

DECISION TREE MEETS OCKHAM'S RAZOR

Problem Solving In The Differential Diagnosis Of Acquired Epileptiform Aphasia (AEA)

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Tweaking the nosology: What acquired epileptiform aphasia (AEA) refers to

- In 1957, William Landau and Frank Kleffner first described acquired aphasia with convulsive disorder in children.
- In 1992, Landau declared the label Landau-Kleffner syndrome (LKS) to be an “eponymic badge of ignorance” (p. 353).
- AEA since then has gained currency as the identifier for language regression secondary to seizure activity, a characteristic of LKS, disintegrative epileptiform regression, and autistic epileptiform regression (Tuchman, 1997; Shinnar et al., 2002; Stefanatos, Kinsbourne & Wasserstein, 2002).

The underdeveloped clinical picture: What Landau (1992) was talking about

- No typical pathogenesis has been established for AEA (Singh, Kalita & Misra, 2002; Kolski & Otsubo, 2003).
- Seizure foci vary (Mizuno-Matsumoto et al., 2001; Robinson et al., 2001).
- Associated seizure episode types vary (Hahn et al., 2001; Shinnar et al., 2002).
- Nocturnal electrophysiological disturbances consistently reported in cases of AEA (Stefanatos, Kinsbourne & Wasserstein, 2002; Kolski & Otsubo, 2003) also are noted in cases of non-aphasic epilepsy (Mendez & Radtke, 2001; Dinner, 2002) and non-convulsive developmental dysphasia (Echenne et al., 1992; Nasr et al., 2001).
- Accounts of language deficits in AEA variously emphasize auditory processing (Plaza et al., 2001), phonology (Majerus et al., 2003), and pragmatics (Taylor & Nance, 2000).

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Additional complexity

- The ultimate impact of AEA upon different linguistic modalities (Glos, Jariabkova & Szabova, 2001; Perez et al., 2001; Sieratzki et al., 2001) and developmental course and outcome (Camfield & Camfield, 2002; Praline et al., 2003) is in dispute.
- Language deficits initially may co-occur and be confounded with (or obscured by)
 - verbal dyspraxia and stuttering (Taylor & Kamhi, 1995; Tutuncuoglu, Serdaroglu & Kadioglu, 2002)
 - psychoemotional disturbances (Devinsky et al., 1989; Alper et al., 1997; Kanemoto, 1997)
 - attention deficits, with or without hyperactivity (Taylor & Kamhi, 1995; Laporte et al., 2002; Richer, Shevell & Rosenblatt, 2002).
- AEA and autistic spectrum disorders (ASD) co-occur with high frequency (Kallen et al., 2001; Stefanatos, Kinsbourne & Wasserstein, 2002; Tuchman & Rapin, 2002).

The consequent problem

AEA continues to be misdiagnosed as everything from sensorineural deafness (Landau & Kleffner, 1957; Nasr et al., 2001) to social deviance (Taylor & Nance, 2000), making inappropriate, and possibly harmful, interventions commonplace. Neuroimaging procedures (e.g.: MRI, nocturnal EEG) ultimately are necessary for differential diagnosis, but can be costly in both time and money. A need for more systematic, reliable and valid screening procedures is therefore obvious.

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Decision-tree induction: Can it facilitate screening of potential AEA cases?

- **Decision- tree induction has facilitated assessment and intervention in cases of cancer (Decaestecker et al., 1997) and pneumonia (Eardley et al., n.d.).**
- **The sheer number of competing hypothetical diagnoses to be ruled out, however, complicates the use of decision-tree algorithms in identifying AEA.**

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The need to simplify: Where to apply Ockham's razor?

- **However desirable conceptual simplicity may seem, arbitrarily and artificially reducing complexity in data interpretation tends to guarantee systematic error in problem solving.**
- **One point at which simplicity can and should be promoted is that of initial interaction with parents (or other caregivers) of the child to be evaluated**
 - **who tend to experience emotional distress upon initially consulting a clinician over inexplicable symptoms observed in a child**
 - **whose disclosure of information tends to become inhibited when such distress is compounded by perplexity over clinical jargon (Taylor, 1995)**
 - **who tend to distrust and resent clinicians that seem to rush to diagnostic judgment.**

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A Clinical Imperative

Use of layperson language to optimize information sharing between clinician and client family becomes imperative if the clinician is to avoid many fruitless paths of diagnostic inquiry that lay open.

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Taylor and Kamhi (1995)

In a quantitative literature review encompassing 76 AEA cases, the following characteristics were identified as clinically significant in 50 percent or more:

- **receptive deficits**
- **expressive deficits**
- **pragmatic deficits**
- **attention deficits (confounded to an undetermined extent with absence seizure episodes)**
- **social adjustment difficulties**
- **emotional lability**
- **disruptive behaviors**
- **sleep disturbances**

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Taylor (1995); Taylor and Nance (2000)

- **In an ethnographic study, Taylor recorded lay terminology used for each characteristic noted by Taylor and Kamhi.**
- **Supplementing case retrospectives with parent/caregiver interviews, Taylor and Nance recorded lay terminology used for hyperlexic behavior (Benson, 1991) and altered speech sound response (Paetau, 1994) that may co-occur with AEA in some cases.**

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The LAIP (2004)

Taylor integrated the lay language from the 1995 and 2000 studies into 12 problem descriptors that comprise the basis of the Linguistic and Affective Indicators Profile (LAIP), a questionnaire designed to supplement the traditional intake interview.

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The 12 Problem Descriptors

- 1. Misunderstanding what I say**
- 2. Frustration in trying to speak**
- 3. Saying the wrong thing at the wrong time**
- 4. Using words I would not expect a child of that age to use**
- 5. Daydreaming during tasks**
- 6. Difficulty making friends of the same age group**
- 7. Avoiding noisy gatherings**
- 8. Crying without apparent cause**
- 9. Changing moods without an apparent reason**
- 10. Breaking things, hitting, biting or screaming when upset**
- 11. Difficulty going to sleep at night**
- 12. Looking, or complaining about feeling, really tired in the morning**

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Shapiro-format items

In 36 questionnaire items, two statements were paired in Shapiro Personalized Questionnaire format (Hobson & Shapiro, 1970), differing only in which of three adverbial modifiers—rarely, often and consistently—were used to indicate frequency of display, as in the following example.

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Shapiro-format items

CHECK THE BOX BY THE ONE STATEMENT THAT, OF THE TWO, MOST ACCURATELY DESCRIBES THIS CHILD:

- THIS CHILD CONSISTENTLY LOOKS, OR COMPLAINS ABOUT FEELING, REALLY TIRED IN THE MORNING.
 THIS CHILD OFTEN LOOKS, OR COMPLAINS ABOUT FEELING, REALLY TIRED IN THE MORNING.

CHECK THE BOX BY THE ONE STATEMENT THAT, OF THE TWO, MOST ACCURATELY DESCRIBES THIS CHILD:

- THIS CHILD RARELY SEEMS TO MISUNDERSTAND WHAT I SAY.
 THIS CHILD CONSISTENTLY SEEMS TO MISUNDERSTAND WHAT I SAY.

CHECK THE BOX BY THE ONE STATEMENT THAT, OF THE TWO, MOST ACCURATELY DESCRIBES THIS CHILD:

- THIS CHILD RARELY SEEMS TO HAVE DIFFICULTY GOING TO SLEEP AT NIGHT.
 THIS CHILD OFTEN SEEMS TO HAVE DIFFICULTY GOING TO SLEEP AT NIGHT.

CHECK THE BOX BY THE ONE STATEMENT THAT, OF THE TWO, MOST ACCURATELY DESCRIBES THIS CHILD:

- THIS CHILD OFTEN SEEMS FRUSTRATED IN TRYING TO SPEAK.
 THIS CHILD RARELY SEEMS FRUSTRATED IN TRYING TO SPEAK.

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Yes/no and VAS-format items

In another 12 items, problem descriptors were presented without the adverbial modifiers, and the respondent indicated whether or not the descriptor represent an actual observed problem.

- Negative responses on these 12 items served as a check against false-positive indications from Shapiro-format items (e.g., those forcing a choice between statements containing often and consistently, when rarely in fact would have been accurate).
- Positive responses were to be elaborated using a visual analog scale (VAS) indicating recency of onset, as in the following example.

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Yes/no and VAS-format items

MISUNDERSTANDING WHAT I SAY

IS NOT A PROBLEM WITH THIS CHILD IS A PROBLEM WITH THIS CHILD

IF THE ABOVE IS A PROBLEM WITH THIS CHILD, THEN PLEASE USE THE SCALE BELOW TO INDICATE WHEN, TO THE BEST OF YOUR KNOWLEDGE, IT FIRST BECAME A PROBLEM. IF EITHER STATEMENT BELOW IS TRUE, THEN CHECK THE BOX BESIDE IT. IF NEITHER STATEMENT IS TRUE, THEN MARK AN 'X' ON THE VERTICAL LINE TO APPROXIMATELY INDICATE WHEN, SINCE YOU FIRST KNEW THE CHILD, THE PROBLEM BEGAN.

THIS HAS BECOME A PROBLEM JUST SINCE THE PAST TWO WEEKS.

THIS HAS BEEN A PROBLEM SINCE I HAVE KNOWN THIS CHILD.

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Subjects

Twelve adults residing in the Wabash Valley region, comprising couples parenting 6 children previously on the caseload of the first author, served as respondents. The children are described in the table below (all ages reported in months as of 1 January 2004).

CHILD	IDENTIFIER	AGE	DEVELOPMENTAL FACTORS	SPEECH-LANGUAGE Dx	RESOLVED	SUSPECTED
1	MALE A	126	1. ADOPTED 2. ATTENTION DEFICIT-HYPERACTIVITY DISORDER	PHONOLOGICAL DELAY		
2	FEMALE A	88	1. ADOPTED 2. BILINGUAL HOUSEHOLD	ATYPICAL STUTTER		TOURETTE SYNDROME
3	FEMALE B	116	HOME-SCHOOLED	ATYPICAL STUTTER	PHONOLOGICAL DELAY	
4	FEMALE C	113	WILLIAMS SYNDROME	PHONOMOTOR DELAY		
5	MALE B	112	PERVASIVE DEVELOPMENTAL DISORDER	EXPRESSIVE REGRESSION		AEA
6	MALE C	80	ADOPTED	PHONOMOTOR DELAY		ADHD

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Preliminary field trials

Trial 1 LAIP questionnaires were mailed out in early January 2004; all were completed and returned by the end of that month. In response to comments regarding the last 12 items (which several respondents declined to complete until uncertainties could be addressed in telephone conversations), these items were revised for Trial 2 as in the following example. (Trial 2 LAIP questionnaires were mailed out in mid-April 2004; all were completed and returned by late May 2004.)

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Revised yes/no and VAS-format items

MISUNDERSTANDING WHAT I SAY

CHECK ALL THAT APPLY, MARK LINES WITH 'X' TO INDICATE APPROXIMATE TIME THIS PROBLEM BEGAN AND/OR ENDED

- NEVER HAS BEEN A PROBLEM FOR AS LONG AS I HAVE KNOWN THIS CHILD, FROM THE EARLIEST TO THE PRESENT
- PROBLEM BEGAN: EARLIEST _____ PRESENT
- PROBLEM ENDED: EARLIEST _____ PRESENT
- HAS BEEN A PROBLEM FOR AS LONG AS I HAVE KNOWN THIS CHILD, FROM THE EARLIEST TO THE PRESENT

FRUSTRATION IN TRYING TO SPEAK

CHECK ALL THAT APPLY, MARK LINES WITH 'X' TO INDICATE APPROXIMATE TIME THIS PROBLEM BEGAN AND/OR ENDED

- NEVER HAS BEEN A PROBLEM FOR AS LONG AS I HAVE KNOWN THIS CHILD, FROM THE EARLIEST TO THE PRESENT
- PROBLEM BEGAN: EARLIEST _____ PRESENT
- PROBLEM ENDED: EARLIEST _____ PRESENT
- HAS BEEN A PROBLEM FOR AS LONG AS I HAVE KNOWN THIS CHILD, FROM THE EARLIEST TO THE PRESENT

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Scoring the LAIP

All questionnaire responses were entered into a separate interactive spreadsheet per respondent per trial. Three separate measures were calculated: severity rating, categorical rating, and acuteness rating.

- On the Shapiro-format items, selected statements containing the modifier rarely were scored 0, selected statements containing the modifier often were scored 1, and selected statements containing the modifier consistently were scored 2. Three raw scores were summed per problem descriptor and then divided by 5 to yield a severity rating score ranging between 0.2 and 1.
- On the 12 items wherein the respondent indicated whether or not a problem descriptor represented an actual observed problem, each negative response was scored 0 and each positive response was scored 1 to yield a categorical rating score.
- VAS responses measured in millimeters x (from the bottom in Trial 1, and from left in Trial 2) were scaled as suggested by Linacre (1998) using an embedded algorithm (if $x=0,0$; if $x<11,0,05$; if $x<31,0,2$; if $x<70,0,5$; if $x<90,0,8$; if $x<100,0,95$; if $x=100,1$) to yield an acuteness rating score.

Scores were imported from the spreadsheets into data files configured for various statistical analyses.

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Hypothesis

The problem profile derived from questionnaires completed by the parents of Male B--a child whose history of seizure episodes, expressive language regression, and recurring autistic behaviors suggests AEA--should be distinct from the problem profiles derived for any of the other 5 children.

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Correlation analyses

Pearson product-moment correlations (r) were calculated to provide estimates of intrarater, interrater, and inter-item reliability.

- **r was calculated across respondents and measures between the two trials.**
- **r was calculated between maternal and paternal responses across trials and measures.**
- **r was calculated between the three measures and child age across respondents and trials.**

Results are presented in the following table.

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Correlation analyses

VARIABLE	VARIABLE	MEASURE	r	p
TRIAL1 RESPONSES	TRIAL 2 RESPONSES	OVERALL	0.587	<0.001
TRIAL1 RESPONSES	TRIAL 2 RESPONSES	SEVERITY RATING	0.657	<0.001
TRIAL1 RESPONSES	TRIAL 2 RESPONSES	CATEGORICAL RATING	0.497	<0.001
TRIAL1 RESPONSES	TRIAL 2 RESPONSES	ACUTENESS RATING	0.409	<0.001
MATERNAL RESPONSES	PATERNAL RESPONSES	OVERALL	0.309	<0.001
MATERNAL RESPONSES	PATERNAL RESPONSES	SEVERITY RATING	0.305	<0.001
MATERNAL RESPONSES	PATERNAL RESPONSES	CATEGORICAL RATING	0.207	0.013
MATERNAL RESPONSES	PATERNAL RESPONSES	ACUTENESS RATING	0.001	0.993
AGE IN MONTHS	SEVERITY RATING		-0.003	0.959
AGE IN MONTHS	CATEGORICAL RATING		-0.098	0.098
AGE IN MONTHS	ACUTENESS RATING		-0.171	0.004
SEVERITY RATING	CATEGORICAL RATING		0.659	<0.001
SEVERITY RATING	ACUTENESS RATING		0.294	<0.001
CATEGORICAL RATING	ACUTENESS RATING		0.567	<0.001

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Changes in the measures

The acuteness rating measure was eliminated from further analysis for the following reasons:

- Lowest intrarater and interrater reliability among the original measures
- Only weak to moderate correlation with the other measures
- Potentially confounding with age
- Derivation from VAS-format items that respondents found confusing in both trials.

A composite measure (c) calculated from severity rating scores (a) and categorical rating scores (b) was used in developing graphic profiles of problems observed in each child, and for the subsequent cluster analysis.

- The formula $c = (((a+b)/2) - 0.1) \times (10/9)$ eliminated false positives and reconciled discrepancies between the parent measures.
- Intrarater and interrater reliability estimates were comparable to those for the severity rating measure: $r = 0.611$ ($p < 0.001$) between Trial 1 and Trial 2 scores $r = 0.277$ ($p = 0.001$) between maternal and paternal scores.
- Correlations with other measures revealed favorable weighting of categorical rating scores: $r = 0.859$ ($p < 0.001$) with the severity rating measure $r = 0.277$ ($p < 0.001$) with the categorical rating measure.

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Cluster analysis

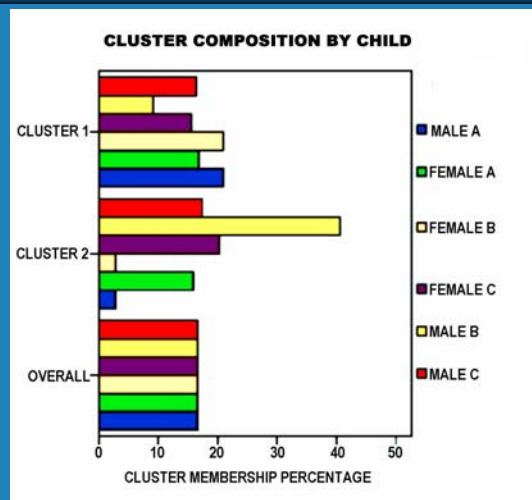
A 2-step cluster analysis was performed to determine the degrees of similarity and difference between the problem profiles for each child.

- Schwarz's Bayesian Criterion was used in defining clusters.
- Categorical variables entered included diagnostic profile and problem.
- Continuous variables entered included the composite measure and its two parent measures (severity rating and categorical rating).
- Spearman's rank-order correlation coefficient (*rho*) was subsequently calculated between
 - the rankings of the first 6 problems defining each cluster
 - the rankings of each child within each cluster.

The results are presented in the following figure and table.

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Cluster analysis



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Cluster analysis

CLUSTER	CHILD	CHILD RANK	PROBLEM (FIRST SIX RANKED FOR CLUSTER)	PROBLEM RANK	<i>r</i> _{pb}	<i>p</i>
1	MALE A	1	Crying without apparent cause	1	0.956	<0.001
	FEMALE B	1	Changing moods without an apparent reason	2		
	FEMALE A	2	Using words I would not expect a child of that age to use	2		
	MALE C	3	Difficulty going to sleep at night	3		
	FEMALE C	4	Misunderstanding what I say	4		
	MALE B	5	Breaking things, hitting, biting or screaming when upset	5		
2	MALE B	1	Daydreaming during tasks	1	0.955	0.003
	FEMALE C	2	Frustration in trying to speak	2		
	MALE C	3	Looking, or complaining about feeling, really tired in the morning	3		
	FEMALE A	4	Avoiding noisy gatherings	4		
	MALE A	5	Difficulty making friends of the same age group	4		
	FEMALE B	5	Saying the wrong thing at the wrong time	4		

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Findings

The problem profile derived from questionnaires completed by the parents of Male B was clearly distinct from the problem profiles derived for any of the other 5 children. This distinction is visible in the graphic profiles, and was confirmed by the results of the cluster analysis.

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Findings

The interrater reliability measure of correlation between maternal and paternal responses, while statistically significant, was weak overall.

Maternal-paternal disparities in both qualitative and quantitative ratings of child behavior are well documented in psychological literature, and recently were analyzed by Treutler and Epkins (2003).

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Limitations and questions

The intrarater reliability measure of correlation between first and second trial responses, while statistically significant, was moderate at best. To what extent does this reflect

- **actual changes in the children profiled over a space of 3 months?**
- **limited sample size?**
- **weaknesses of the instrument?**

The present study was a pilot in which a very limited sample was utilized. How should the sample be expanded and diversified in subsequent trials of the LAIP?

The LAIP is an initial prototype of a screening instrument. What changes might improve the reliability and validity of this instrument?

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Directions for continued research

Immediate revision of the LAIP

- Changing the adverbial modifiers used in Shapiro-format items
- Replacing yes/no and VAS-format items with multiple-choice items
- Graphic and ergonomic modifications
- Algorithms for reconciling maternal and paternal responses

Subsequent revision of the LAIP

- Per suggestions elicited in subsequent field trials
- Wording changes as suggested by an expanded ethnographic research base

Formulation of decision-tree algorithms

- Deriving criteria from clustering factors consistently associated with AEA (and other neurodevelopmental conditions).³¹

Clinical implications

The need for clinicians to develop facility in code-switching from clinical to lay terminology complements the need among laypersons to learn more about neurodevelopmental conditions such as AEA.

Where possible, case histories should be derived from more than one reporter.

Case history interviews should be repeated on a continuing basis.

Summary

The present study represents, at best, a first step towards resolving a clinical problem of undetermined magnitude and complexity.

The importance of a well-articulated lay perspective in helping to direct the path of diagnostic inquiry has been demonstrated nonetheless.

Systematic elicitation and elaboration of the lay perspective in lay terms is a science essential to clinical practice.

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In closing—a cautionary graphic parable about diagnostic categories



“Well—it does fit.”

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