Evidence-Based Practice Research: Educational and Therapeutic Interventions for Persons with Fragile X Syndrome

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Evidence-Based Practice and Fragile X Intervention

- Where and Why We Began This Project:

  Our purpose is to seek out and establish a data-base of effectiveness research in the areas of early intervention, speech-language intervention, occupational therapy, academic intervention, behavioral approaches, and so on. It is not to review medical/pharmacological interventions.

  Last year, Dr. Harris-Schmidt heard Dr. Patricia Prelock, of the University of Vermont, speak about intervention for children with autism. She discussed the National Autism Center's National Standards Report Findings and Conclusions.

Evidence-Based Practice and Fragile X Intervention

- The NAC's purpose was to identify the research support available for a variety of interventions currently promoted for children with autism.
- They reviewed thousands of articles, devised a rating scale for their scientific merit, and ultimately classified the methods as: Established, Emerging, Unestablished, Ineffective/Harmful.
- The NAC has shared with us their findings and their coding manual.

Evidence-Based Practice and Fragile X Intervention

- So…we began a search for information about interventions for children and adults with fragile X syndrome, a search for research-based evidence about “what works” for those with fragile X.

- This search was based on the principles of **Evidence-Based Practice.**

Evidence-Based Practice

- Definition:
- "The integration of best research evidence with clinical expertise and patient values" (Sackett, Strauss, and Richardson, 2000).
- Evidence-based practice involves a five-step process (ASC-AE):
  - Ask a question that is relevant for meeting a particular student, client, or group's needs
  - Search for available evidence
  - Critique the quality of the evidence
  - Apply the evidence to one's own practice
  - Evaluate effectiveness in terms of outcomes for a particular student, client or group (ASHA, 2005).

Steps in Evidence-Based Practice

- The first step is to ask a good questions, thinking about PICO (Nelson, 2010):
  - **P**erson, population, problem, and perspective
  - **I**ntervention: What procedures are most likely to achieve the outcome/goal for this particular person?
  - **C**omparison/contrast: Alternative interventions
  - **O**utcomes: What functional outcomes are observable and measurable and can provide evidence for how the intervention works?

Steps in Evidence-Based Practice

- Within the field of fragile X, there are many questions to be asked. They might be concerning best practice in **intervention** for behaviors, language development, social interaction, reading instruction, learning styles, and on and on.
- After formulating relevant questions, our next step is to search for evidence about the particular area about which we are interested and then to critique that evidence.

Search for the Evidence about Treatment

- For our search for evidence, we did the following:
- We used appropriate search engines, databases, and online resources. These included:
  - Worldcat (www.wordcat.org, which includes MedLine and other databases)
  - Cochrane Collection (www.cochrane.org/reviews)
  - What Works Clearinghouse (www.whatworks.ed.gov)
  - American Speech Language Hearing Association (www.asha.org)

The search included mainly peer-reviewed journals, not books or other publications.

Search for the Evidence about Treatment

- Within these databases, we put in terms that we hoped would lead us to articles about treatment:
- “fragile x” therapy’
- “fragile x” education’
- “fragile x” early intervention’
- “fragile x” vocational’
- “fragile x” parents’
- “fragile x” language’

Search for the Evidence about Treatment

- We then went back and conducted searches using various researchers' and authors' names from books and authors of articles in *The National Fragile X Foundation Quarterly*.

Search for the Evidence about Treatment

- We found many articles about fragile X, but most were about:
  - Characteristics
  - Development
  - Diagnosis
  - Assessment of various areas
  - Deficit areas
  - Lifespan changes

  - Most were **not** about the **effects of various types of intervention.**

Search for the Evidence about Treatment

- With the articles that we did find, we organized them by topic area and type of study.
- This list is FAR FROM complete—we are asking for your help in compiling intervention studies.

Critique of the Evidence

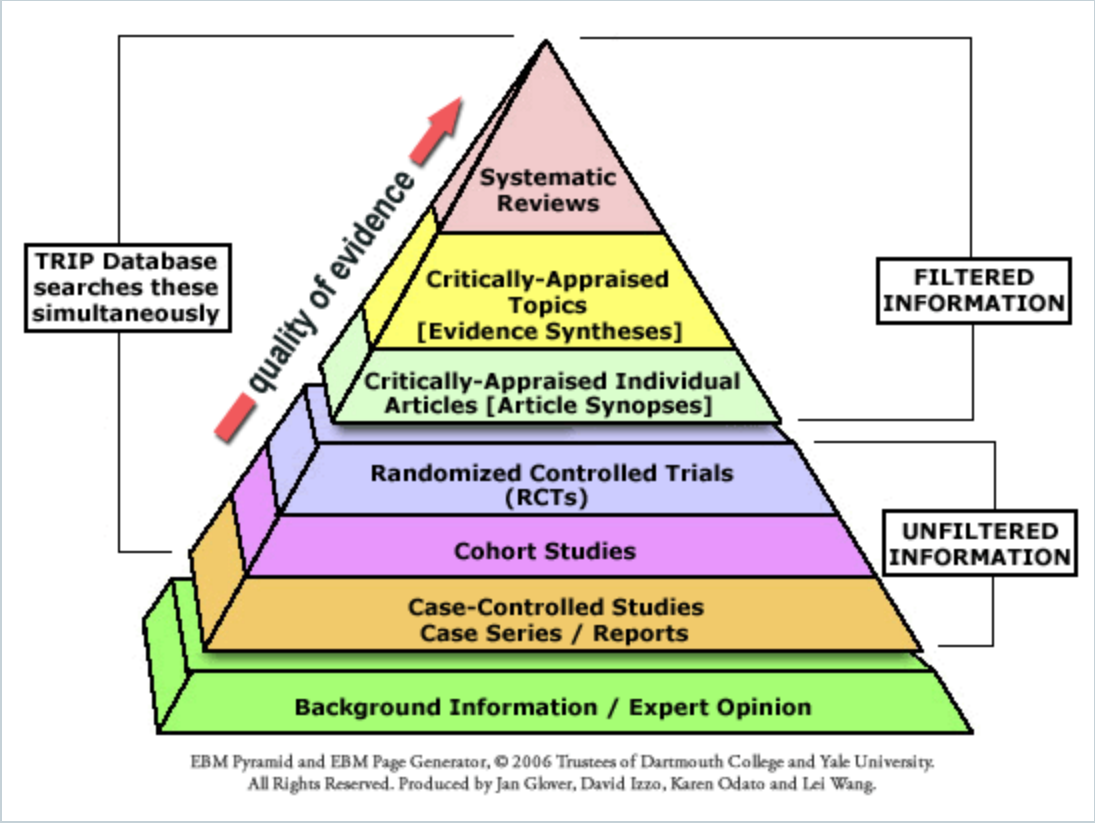
- We conducted a preliminary ranking of the research , using the pyramid levels.
- There are several examples of pyramids on the following slides, most of which originated from the field of medicine.
- Within the pyramids, highest rankings are given to meta-analyses that compare several randomly controlled trials.
- The next highest ranking is for randomly controlled trials (RCTs).

Evidence-Based Practice Pyramid

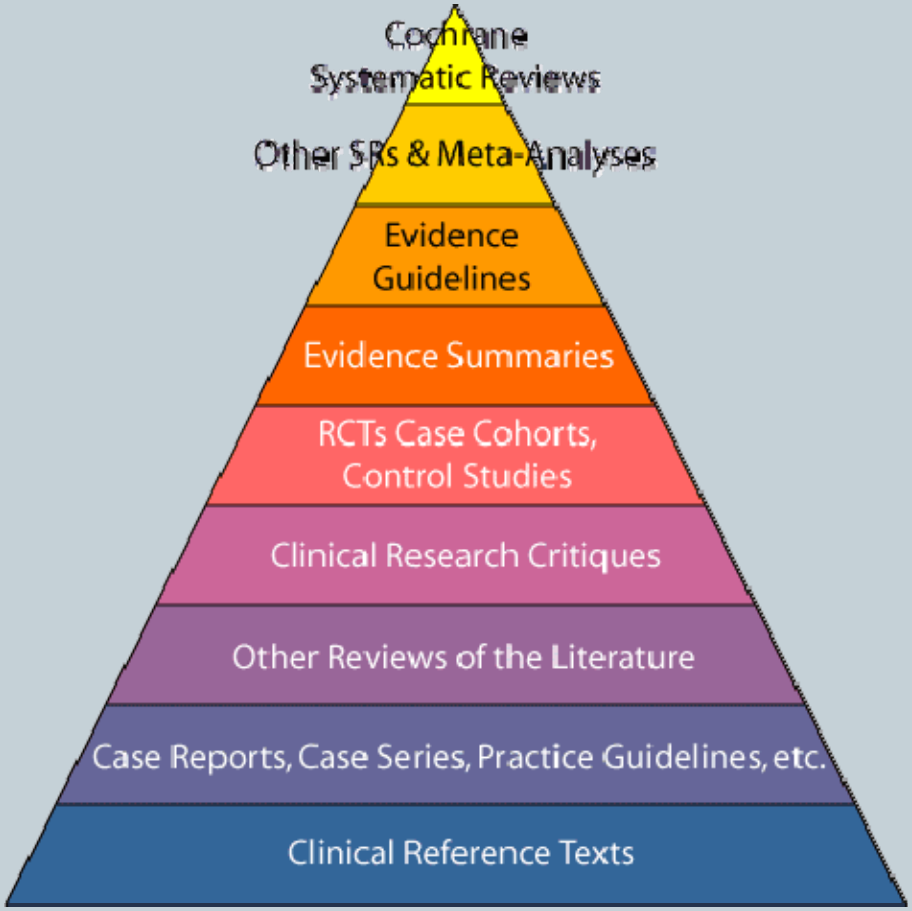
- http://ebp.lib.uic.edu/nursing

Evidence-Based Practice Pyramid

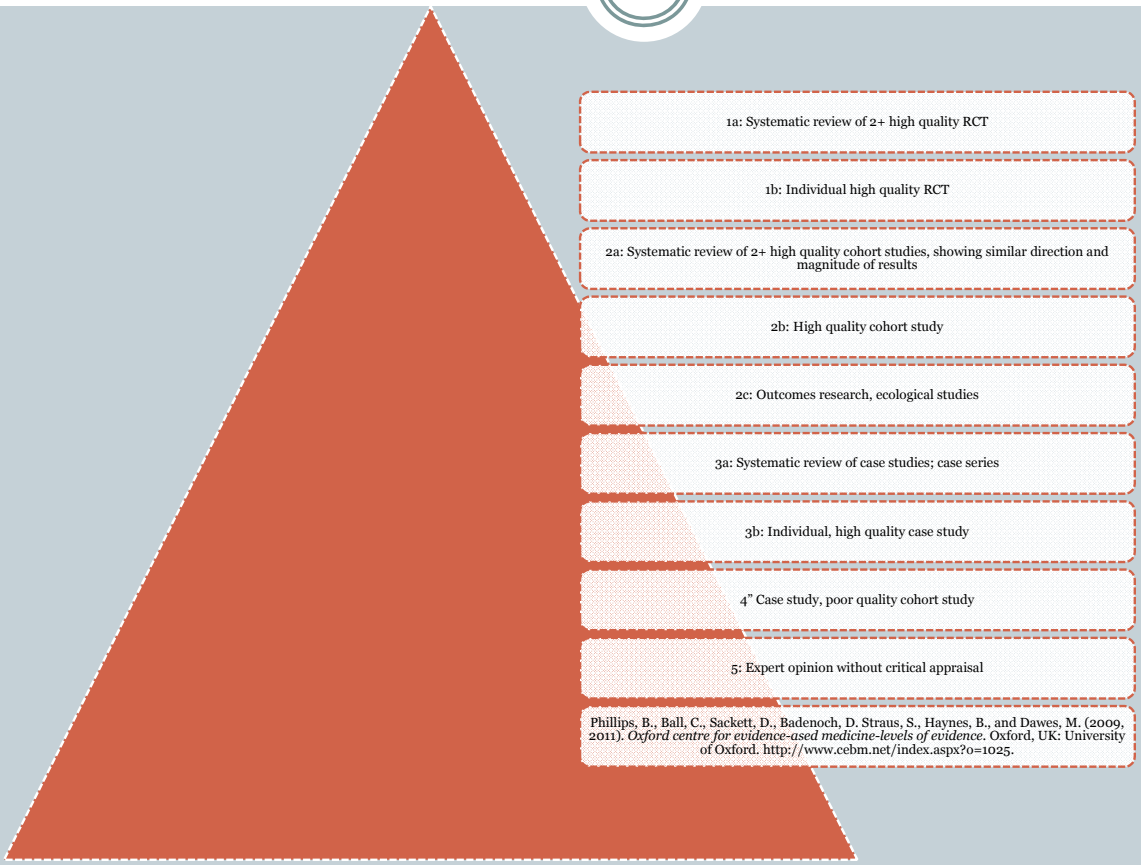
Hierarchy of Evidence-Based Medicine



Evidence-Based Practice Pyramid



Oxford Pyramid



1a: Systematic review of 2+ high quality RCT

1b: Individual high quality RCT

2a: Systematic review of 2+ high quality cohort studies, showing similar direction and magnitude of results

2b: High quality cohort study

2c: Outcomes research, ecological studies

3a: Systematic review of case studies; case series

3b: Individual, high quality case study

4" Case study, poor quality cohort study

5: Expert opinion without critical appraisal

Phillips, B., Ball, C., Sackett, D., Badenoch, D. Straus, S., Haynes, B., and Dawes, M. (2009, 2011). *Oxford centre for evidence-ased medicine-levels of evidence*. Oxford, UK: University of Oxford. http://www.cebm.net/index.aspx?o=1025.

Critique of the Evidence

- The problem in fragile X research is that randomly controlled trials are expensive and require experimental and control groups (one receiving the intervention being tested and the other either not receiving it or receiving another treatment).
- Therefore, we do not have many RCTs in our data base.
- We may need to move down the pyramid to look for studies that are non-experimental case studies and other small quasi-experimental studies.

Critique of the Evidence

- There is a group of consensus documents from our Fragile X Clinical and Research Consortium on five topics related to our searches:
  - Behavior Problems in Fragile X Syndrome
  - Hyperarousal in Fragile X Syndrome
  - Sleep in Children with Fragile X Syndrome
  - Toileting Issues in Fragile X Syndrome
  - Use of Complementary and Alternative Therapies in the Treatment of Fragile X Syndrome

Critique of the Evidence

- Each of the Consensus Documents lists an author and references, and each states that it was reviewed by members of the Fragile X Clinical and Research Consortium. These documents were included in the table (separate document) for review.
- The table (separate document) given shows the articles we have searched out or received from others so far.

Future Steps

- We did not complete the final two steps of applying the evidence and evaluating outcomes, as we leave those to the teachers, clinicians, clients, and families who are involved in education and therapy.
- We ask that special educators and therapists use evidence to design their teaching and therapy and conduct systematic, on-going assessments of students'/clients' progress (Nippold, 2011).

Where Do We Go From Here?

- Where are we in regard to fragile X and best evidence
  - Relatively young field
  - Focus has been on:
    - Identification
    - Causes
    - Characteristics
    - Developmental trajectories
    - Medical/drug treatments

Where Do We Go From Here?

- We invite researchers and clinicians to add to our list of research articles.
- We invite researchers and clinicians to rank and critique articles and to give us feedback about our rankings.
- We may ask the National Fragile X Foundation to find a place on the website for what we hope will be a growing body of educational/therapy intervention studies.

Where Do We Go From Here?

- Clearly, there is a need for research at every level about educational and therapeutic interventions for children and adults with fragile X syndrome.
- What are the questions that need answers?
- We need to have more published research in peer-reviewed journals both to help special educators and clinicians who see persons with fragile X, but also to "spread the word" about fragile X.
- How can we encourage clinicians to publish case studies, small group work etc.?
- Are there ways by which we can foster research across geographic lines, encouraging educators and clinicians to contribute pieces to collaborative projects?

Where Do We Go From Here?

- How do we help practitioners collaborate with researchers in order to generate questions and foster research? (Nelson, 2010)
- How do we encourage researchers to relate their questions to clinical concerns? (Apel, 2001)

How Evidence Based Practice Can Be Applied To Real Life Scenarios

- Requests for specific methodology in IEP meetings
- Legal support for specific behavioral intervention, placement and/or strategies for due process hearing
- Rationale for funding of training in specific strategies
- Rationale for Independent Education Evaluation (IEE)
- Support for private therapies and placements
- Overall outcome for students will be improved. IDEA requires EBP as RtI is implemented

Symons, Clark & Roberts 2001

- Classroom engagement of elementary school children with FXS is strongly related to the environmental and instructional quality of the teachers and classroom
- The ways the teachers structured and arranged the classroom environment was much more important to student engagement than specific aspects of the child's FX status, medication use or dual diagnosis

Symons, Clark & Roberts 2001

- Videos will be embedded here to illustrate how different the level of engagement is when the environment and instructional model is consistent with evidence based practice

Where Do We Go From Here?

- Open for Discussion!

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