures

Standardized Reading Comprehension Test

Reading comprehension was assessed through standardized administration of the Gates-MacGinitie Reading Test (GMRT) 3rd Edition Passage Comprehension subtest (MacGinitie & MacGinitie, 1989). Z scored in the below average range on this subtest, indicating specific difficulty answering reading comprehension questions.

Control Measure

The Peabody Picture Vocabulary Test – 3 (PPVT-3) (Dunn & Dunn, 1981) was administered as a pre- and post-treatment control measure. While not an identical task to answering comprehension questions, both are receptive language skills for which application of contextual knowledge can be a contributor and are reliable standardized tests on which student's performance would typically remain fairly consistent. Receptive vocabulary was not targeted during intervention; thus, we conceptualized this task as a quasi-control for developmental changes to increase our confidence that any change in inferential question-answering ability could be attributed to the treatment targeting it. We note that there are flaws in this argument due to differences in the types of knowledge required for inferential comprehension versus comprehension of vocabulary. Additionally, the use of standardized pre- and post-test measures creates the risk of a practice effect that offsets a change in behavior that is not attributable to the treatment. If this were the case, improved performance on both standardized measures would be expected.

Standardized pre-test scores are detailed in Table 1.

Informal Inferential Measure

To further examine inferential abilities in reading comprehension, an informal inventory of Z's errors on the Gates-MacGinitie Reading Comprehension subtest (MacGinitie & MacGinitie, 1989) was completed. This subtest is comprised of literal and inferential question types, both of which are defined in the test manual. At pretest, Z answered 34% (9/26) of the inferential questions correctly and 52% (10/19) of the literal questions correctly.

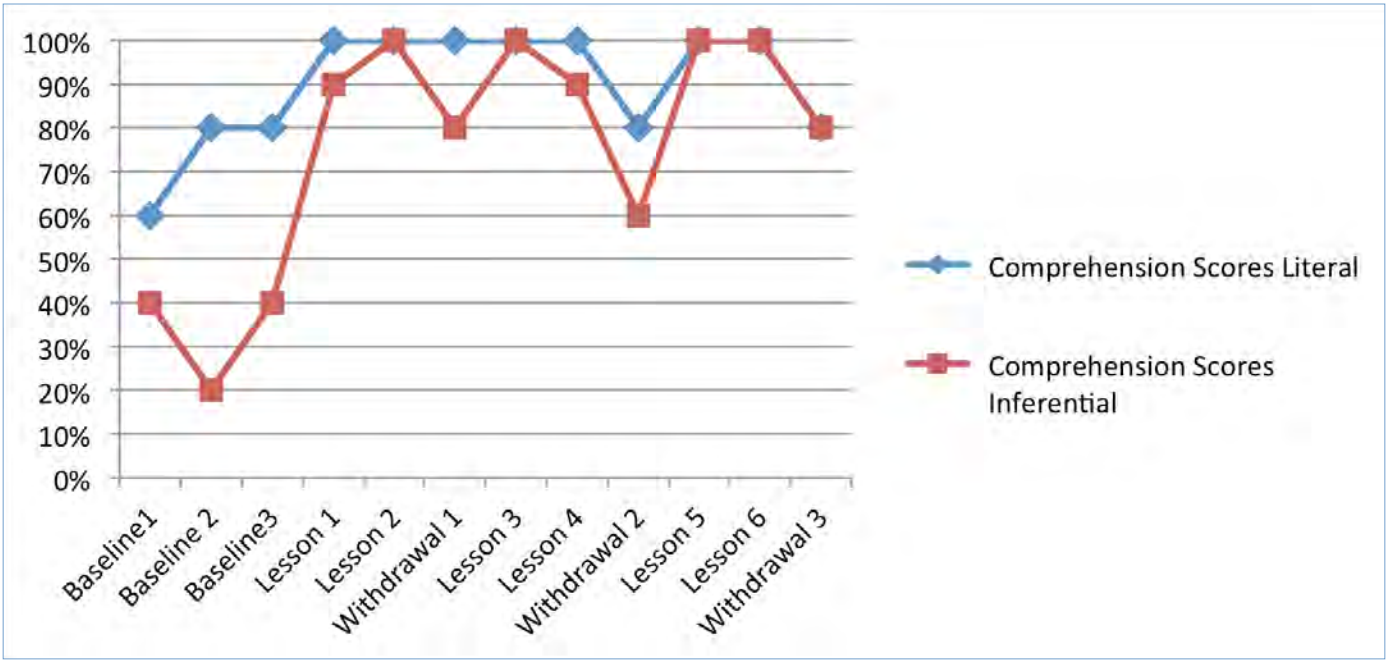
Baseline, Treatment and Withdrawal Reading Comprehension Tasks

The baseline, treatment and withdrawal comprehension tasks utilized reading materials taken from the Steck-Vaughn Level 3 Reading Comprehension workbook (Steck-Vaughn Company, 1999). The passages were narrative in nature, were written at a third grade reading level, and had a mean length of 250 words. Given Z's below average score on the GMRT subtest, the third grade readability

Table 1: Pre- and Post-Treatment Standardized Test Scores (percentile ranks)

Test	Percentile Rank scores (Pre)	Percentile Rank Scores (Post)
PPVT-III (Control Measure)	39	32
Gates-MacGinitie Reading Comprehension Subtest	3	30

Figure 1: Baseline, Treatment and Withdrawal Data: Percentage of Literal and Inferential Comprehension Questions Answered Correctly



level was selected to ensure Z's ability to focus on his use of the new comprehension strategies rather than struggle with vocabulary or decoding. Z was able to read the passages without assistance. Each passage was followed by ten reading comprehension questions written by both authors, five of which were literal (e.g., "Where did Joe go?") and five of which were inferential (e.g., "Why did Joe feel sad?"). Specifically, questions were classified as literal "if the student could answer by choosing a restatement of something stated explicitly in the passage." Questions were inferential if "the student could not answer the question by choosing a restatement of something stated explicitly in the passage (MacGinitie & MacGinitie, 1989). Once the 10 questions were formulated for each passage, they were rated as literal or inferential by an independent rater (a licensed speech/language pathologist) who was provided with the definitions described above. Only questions with 100% agreement between the authors and the independent rater were included in the study. See Appendix B for an example stories and questions.

Treatment

The participant received the inferential comprehension intervention during one weekly 40-minute session with another student in the speech room at his school during an 8 week period (6 weeks of intervention and 2 weeks with no treatment). See Appendix A for this schedule. The other student received the same intervention but did not participate in the study. Treatment was conducted by the second author, a certified speech/language pathologist. During the first treatment session, the 4-step inferential question-answering process was introduced and the students then utilized it while completing the first two reading passages and the ten comprehension questions that followed. Instruction, feedback, and scaffolding were provided throughout. Specifically, before reading the passage, the students were first asked to make predictions about the story based on the title, thus activating their background knowledge. After reading the passage silently, the clinician introduced the five literal and five inferential questions, which were in a random order. The clinician

explained the difference between literal and inferential ("puzzle") questions. They were taught that, if it was not a puzzle question (but a literal one), then they should be able to go back and find the answer directly written in the story. The clinician then explained that the answers to some questions were not found explicitly in the passage and were therefore "puzzle questions" (e.g., inferential questions). The students read the questions aloud and discussed whether each was a "puzzle question" or not. Once they demonstrated understanding of these two question types, the "puzzle question" strategy was introduced. When the students came to these inferential questions they were guided to do the following:

1. Find information in the book that will help (e.g., use key words in sentences as clues).
2. Think about information that they already know that will help (e.g., Ask yourself, "What do I already know about this?")
3. Then, talk to yourself about how the information fits together logically to help you figure out the answer (e.g., "How does what I know fit together with what I read?")
4. Then think to yourself, "Based on what I know and what I read, does my answer make sense?"

The clinician modeled the process by thinking aloud. She then prompted students to make predictions, consider the puzzle metaphor, and think about specific words in a sentence that might provide clues to the answer. Scaffolding (e.g., Cues such as, *What kind of question is this? What should you do to find the answer?*) was provided as necessary. When students accurately answered inferential questions, they were asked to state the "clues" that helped solve the "puzzle question." This allowed the clinician to ensure that they had used their strategies successfully.

Once the predicting, question identification, and "puzzle question" processes were initially taught, each treatment session followed the same schedule: brief review of literal and inferential question types, review of the strategies for answering literal and "puzzle" questions, and completion of two passage-reading and question-answering activities. Clinician modeling and scaffolding were provided as needed.

Data Collection and Analysis

Prior to, throughout, and following completion of treatment, data were collected on the total number of literal and inferential comprehension questions answered correctly on the passage-reading and question-answering activities. During treatment, these tasks were initially completed with clinician instruction, modeling, scaffolding (e.g., *Since this is a puzzle question, what is the next step?*) and feedback, which was faded as intervention progressed. During the treatment withdrawal weeks, Z completed the same type of passage-reading, question-answering task with no assistance from the clinician. Percentage correct scores were calculated separately for the literal questions and for the inferential questions. Data analysis consisted of visual inspection of baseline, treatment and withdrawal data, along with calculation of effect size and comparison of pre- and post-test measures.

Results

The purpose of this study was to determine if the inferential reading comprehension skills of a fourth grader with a language/learning disability could be improved using background knowledge activation, instruction in literal and inferential question types, and a "puzzle question" strategy (Milosky & Ford, 1996).

Baseline, Treatment and Withdrawal Data

Z completed the exact same type of reading and question-answering activity during the baseline, treatment and withdrawal phases of the study. Three of these reading and question-answering tasks were completed prior to treatment. Then, treatment data were taken on two of these tasks during each session as the clinician provided instruction and scaffolding in applying new knowledge and using the puzzle question strategy. After the 2nd, 4th and 6th weeks of treatment, the intervention was withdrawn and the same reading and question-answering task was completed with no assistance from the clinician. Z's literal and inferential comprehension scores are detailed in Figure 1. Per visual inspection of the data, it appears that there was a trend toward improvement once treatment began, with 100% non-overlapping data for inferential questions and 78% non-overlapping data for literal questions. A decrease in performance on both question types was noted during the withdrawal weeks (with the exception of performance on literal questions during the first withdrawal phase), also providing support for a treatment effect. In order to quantify the magnitude of the change in level of performance on inferential questions, a variation of Cohen's d was used to calculate an effect size (Kromrey & Foster-Johnson, 1996). While not ideal, this measure can be calculated with only one post-intervention measure (Beeson & Robey, 2006). The standardized mean difference formula

$$\frac{\text{X post-intervention} - \text{X pre-intervention}}{\text{SD pre-intervention}}$$

was applied using a post intervention mean of 80, a pre-intervention mean of 33, and a SD pre-intervention of 11.5, which yielded an effect size of 4.08. This effect size

is considered medium based on those reported for single-subject studies by Robey, Schultz, Crawford, & Sinner (1999).

Post Test Data

On the Gates-MacGinitie Passage Comprehension subtest, Z's percentile rank went from 3 to 30, the latter of which indicated performance within the average range. Post test scores are detailed in Table 1. Informal documentation of performance on inferential questions provides additional support for a treatment effect. At post-test, Z answered 61% (16/26) of the inferential questions correctly and 73% (14/19) of the literal questions correctly (an increase from his pretest 34% and 52% respectively).

Control Measure

The PPVT-III was administered pre and post treatment as a control measure. Given that receptive vocabulary was a comprehension skill not being taught, it was predicted that the participant's performance on this instrument would not change over the course of the treatment period. The post-treatment percentile rank was actually slightly lower than the pre-treatment score, indicating no change in receptive vocabulary performance.

Qualitative Observations

While data were not systematically collected on strategy use, the level of scaffolding provided, and the amount of cueing required during treatment, informal behavioral observations were made throughout. With respect to the processes for answering literal and inferential questions, Z initially relied on step-by-step instruction for answering both question types. After the first two sessions he was able to independently identify both the literal and inferential question types correctly. In addition, when he encountered a literal question, he consistently verbalized the definition (e.g., that he "could find the answer within the story"). After lesson 3, he was able to self-cue to return to the story and find clues to answer the inferential questions. Scaffolding was faded between lessons 2 and 4 and accurate responses to both question types were provided independently during lessons 5 and 6. With regard to stating the clues that supported his inferential answer choices, Z initially did not consistently relate relevant world knowledge (e.g., he made tangential, unrelated comments) and relied on clinician cueing to remain focused on the question. By lesson 5, however, he was more accurate at providing appropriate information support when answering an inferential comprehension question.

Discussion

This study provides preliminary evidence that use of an inferential comprehension intervention program can have a positive effect on student performance. Data from three of the analyses support this conclusion. First, improved performance (from the initial baseline) was seen on the total number of inferential questions answered correctly on the measures completed during both treatment and withdrawal (i.e., no treatment) phases and the effect size for the change in performance was considered medium. Interestingly, a slight decrease in the percentage correct scores was noted when the treatment was withdrawn. The decreases could support the treatment effect such that strategy use and subsequent comprehension performance were benefitting from the intervention and lagged slightly when it was removed. These decreases in comprehension scores could also be indicative of the length of time required for effective strategy learning and use. Given that the treatment involved activation of background knowledge, use of context clues, and implementation of the newly learned "puzzle question" strategy, a great deal of self-regulation was required. While the long term goal of strategy instruction is automatic application in appropriate contexts, this process takes time. When a procedure is newly learned, it requires more effort to carry out, it competes with old, familiar strategies and concepts, and it is not widely adaptable (Pressley, 1995). The dip in inferential comprehension performance on the withdrawal probes provides evidence that, although improvement from baseline was noted, additional clinician support and structured practice beyond six treatment sessions may be necessary for ultimate inferential comprehension success.

A second piece of evidence is found in the change that was seen in standardized test performance, with the percentile rank score on the GMRT Passage Comprehension subtest moving from the below average range to the average range. There was also a decrease in the number of both inferential and literal question errors made on this same instrument at post-test. Lastly, the pre/post-test receptive vocabulary control measure (PPVT-III score) did not show change from pre- to post-testing. A control measure such as this one provides some, although limited, reassurance that the changes seen in inferential comprehension performance on the GMRT were not a result of maturation, practice effects, or participation in the classroom curriculum.

In addition to the changes seen in test and probe scores, qualitative changes were observed by both the clinician/second author and Z's classroom teacher. For example, the second author noted that Z required less and less cueing and scaffolding to successfully answer the comprehension questions during the treatment activities. Additionally, his teacher commented that Z demonstrated "increased confidence during reading activities" and improved comprehension performance in the classroom after treatment began.

One last consideration with regard to the findings, especially from a public school therapy planning standpoint, is the change in inferential comprehension performance with respect to the treatment schedule. When considering how much therapy is necessary (i.e., treatment "dosage") for students with language/learning disabilities to be successful with a new concept or skill, this study provides some important insight and a direction for future research. Improvement was demonstrated after six sessions of treatment, indicating that learning can take place during this time frame. Worth noting too, though, is that more intervention time may be necessary to foster completely independent application of new information and strategies. In this era of treatment accountability, the issue of optimal treatment intensity is an important area of investigation.

There were several limitations to this study. First, the design would have been stronger with a multiple baseline across participants or behaviors, thereby providing better evidence to support treatment effects. A single participant limits generalizability and provides only a weak form of clinical evidence. Also, the school/student schedule prohibited a longer treatment period and efficient data-gathering, as we were only able to treat for 8 weeks and obtain one post-treatment withdrawal measure. There was also a risk for examiner bias as the second author was the only speech/language pathologist at the school and therefore completed all data collection. Additionally, it cannot be assumed that inferential comprehension ability would improve in children with other disabilities or that performance on the question-answering task in this study would generalize to other inferential tasks. Thus, this inferential comprehension intervention procedure should be investigated in a study with greater experimental control, a longer treatment period, and a variety of inferential comprehension tasks. Additionally, examining its use in an inclusive classroom setting would be a study with practical implications.

Lastly, as speech-language pathologists, we need to hold ourselves accountable for the success of our treatment programs (Damico, 1988; Meline & Paradiso, 2003; Zippoli & Kennedy, 2005). Clinical effects need to be demonstrated empirically, which can be a challenging process in school-based settings. As we seek to close the gap between the small evidence base of Level 1 and 2- studies in child language research (Cirrin & Gillam, 2008) and the actual research being conducted with primary and intermediate school-age children, we will need to explore how manageable but carefully designed individual treatment studies can serve as a realistic starting point. While speech/language pathologists in the schools are already gathering baseline data, implementing treatment as a result of the data, and measuring progress in consistent ways, they must now determine how to create additional experimental control and be ready to publish their findings. Given the logistics involved in providing this additional control, preliminary evidence demonstrating a treatment program's success can provide confidence that the intervention is worth the time and effort necessary for continued study and attainment of a higher level of evidence.

Language treatment studies are an increasingly important objective for clinicians in the public schools and, as this study shows, a doable endeavor in that setting. Data can be carefully collected and evaluated and intervention programs such as the Milosky and Ford (1996) inferential comprehension paradigm can, over time, become evidence-based options for school age children with language/learning disabilities.

References

Adams, C., Clarke, E., & Haynes, R. (2009). Inference and sentence comprehension in children with specific or pragmatic language impairments. *International Journal of Language and Communication Disorders, 44* (3), 301-381.

American Speech-Language-Hearing Association (2005a). Evidence-based practice in communication disorders: An introduction [Position Statement]. Retrieved from http://www.asha.org/members/deskref-journals/deskref/default

Bashir, A., & Singer, B.D. (2006). Assisting students in becoming self-regulated writers. In T. Ukrainetz (Ed.), *Contextualized language intervention: Scaffolding pre-k-12 literacy achievement* (pp. 565-598) Eau Claire, WI: Thinking Publications.

Beeson, P., & Robey, R. (2006). Evaluating single subject treatment research: Lessons learned from the Aphasia literature. *Neuropsychological Review, 16*, 161-169.

Brown, A. L., Palinscar, A. S., & Armbruster, B.B. (1984). Instructing comprehension-fostering activities in interactive learning situations. In H. Mandl, N.L., Stein, & T. Trabasso (Eds.), *Learning and comprehension of text.* (pp. 121-123). Hillsdale: Erlbaum.

Cain, K., Oakhill, J. V., & Elbro, C. (2003). The ability to learn new word meanings from context by school-age children with and without language comprehension difficulties. *Journal of Child Language, 30* (3). 681-694.

Casteel, M. (1993). Effects of inference necessity and reading goal on children's inferential generation. *Developmental Psychology, 29*, 2, 36-357.

Carr, E., Dewitz, P., & Patberg, J. (1989). Using cloze for inference training with expository text. *Reading Teacher, 42* (6) 380-385.

Carr, E., & Thompson, B. (1996). The effects of prior knowledge and schema activation strategies on the inferential reading comprehension of children with and without learning disabilities. *Learning Disability Quarterly, 19*, 48-61.

Cirrin, F., & Gillam, R. (2008). Language intervention practices for school-age children with spoken language disorders: A systematic review. *Language Speech and Hearing Services in Schools, 39*, 110-137.

Damico, J. (1988). The lack of efficacy in language therapy: A case study. *Language, Speech, and Hearing Services in Schools, 19*, 51-66.

Dodwell, K., & Bavin, E. L. (2008). Children with specific language impairment: An investigation of their narratives and memory. *International Journal of Language and Communication Disorders, 43* (2), 201-218.

Dunn, L., & Dunn, L. (1981). *Peabody Picture Vocabulary Test - Revised*. Circle Pines: American Guidance Service.

Garner, R. (1990). When children and adults do not use learning strategies: Toward a theory of settings. *Review of Educational Research, 60*, 517-529.

Gillam, S. (2007). Understanding and remediating text comprehension problems in school-age children. In A.G. Kamhi, J.J. Masterson & K. Apel (Eds.), *Clinical decision making* (pp. 267-283). Maryland: Paul H. Brookes Publishing Co.

Graham, S., & Harris, K. H. (2012). The role of strategies, knowledge, will and skills in a 30-year program of writing research (with homage to Hayes, Fayol, and Boscolo). In V.W. Berninger (Ed.), *Past, present, and future contributions of cognitive writing research to cognitive psychology* (pp. 177-196). New York: Psychology Press.

Guszak, F. J. (1967). Teacher questioning and reading. *Reading Teacher, 21*, 227-234.

Hansen, C. (1981). The effects of inference training and practice on young children's comprehension. *Reading Research Quarterly, 16*, 391-417.

Hansen, J., & Pearson, P. (1983). An instructional study improving the inferential comprehension of good and poor fourth-grade readers. *Journal of Educational Psychology, 75*, 821-829.

Holmes, B. (1984). The effect of prior knowledge on the question answering of good and poor readers. *Journal of Reading Behavior, 15*, 1-18.

Kromrey, J. D., & Foster-Johnson, L. (1996). Determining the efficacy of intervention: The use of effect sizes for data analysis in single subject research. *Journal of Experimental Education, 65* (1), 73-93.

Laing, S., & Kamhi, A. (2002). The use of think-aloud protocols to compare inferencing abilities in average and below-average readers. *Journal of Learning Disabilities, 35*, 436-447.

Maria, K. (1990). *Reading comprehension instruction: Issues & Strategies*. Maryland: York Press.

MacGinitie, W., & MacGinitie, R. (1989). *Gates MacGinitie Reading Tests, Third Edition*. Itasca, IL: Riverside Publishing.

McGee, A., & Johnson, H. (2003). The effect of inference training on skilled and less skilled comprehenders, Educational Psychology, 23, 1, 49-59.

Meline, T., & Paradiso, T. (2003). Evidence-based practice in schools: Evaluating research and reducing barriers. *Language, Speech, & Hearing Services in Schools, 34*(4), 273-283.

Milosky, L., & Ford, J. (1996). Comprehending what is not said: Critical inferencing skills in comprehension. Presentation at the American Speech and Hearing Association Convention. Seattle, WA.

Oakhill, J. (1984). Inferential and memory skills in children's comprehension of stories. *British Journal of Educational Psychology, 54*, 31-39.

Omanson, R. C., Warren, W. H., & Trabasso T. (1978). Goals, inferential comprehension, and recall of stories by children. *Discourse Process, 1*, 337-354.

Phillips, B., Ball, C., Sackett, D., Badenoch, D., Straus, S., Haynes, B., & Dawes, M. (2001, May). Oxford Centre for evidence-based medicine levels of evidence, Retrieved from http://www.cemb.net/levels_of_evidence.asp

Pressley, M. (1995). More about the development of self-regulation: Complex, long-term, and thoroughly social. *Educational Psychologist, 30* (4), 207-212.

Reder, L. M. (1980). The role of elaboration in the comprehension and retention of prose: A critical review. *Review of Educational Research, 50*, 5-53.

Robey, R., Schultz, M., Crawford, A., & Sinner, C. (1999). Single-subject clinical outcome research: Designs, data, effect sizes, and analyses. *Aphasiology, 13*, 445-473.

Sackett, D. L., Rosenberg, W. M., Gray, J. A., Haynes, R. B., & Richardson, W.S. (1996). *Evidence-based medicine: How to practice and teach EBM*. Edinburgh, Scotland: Churchill Livingstone.

Sadker, M., & Sadker, D. (1982). *Sex equity handbook for schools.* New York: Longman, Scannell-Miller, M. (1982). A comparison of the development and use of inference: A mnemonic strategy for the disabled and normal-achieving students. *Dissertation Abstracts International, 43*, 12.

Schunk, D.H., & Ertmer, P.A. (2000). Self-regulation and academic learning: Self-efficacy enhancing interventions. In M. Boekaergs, P.R. Pintrich, & M. Zeidner (Eds.), *Handbook of self-regulation* (pp. 631-649). San Diego: Academic Press.

Snyder, L. (1984). Developmental language disorders: Elementary school age. In A. Holland (ed.), *Language disorders in children: Recent advances* (pp. 129-158). San Diego: College Hill Press.

Steck-Vaugh Company (1999). *Improving Reading Comprehension 3*. Orlando: Houghton Mifflin Harcourt.

Tierney, R. J., Bridge, C., & Cera, M. J. (1978). The discourse processing operations of children. *Reading Research Quarterly, 4*, 541-569.

Troia, G. (2002). Teaching writing strategies to children with disabilities: Setting generalizations as the goal. *Exceptionality, 10*, 249-269.

Williams, J. P. (1993). Comprehension of students with and without learning disabilities: Identification of narrative themes and idiosyncratic text representations. *Journal of Educational Psychology, 85*, 631-641.

Wilson, M. M. (1979). The processing strategies of average and below average readers answering factual and inferential questions on three equivalent passages. *Journal of Reading Behavior, 11*, 235-245.

Wong, B. Y. L. (1988). Research and educational implications of some recent conceptualizations in learning disabilities. Learning *Disability Quarterly, 11* (3), 189-194.

Wong, B. Y. L. (1994). Instructional parameters promoting transfer of learned strategies in students with learning disabilities. *Learning Disability Quarterly, 17* (2), pp. 110-120.

Zabrusky, K. (1986). The role of factual coherence in discourse comprehension. *Discourse Processes, 9*, 197-220.

Zippoli, R., & Kennedy, M. (2005). Evidence-based practice among speech-language pathologists: Attitudes, utilization, and barriers. *American Journal of Speech-Language Pathology, 14*, 208-220.

Authors' Note

Correspondence concerning this article should be addressed to Laura B. Green, Department of Communication Sciences and Disorders, Texas Woman's University P. O. Box 425737 Denton, TX 76204 U.S.A. Email: LGreen@mail.twu.edu

Received date: June 19, 2012

Accepted date: April 16, 2013

Appendix A: Pre-test, Treatment and Post-test Schedule (January-April)

Administration of pre-test measures

January 28, February 15

Baseline data collection

February 1, 4 and 11

Lesson and Withdrawal measure dates

2/18 Lesson 1

2/29 Lesson 2

3/2 Withdrawal 1

3/10 Lesson 3

3/20 Lesson 4

3/27 Withdrawal 2 (administered prior to Lesson 5)

3/27 Lesson 5

3/31 Lesson 6

4/7 Withdrawal 3

Administration of post-test measures

April 14

Appendix B: Example Stories (Steck-Vaughn Company, 1999) and Comprehension Questions

A New Name

Little Deer was tired of his name. It was a name for a young boy. Now that Little Deer was ten summers old, he no longer thought he was a little boy. Little Deer thought he was old enough to be given a powerful man's name. Little Deer knew he could not just change his name, so he talked to the elders of the tribe.

The elders of the tribe said Little Deer could earn a man's name by doing a brave deed. Little Deer could not think of a brave deed to do. Then one day Little Deer saw a wild horse charge toward his little sister. Without thinking of his own safety, Little Deer ran toward the horse, shouting and waving his arms. Just before the horse reached her, it turned away. The people of the tribe were so grateful to Little Deer that they changed his name to Wild Horse.

1. What was Little Deer tired of? (Literal)
2. Why did Little Deer think his name was for a young boy? (Inferential)
3. How old was Little Deer? (Literal)
4. Who did Little Deer talk to about changing his name? (Literal)
5. How do you think the people of the tribe thought of the name Wild Horse? (Inferential)
6. What did Little Deer hope to do by running toward the horse waving his arms? (Inferential)
7. What did the elders say Little Deer needed to do in order to change his name? (Literal)
8. Who was the horse charging toward? (Literal)
9. Why do you think Little Horse's sister didn't run away from the horse? (Inferential)
10. Why couldn't Little Deer just change his name? (Inferential)

Aunt Kate's Cottage

Lana and her family were packing the car. Today was the first day of summer vacation. Every year, for as long as Lana could remember, her family had gone to the lake for the whole summer. Lana's Aunt Kate owned a cottage there. She always had invited Lana's family to stay with her for the summer.

Lana loved to go to visit her aunt's cottage. It was surrounded by fir trees that were full of birds and their nests. Lana and her brother would spend all day swimming and fishing. At night they would cook dinner over a fire and tell stories by the lake.

As Lana looked out of the car window, she was both happy and sad. She was looking forward to spending the summer at the lake, but this would be their last summer there. Aunt Kate had decided to sell the cottage. She told the family she would keep it until the end of the summer. Then she was putting the cottage up for sale. Lana would miss the lake and the cottage very much. She promised herself that this summer would be the best summer of all at the lake.

1. How long do Lana and her family typically stay at the lake during the summer? (Literal)
2. Why did Lana promise herself to make this summer the best? (Inferential)
3. Who owned the cottage? (Literal)
4. How did Lana get to her aunt's cottage? (Inferential)
5. Where did Lana's family cook dinner? (Literal)
6. Who was always invited to stay the summer at the lake? (Literal)
7. What kinds of things did Lana and her family pack in the car? (Inferential)
8. How did Lana's family feel about visiting the cottage? (Inferential)
9. Why did Aunt Kate keep the cottage until the end of the summer? (Inferential)
10. How did Lana feel as she looked out the car window? (Literal)

Book Review
Évaluation de Livre

Title: Cochlear implants and other implantable hearing devices

Publisher: Plural Publishing Inc.

Author: Michael J. Ruckenstein

Cost: $110.00

ISBN: 159756432X

Reviewer: François Bergeron, Laval University

Objectives or purpose

The book regroups 21 chapters written by a group of experts reviewing contemporary data on cochlear and other implantable devices.

Intended audience

The target audience is not clearly specified. Considering that the content covers broad topics ranging from the history of cochlear implants to rehabilitation, including surgery and programming, the book would be of interest to non-experts in this field. This is emphasized in the preface where the editor highlights the practical format of the chapters providing students with the necessary background to understand and contribute to the field.

General quality and organization

The textbook is of good quality, including hard cover, good quality paper, clear photos and readable figures. Globally, chapters are organized in a logical way, which is closely related to the sequence of interventions in the implantation process.

Evaluation of the product

As the book regroups contributions from different authors, the quality of the content fluctuates from one chapter to the other: some have a strong science basis and focus on more recent developments in the field, while others appear more superficial, some even presenting incomplete or biased information. Chapter 1, for example, misses important milestones in the history of cochlear implants. While the author introduces the Audiant and the Baha systems, Ponto and Sophono devices are ignored even though these systems were introduced well before 2012. The reader is also not informed that the Symphonic middle ear implant is now a Med-El device, or that the Otologic device is now available as a completely implantable system. Further, the author states that cochlear implants are mass-produced, which is not the case. Additionally, the author states that cochlear implants are designed to last for a lifetime, which, in 2012, is not accurate when considering the expected technological improvements in the lifetime of users. Then the historical review that follows completely ignores the work done in France by Chouard & colleagues and in Austria by Hochmair & Hochmair. The section on the advancement of speech processing strategies is rather light in view of the tremendous work conducted over the years on this issue. Finally the author neglect to specify that initial candidacy was reserved to adults with acquired deafness.

Chapter 2 appears awkwardly placed. A discussion on designing a cochlear implant program requires that one understand all aspects of the process of cochlear implantation, including the professionals involved, the specifics of the different devices and their maintenance, candidacy, protocols, etc. These issues are covered further in the textbook; consequently, this chapter would be expected to appear toward the end of the book. Pertinence of the long section on US insurance considerations, including the national Current Procedural Terminology codes, is questionable, specifically from an international point of view. In the evaluation protocol section, the prescribed testing condition is quite surprising: what does 60dB SPL (A) refer to? 60 dBSPL or 60 dBA? What is the rationale behind a signal-to-noise ratio of +8 dB? What is the rationale behind the prescribed follow-up schedule (2 weeks, 1, 3, 6, 9, 12 months)?

Chapters 3 and 4 on electrode design and signal processing strategies presents exhaustive and up to date discussions on these issues. The only drawback is the focus on USA available devices only, ignoring the contribution from other international manufacturers.

Chapter 5 presents the candidate selection process. Some discussion on the impact of neonatal auditory screening programs on paediatric candidacy would have been pertinent, bringing some recognition to the fact that family arrives in cochlear implant clinics with knowledge and acceptance of the diagnosis. The emphasis on testing

thresholds at 125 Hz is welcome considering the trend of implanting candidates with more and more residual hearing, as well as assessing both ears individually in order to assure that each ear will be optimally fitted. While quality of life appears as a pertinent outcome to consider, some thoughts on the sensitivity of this measure for cochlear implant candidates would have been a nice addition to this chapter. It is also of importance to understand that the role of the speech language pathologist cannot be limited to language assessment; he/she must also be involved in identifying any possible, specific speech-language impairments that can limit the use of the device. Again, cochlear implants, and bilateral implants, are only considered in a US context; an international perspective would have been appreciated. Finally, the proposition of assessing listening effort as a potential outcome of cochlear implants needs to be explored.

Imaging in cochlear implantation constitutes the main theme of chapter 6. As such, the first section dealing with hearing devices appears redundant with other chapters of the book; furthermore, the information is incomplete or misleading. Indeed, even if the textbook covers devices such as bone anchored hearing aids and middle ear implants, the author does not mention them as "hearing improvement devices". Also, the author's list of cochlear implant systems in use worldwide is completely out-dated. In the candidate section, the author refers to "... the rate of success of implantation..." without defining his own perception of this concept, which can be quite different from one person to another. When the reader finally enters the main topic of the chapter, he is faced with over-summarized information, often referring to technical details that are not of common knowledge. More detailed explanations would have been appropriate.

Surgical issues are discussed in chapters 7, 8, 9 and 10. As these issues are mainly examined from a US point of view, indications and techniques presented only reflect the American standard of practice.

A discussion on cost-effectiveness would have been a nice addition to chapter 11 on bilateral cochlear implantation. The reader is exposed to the positive outcomes of this approach; research results are not always clear. The concept of rehabilitation for bilateral implant users is original and interesting.

Basic information on programming for adults and children are presented in chapters 12 and 13. Again a US point of view is privileged.

Chapters 14, 15 and 16 present an overview of outcome measures in adults and children. While the more frequent tests are introduced, a critical discussion, emphasising forces and weaknesses of each of those tools, would have been pertinent. The section on electrophysiological measures of auditory development following cochlear implantation is innovative, interesting and pertinent. The same is true for the chapter on music perception with cochlear implants. This issue is expected to be more and more relevant as technology continues to move forward.

Chapter 17 addresses the critical issue of implant reliability. A large part of the chapter is devoted to the conception and evolution of implants; reliability is viewed on a theoretical basis but no actual metrics are given. Again, a US point of view is privileged which does not necessarily reflect the international reality. For example, if the first American child received a Nucleus multi-channel CI in 1989, this was realised earlier elsewhere. The international reader can also note some difficulty with foreign language use; "secousse dans la tate" should read "secousse dans la tête" (p.304).

The textbook concludes with a good overview of the other implantable solutions for hearing impaired individuals (chapters 18, 19, 20 and 21). While there is variation in completeness and redundancy in these chapters, the reader can derive a fair perspective on these technologies and their results.

Recommendations

Globally, the textbook can be considered among one of the many basic references in the field of cochlear and other implantable technology. It presents an overview of the field, sometimes with up-to-date and scientifically grounded information, sometimes with incomplete, out-dated or biased information. As such, it cannot be considered as a main and only reference for the non-expert in the field. Its utility for already well-established cochlear implant teams is, on the other hand, not clear; some chapters are very informative, some not.

Information for Contributors

The Canadian Journal of Speech-Language Pathology and Audiology (CJSLPA) welcomes submissions of scholarly manuscripts related to human communication and its disorders broadly defined. This includes submissions relating to normal and disordered processes of speech, language, and hearing. Manuscripts that have not been published previously are invited in English and French. Manuscripts may be tutorial, theoretical, integrative, practical, pedagogic, or empirical. All manuscripts will be evaluated on the basis of the timeliness, importance, and applicability of the submission to the interests of speech–language pathology and audiology as professions, and to communication sciences and disorders as a discipline. Consequently, all manuscripts are assessed in relation to the potential impact of the work on improving our understanding of human communication and its disorders. All categories of manuscripts submitted will undergo peer-review to determine the suitability of the submission for publication in CJSLPA. The Journal has established multiple categories of manuscript submission that will permit the broadest opportunity for dissemination of information related to human communication and its disorders. The categories for manuscript submission include:

Tutorials: Review articles, treatises, or position papers that address a specific topic within either a theoretical or clinical framework.

Articles: Traditional manuscripts addressing applied or basic experimental research on issues related to speech, language, and/or hearing with human participants or animals.

Clinical Reports: Reports of new clinical procedures, protocols, or methods with specific focus on direct application to identification, assessment and/or treatment concerns in speech, language, and/or hearing.

Brief Reports: Similar to research notes, brief communi-cations concerning preliminary findings, either clinical or experimental (applied or basic), that may lead to additional and more comprehensive study in the future. These reports are typically based on small "n" or pilot studies and must address disordered participant populations.

Research Notes: Brief communications that focus on experimental work conducted in laboratory settings. These reports will typically address methodological concerns and/or modifications of existing tools or instruments with either normal or disordered populations.

Field Reports: Reports that outline the provision of services that are conducted in unique, atypical, or nonstandard settings; manuscripts in this category may include screening, assessment, and/or treatment reports.

Letters to the Editor: A forum for presentation of scholarly/clinical differences of opinion concerning work previously published in the Journal. Letters to the Editor may influence our thinking about design considerations, methodological confounds, data analysis, and/or data interpretation, etc. As with other categories of submissions, this communication forum is contingent upon peer-review. However, in contrast to other categories of submission, rebuttal from the author(s) will be solicited upon acceptance of a letter to the editor.

Submission of Manuscripts

Contributors should use the electronic CJSLPA manuscript submission system at www.cjslpa.coverpage.ca to submit articles. If you are unable to use the electronic system, please send a file containing the manuscript, including all tables, figures or illustrations, and references in Word via e-mail to the editor at cjslpa.rcoa@caslpa.ca.

Along with copies of the manuscript, a cover letter indicating that the manuscript is being submitted for publication consideration should be included. The cover letter must explicitly state that the manuscript is original work, that it has not been published previously, and that it is not currently under review elsewhere. Manuscripts are received and peer-reviewed contingent upon this understanding.

The author(s) must also provide appropriate confirmation that work conducted with humans or animals has received ethical review and approval. Failure to provide information on ethical approval will delay the review process. Finally, the cover letter should also indicate the category of submission (i.e., tutorial, clinical report, etc.). If the editorial staff determines that the manuscript should be considered within another category, the contact author will be notified.

All submissions should conform to the publication guidelines of the Publication Manual of the American Psychological Association (APA), 6th Edition. A confirmation of receipt for all manuscripts will be provided to the contact author prior to distribution for peer review. CJSLPA seeks to conduct the review process and respond to authors regarding the outcome of the review within 90 days of receipt. If a manuscript is judged as suitable for publication in CJSLPA, authors will have 30 days to make necessary revisions prior to a secondary review.

The author is responsible for all statements made in his or her manuscript, including changes made by the editorial and/or production staff. Upon final acceptance of a manuscript and immediately prior to publication, the contact author will be permitted to review galley proofs and verify its content to the publication office within 72 hours of receipt of galley proofs.

Organization of the Manuscript

All copies should be typed, double-spaced, with a standard typeface (12 point, non-compressed font) on 8 ½ x 11 paper size. All margins should be at least one (1) inch. An electronic copy of the manuscript should be submitted directly to the editor. Author identification for the review process is optional; if blind-review is desired, the documents should be prepared accordingly (cover page and acknowledgments blinded). Responsibility for removing all potential identifying information rests solely with the author(s). All submissions should conform to the publication guidelines of the most current edition of the Publication Manual of the American Psychological Association (APA. The APA manual is available from most university and commercial bookstores. Generally, the following sections should be submitted in the order specified.

Title Page: This page should include the full title of the manuscript, the full names of the author(s) with academic degrees, each author's affiliation, and a complete mailing address for the contact author. An electronic mail address also is recommended.

Abstract: On a separate sheet of paper, a brief yet informative abstract that does not exceed one page is required. The abstract should include the purpose of the work along with pertinent information relative to the specific manuscript category for which it was submitted.

Key Words: Following the abstract and on the same page, the author(s) should supply a list of key words for indexing purposes.

Tables: Each table included in the manuscript must typedwritten double-spaced and placed at the end of the document. Tables should be numbered consecutively beginning with Table 1. Each table must have a descriptive caption. Tables should serve to expand the information provided in the text of the manuscript, not to duplicate information.

Illustrations: All illustrations to be included as part of the manuscript must also be submitted in their original file format separate from the manuscript. High resolution (at least 300 dpi) files in any of the following formats must be submitted for each graphic and image: JPEG, TIFF, AI, PSD, GIF, EPS or PDF. For other types of computerized illustrations, it is recommended that CJSLPA production staff be consulted prior to preparation and submission of the manuscript and associated figures/illustrations.

Legends for Illustrations: Legends for all figures and illustrations should be typewritten (double-spaced) on a separate page with numbers corresponding to the order in which figures/illustrations appear in the manuscript.

Page Numbering and Running Head: The text of the manuscript should be prepared with each page numbered, including tables, figures/illustrations, references, and appendices. A short (30 characters or less) descriptive running title should appear at the top right hand margin of each page of the manuscript.

Acknowledgments: Acknowledgments should be typewritten (double-spaced) on a separate page. Appropriate acknowledgment for any type of sponsorship, donations, grants, technical assistance, and to professional colleagues who contributed to the work, but are not listed as authors, should be noted.

References: References are to be listed consecutively in alphabetical order, then chronologically for each author. Authors should consult the most current edition of the APA publication manual for methods of citing varied sources of information. Journal names and appropriate volume number should be spelled out and italicized. All literature, tests and assessment tools, and standards (ANSI and ISO) must be listed in the references. All references should be double-spaced.

Potential Conflicts of Interest and Dual Commitment

As part of the submission process, the author(s) must explicitly identify if any potential conflict of interest or dual commitment exists relative to the manuscript and its author(s). Such disclosure is requested so as to inform CJSLPA that the author or authors have the potential to benefit from publication of the manuscript. Such benefits may be either direct or indirect and may involve financial and/or other nonfinancial benefit(s) to the author(s). Disclosure of potential conflicts of interest or dual commitment may be provided to editorial consultants if it is believed that such a conflict of interest or dual commitment may have had the potential to influence the information provided in the submission or compromise the design, conduct, data collection or analysis, and/or interpretation of the data obtained and reported in the manuscript submitted for review. If the manuscript is accepted for publication, editorial acknowledgement of such potential conflict of interest or dual commitment may occur within the publication.

Participants in Research Humans and Animals

Each manuscript submitted to CJSLPA for peer-review that is based on work conducted with humans or animals must acknowledge appropriate ethical approval. In instances where humans or animals have been used for research, a statement indicating that the research was approved by an institutional review board or other appropriate ethical evaluation body or agency must clearly appear along with the name and affiliation of the research ethics and the ethical approval number. The review process will not begin until this information is formally provided to the Editor.

Similar to research involving human participants, CJSLPA requires that work conducted with animals state that such work has met with ethical evaluation and approval. This includes identification of the name and affiliation of the research ethics evaluation body or agency and the ethical approval number. A statement that all research animals were used and cared for in an established and ethically approved manner is also required. The review process will not begin until this information is formally provided to the Editor.

Renseignements à l'intention des collaborateurs

La Revue canadienne d'orthophonie et d'audiologie (RCOA) est heureuse de se voir soumettre des manuscrits de recherche portant sur la communication humaine et sur les troubles qui s'y rapportent, dans leur sens large. Cela comprend les manuscrits portant sur les processus normaux et désordonnés de la parole, du langage et de l'audition. Nous recherchons des manuscrits qui n'ont jamais été publiés, en français ou en anglais. Les manuscrits peuvent être tutoriels, théoriques, synthétiques, pratiques, pédagogiques ou empiriques. Tous les manuscrits seront évalués en fonction de leur signification, de leur opportunité et de leur applicabilité aux intérêts de l'orthophonie et de l'audiologie comme professions, et aux sciences et aux troubles de la communication en tant que disciplines. Par conséquent, tous les manuscrits sont évalués en fonction de leur incidence possible sur l'amélioration de notre compréhension de la communication humaine et des troubles qui s'y rapportent. Peu importe la catégorie, tous les manuscrits présentés seront soumis à une révision par des collègues afin de déterminer s'ils peuvent être publiés dans la RCOA. La Revue a établi plusieurs catégories de manuscrits afin de permettre la meilleure diffusion possible de l'information portant sur la communication humaine et les troubles s'y rapportant. Les catégories de manuscrits comprennent :

Tutoriels : Rapports de synthèse, traités ou exposés de position portant sur un sujet particulier dans un cadre théorique ou clinique.

Articles : Manuscrits conventionnels traitant de recherche appliquée ou expérimentale de base sur les questions se rapportant à la parole, au langage ou à l'audition et faisant intervenir des participants humains ou animaux.

Comptes rendus cliniques : Comptes rendus de nouvelles procédures ou méthodes ou de nouveaux protocoles cliniques portant particulièrement sur une application directe par rapport aux questions d'identification, d'évaluation et de traitement relativement à la parole, au langage et à l'audition.

Comptes rendus sommaires : Semblables aux notes de recherche, brèves communications portant sur des conclusions préliminaires, soit cliniques soit expérimentales (appliquées ou fondamentales), pouvant mener à une étude plus poussée dans l'avenir. Ces comptes rendus se fondent typiquement sur des études à petit « n » ou pilotes et doivent traiter de populations désordonnées.

Notes de recherche : Brèves communications traitant spécifiquement de travaux expérimentaux menés en laboratoire. Ces comptes rendus portent typiquement sur des questions de méthodologie ou des modifications apportées à des outils existants utilisés auprès de populations normales ou désordonnées.

Comptes rendus d'expérience : Comptes rendus décrivant sommairement la prestation de services offerts en situations uniques, atypiques ou particulières; les manuscrits de cette catégorie peuvent comprendre des comptes rendus de dépistage, d'évaluation ou de traitement.

Courrier des lecteurs : Forum de présentation de divergences de vues scientifiques ou cliniques concernant des ouvrages déjà publiés dans la Revue. Le courrier des lecteurs peut avoir un effet sur notre façon de penser par rapport aux facteurs de conception, aux confusions méthodologiques, à l'analyse ou l'interprétation des données, etc. Comme c'est le cas pour d'autres catégories de présentation, ce forum de communi-cation est soumis à une révision par des collègues. Cependant, contrairement aux autres catégories, on recherchera la réaction des auteurs sur acceptation d'une lettre.

Présentation de manuscrits

Pour soumettre un article, les auteurs doivent utiliser le système de soumission électronique de l'ACOA à l'adresse www.cjslpa.coverpage.ca. Si vous ne pouvez pas utiliser le système électronique, veuillez envoyer par courriel un fichier Word contenant le manuscrit, y compris tous les tableaux, les figures ou illustrations et la bibliographie. Adressez le courriel au rédacteur en chef à l'adresse cjslpa.rcoa@caslpa.ca.

On doit joindre aux exemplaires du manuscrit une lettre d'envoi qui indiquera que le manuscrit est présenté en vue de sa publication. La lettre d'envoi doit préciser que le manuscrit est une œuvre originale, qu'il n'a pas déjà été publié et qu'il ne fait pas actuellement l'objet d'un autre examen en vue d'être publié. Les manuscrits sont reçus et examinés sur acceptation de ces conditions. L'auteur (les auteurs) doit (doivent) aussi fournir une attestation en bonne et due forme que toute recherche impliquant des êtres humains ou des animaux a fait l'objet de l'agrément d'un comité de révision déontologique. L'absence d'un tel agrément retardera le processus de révision. Enfin, la lettre d'envoi doit également préciser la catégorie de la présentation (i.e. tutoriel, rapport clinique, etc.). Si l'équipe d'examen juge que le manuscrit devrait passer sous une autre catégorie, l'auteur-contact en sera avisé.

Toutes les présentations doivent se conformer aux lignes de conduite présentées dans le publication Manual of the American Psychological Association (APA), 6e Édition. Un accusé de réception de chaque manuscrit sera envoyé à l'auteur-contact avant la distribution des exemplaires en vue de la révision. La RCOA cherche à effectuer cette révision et à informer les auteurs des résultats de cette révision dans les 90 jours de la réception. Lorsqu'on juge que le manuscrit convient à la RCOA, on donnera 30 jours aux auteurs pour effectuer les changements nécessaires avant l'examen secondaire.

L'auteur est responsable de toutes les affirmations formulées dans son manuscrit, y compris toutes les modifications effectuées par les rédacteurs et réviseurs. Sur acceptation définitive du manuscrit et immédiatement avant sa publication, on donnera l'occasion à l'auteur-contact de revoir les épreuves et il devra signifier la vérification du contenu dans les 72 heures suivant réception de ces épreuves.

Organisation du manuscrit

Tous les textes doivent être écrits à double interligne, en caractère standard (police de caractères 12 points, non comprimée) et sur papier 8 ½" X 11" de qualité. Toutes les marges doivent être d'au moins un (1) pouce. Un fichier électonique du manuscrit doit être présenté directement au rédacteur en chef. L'identification de l'auteur est facultative pour le processus d'examen : si l'auteur souhaite ne pas être identifié à ce stade, il devra préparer un fichier électronique dont la page couverture et les remerciements seront voilés. Seuls les auteurs sont responsables de retirer toute information identificatrice éventuelle. Tous les manuscrits doivent être rédigés en conformité aux lignes de conduite les plus récentes de l'APA. Ce manuel est disponible dans la plupart des librairies universitaires et commerciaux. En général, les sections qui suivent doivent être présentées dans l'ordre chronologique précisé.

Page titre : Cette page doit contenir le titre complet du manuscrit, les noms complets des auteurs, y compris les diplômes et affiliations, l'adresse complète de l'auteur-contact et l'adresse de courriel de l'auteur contact.

Abrégé : Sur une page distincte, produire un abrégé bref mais informateur ne dépassant pas une page. L'abrégé doit indiquer l'objet du travail ainsi que toute information pertinente portant sur la catégorie du manuscrit.

Mots clés : Immédiatement suivant l'abrégé et sur la même page, les auteurs doivent présenter une liste de mots clés aux fins de constitution d'un index.

Tableaux : Tous les tableaux compris dans un même manuscrit doivent être écrits à double interligne sur une page distincte. Les tableaux doivent être numérotés consécutivement, en commençant par le Tableau 1. Chaque tableau doit être accompagné d'une légende et doit servir à compléter les renseignements fournis dans le texte du manuscrit plutôt qu'à reprendre l'information contenue dans le texte ou dans les tableaux.

Illustrations : Toutes les illustrations faisant partie du manuscrit doivent être annexer avec chaque exemplaire du manuscrit. Chaque manuscrit doit être accompagné d'un fichier électronique pour chaque image et graphique en format JPEG, TIFF, AI, PSD, GIF, EPS ou PDF, compression minimale 300 ppp. Pour les autres types d'illustrations informatisées, il est recommandé de consulter le personnel de production de la RCOA avant la préparation et la présentation du manuscrit et des figures et illustrations s'y rattachant.

Légendes des illustrations : Les légendes accompagnant chaque figure et illustration doivent être écrits à double interligne sur une page distincte et identifiées à l'aide d'un numéro qui correspond à la séquence de parution des figures et illustrations dans le manuscrit.

Numérotation des pages et titre courant : Chaque page du manuscrit doit être numérotée, y compris les tableaux, figures, illustrations, références et, le cas échéant, les annexes. Un bref (30 caractères ou moins) titre courant descriptif doit apparaître dans la marge supérieure droite de chaque page du manuscrit.

Remerciements : Les remerciements doivent être écrits à double interligne sur une page distincte. L'auteur doit reconnaître toute forme de parrainage, don, bourse ou d'aide technique, ainsi que tout collègue professionnel qui ont contribué à l'ouvrage mais qui n'est pas cité à titre d'auteur.

Références : Les références sont énumérées les unes après les autres, en ordre alphabétique, suivi de l'ordre chronologique sous le nom de chaque auteur. Les auteurs doivent consulter le manuel de l'APA le plus récent pour obtenir la façon exacte de rédiger une citation. Les noms de revues scientifiques et autres doivent être rédigés au long et imprimés en italiques. Tous les ouvrages, outils d'essais et d'évaluation ainsi que les normes (ANSI et ISO) doivent figurer dans la liste de références. Les références doivent être écrits à double interligne.

Conflits d'intérêts possibles et engagement double

Dans le processus de présentation, les auteurs doivent déclarer clairement l'existence de tout conflit d'intérêts possibles ou engagement double relativement au manuscrit et de ses auteurs. Cette déclaration est nécessaire afin d'informer la RCOA que l'auteur ou les auteurs peuvent tirer avantage de la publication du manuscrit. Ces avantages pour les auteurs, directs ou indirects, peuvent être de nature financière ou non financière. La déclaration de conflit d'intérêts possibles ou d'engagement double peut être transmise à des conseillers en matière de publication lorsqu'on estime qu'un tel conflit d'intérêts ou engagement double aurait pu influencer l'information fournie dans la présentation ou compromettre la conception, la conduite, la collecte ou l'analyse des données, ou l'interprétation des données recueillies et présentées dans le manuscrit soumis à l'examen. Si le manuscrit est accepté en vue de sa publication, la rédaction se réserve le droit de reconnaître l'existence possible d'un tel conflit d'intérêts ou engagement double.

Participants à la recherche – êtres humains et animaux

Chaque manuscrit présenté à la RCOA en vue d'un examen par des pairs et qui se fonde sur une recherche effectuée avec la participation d'être humains ou d'animaux doit faire état d'un agrément déontologique approprié. Dans les cas où des êtres humains ou des animaux ont servi à des fins de recherche, on doit joindre une attestation indiquant que la recherche a été approuvée par un comité d'examen reconnu ou par tout autre organisme d'évaluation déontologique, comportant le nom et l'affiliation de l'éthique de recherche ainsi que le numéro de l'approbation. Le processus d'examen ne sera pas amorcé avant que cette information ne soit formellement fournie au rédacteur en chef.

Tout comme pour la recherche effectuée avec la participation d'êtres humains, la RCOA exige que toute recherche effectuée avec des animaux soit accompagnée d'une attestation à l'effet que cette recherche a été évaluée et approuvée par les autorités déontologiques compétentes. Cela comporte le nom et l'affiliation de l'organisme d'évaluation de l'éthique en recherche ainsi que le numéro de l'approbation correspondante. On exige également une attestation à l'effet que tous les animaux de recherche ont été utilisés et soignés d'une manière reconnue et éthique. Le processus d'examen ne sera pas amorcé avant que cette information ne soit formellement fournie au rédacteur en chef.

CASLPA | ACOA