Biomedical Update:

SIBIS: long-term effectiveness seen

When SIBIS (the Self-Injurious Behavior Inhibiting System) was introduced in 1987, opponents of aversives criticized it as "inhumane" (the device delivers a .2-second mild shock after each head-bang), and were skeptical about its long-term effectiveness. While the device has been little-used in recent years because of bans on aversives by many states and school districts, new research shows that its effects are durable and that treatment results in positive, not negative, effects on behavior and emotional state.

Thomas Linscheid and colleagues recently offered follow-up data on three children whose severe, life-threatening self-injurious behavior (SIB) led to their selection as SIBIS candidates. Their findings:

—One subject whose self-injury was successfully controlled with SIBIS was not allowed to use the device after an initial trial, because of objections by aversives opponents. Without SIBIS her SIB continues, despite non-aversive interventions, and she remains institutionalized.

—A second subject, who averaged 300 head hits per hour before SIBIS was introduced, has virtually ceased hitting his head. SIBIS is being successfully "faded," and he now wears the device only about 20% of the time. He has been taught a hand sign which he uses to request that the device be put on. (A number of children using SIBIS ask to wear the device when it is removed.)

Both at home and in the classroom, the researchers say, this subject "has made significant progress in functional and adaptive areas .... He regularly goes into the community on field trips, to restaurants, etc., and has acquired limited single word language."

—The third subject, whose long-term self-injury had seriously damaged her ears and cheeks, showed a 90% drop in SIB during initial SIBIS trials. After five years, her self-injury remains at near-zero levels.

Two other individuals participated in the initial trials; their data were reported elsewhere and were only briefly summarized in this report. In one of these cases, SIBIS successfully suppressed self-injury and trainers were able to fade the device. In the other case, the subject's SIB dropped but then increased slowly, possibly because the device was used only during the staff's regular weekday work shift.

In another study, Linscheid and colleagues specifically studied the behavioral side effects, both positive and negative, of SIBIS use. Their subject was an eight-year-old, nonverbal, profoundly retarded boy with hydrocephalus, whose neurosurgeon recommended SIBIS because the child's head-banging threatened to destroy his brain shunt and injure or kill him.

In addition to suppressing the boy's head-banging, the researchers say, SIBIS treatment resulted in improvements in the boy's mood and in his interactions with his environment. There was no increase in crying, which Linscheid et al. say "suggests that [the boy] was not specifically distressed by treatment with SIBIS."

The data, the researchers say, cast doubt on several popular theories about self-injury. If SIB is an attempt to communicate, they say, "then why does its reduction result in a happier person who interacts more with the environment"—a reaction seen almost immediately, before increases in appropriate communication can occur? And if individuals injure themselves to increase their bodies' production of opium-like chemicals and cause a "high," the researchers say, then it is unlikely that reducing self-injury would lead to improved mood, as it did in their subject.

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"Are aversive procedures durable? A five year follow-up of three individuals treated with contingent electric shock," Thomas Linscheid, Fred Hartel, and Nannette Cooley; *Child and Adolescent Mental Health Care*, Vol. 3, No. 2, 1993; and "Positive side effects in the treatment of SIB using the Self-Injurious Behavior Inhibiting System (SIBIS): implications for operant and biochemical explanations of SIB," Thomas Linscheid, Carrie Pejeau, Sheila Cohen and Marianna Footo-Lenz; *Research in Developmental Disabilities*, Vol. 15, No. 1, 1994, pp. 81-90. Address: Thomas Linscheid, Dept. of Psychology, Children's Hospital, 700 Children's Drive, Columbus, OH 43205.

Williams and autism: a cerebellar link?

It's hard to imagine two more different disorders than autism—a syndrome characterized by aloofness and severe language problems—and Williams syndrome, which has been dubbed "cocktail party syndrome" because people with the disorder are fluent chatterers with remarkable vocabularies. But Swedish researchers say that Williams and autism occur together much more often than chance would explain, and that the two disorders have notable similarities.

Christopher Gillberg and Peder Rasmussen have diagnosed four autistic children (out of a group of about 600 seen at their clinic) who also have Williams syndrome. Prevalence figures, they say, suggest that the disorders would occur together by chance in only 3.5 per 100 million births. Interestingly, the family histories of all four children include learning disabilities, anorexia nervosa, Asperger's syndrome, schizophrenia and/or manic depression.

While Williams and autism appear to be a medical "odd couple," Gillberg and Rasmussen say that other cases of co-occurrence have been documented. Furthermore, they note, symptoms of Williams syndrome include "hyperacusis [over-sensitive hearing], social isolation, and other types of social impairment (such as indiscriminately approaching total strangers), distractibility, inflexibility, ritualism, obsessiveness, and pragmatic deficits (in spite of relatively excellent superficial expressive speech and language skills), all of which can be hallmarks of the autistic syndrome."

They note, in addition, that recent brain studies of individuals with Williams syndrome reveal defects of the cerebellar vermis—the same area that magnetic resonance imaging studies show is defective in autism (see cover story). People with Williams syndrome show over-development or *hyperplasia* of this area, a finding also seen in a significant minority of autistic subjects with cerebellar abnormalities.

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"Brief report: four case histories and a literature review of Williams syndrome and autistic behavior," Christopher Gillberg and Peder Rasmussen; *Journal of Autism and Developmental Disorders*, Vol. 24, No. 3, 1994, pp. 381-393. Address: Christopher Gillberg, Child Neuropsychiatry Clinics, Annedals Clinics, Goteborg, Sweden.

More clues point to immune disorder

New research adds weight to the theory that some cases of autism are caused by an autoimmune disorder—that is, by the body mistaking its own cells for "enemy" cells and attacking them.

Audrius Plioplys and colleagues recently reported a significantly increased incidence of abnormal immune reaction to cerebellar tissue in autistic subjects compared to non-disabled controls. No abnormal reaction to frontal cortex tissue occurred.

The researchers say that "the cerebellar specificity of our findings is particularly intriguing" in light of research by Eric Courchesne et al. (see cover story) implicating cerebellar abnormalities in autism.

In separate research involving the same autistic subjects, Plioplys found abnormal increases in DR+ but not IL-2 receptor+ lymphocytes, a finding which they say "suggest[s] 'incomplete' activation, a finding which is seen in autoimmune diseases." Similar patterns, they note, are seen in juvenile arthritis, rheumatoid arthritis, and multiple sclerosis, all immune system disorders.

"The percentage of DR+ T lymphocytes decreased with increasing age" in autistic subjects, Plioplys et al. note, saying that "this result suggests the possibility of a much more active immune system process early in life, in a subset of autistics, which with aging progressively becomes more quiescent." ***(Editor's note: see editorial, page 3.)***

Both of the studies also tested subjects with Rett syndrome (a progressive disorder which often resembles autism in its early stages). No immune system abnormalities were seen in these subjects.

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"Immunoglobin reactivity in autism and Rett's syndrome," Audrius V. Plioplys, Adonna Greaves, Kamyar Kazemi, and Earl Silverman; *Developmental Brain Dysfunction*, 7, 1994, pp. 12-16; and "Lymphocyte function in autism and Rett syndrome," same authors, *Neuropsychobiology*, 29, 1994, pp. 12-16. Address for both: Audrius V. Plioplys, Division of Neurology, Mercy Hospital and Medical Ctr., Stevenson Expressway at King Drive, Chicago, IL 60616.