
Possible Pubertal Effect on Therapeutic Gains in an Autistic Girl

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A deaf, partially sighted, severely retarded autistic girl, 11 years, 6 months of age, received approximately 2 years of occupational therapy, where sensory integration procedures were employed to reduce self-stimulation. Video-taped time samples of the amount of stereotypies showed a consistent reduction from the time of starting therapy to an interruption for vacation and surgery for scoliosis 46 weeks later. On returning to therapy after a 9-week break, self-stimulation had greatly increased

and did not return to the pre-surgery level during an additional 55 weeks of therapy, 30 of which followed the removal of a cast. Menarche occurred 1 week after removing the cast. Increased self-stimulation is linked to reduced inclination toward environmental interaction as well as to an interruption of intervention and possible pubertal effect. Brief reports on four other autistic adolescents who received similar therapy are consistent with the conjecture of frequent pubertal regression.

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In spite of professional efforts to help autistic children throughout childhood, a number of individuals with this disorder are reported to have increased evidence of neurological abnormalities and regression of behavior at puberty. Onset of seizures at adolescence is not uncommon (1-3). Rutter's (3) review of long-term follow-up studies of autistic children found that adolescence did not bring about major changes in most autistic children, but some did show increased behavioral difficulties. Inertia, sometimes followed by general intellectual decline, was also reported at adolescence. Gillberg and Schaumann (4) focused attention on deterioration and symptom aggravation at the onset of puberty in five cases, three of whom were girls.

This single case report of a deaf, partially sighted, severely retarded autistic girl, age 11 years, 6 months, is consistent with the information accumulating on a possible pubertal regression effect. The client was referred to occupational therapy for the purpose of reducing constant,

intensive self-stimulation, which severely impaired her ability to interact in her social and physical environment. Initially, the client did not engage in voluntary adaptive behavior in the occupational therapy setting. When there was no intervention, she lay on her back and self-stimulated.

Data collection was begun to assess the influence a prolonged interruption of occupational therapy had on the effectiveness of the therapeutic program. Menarche occurred during the assessment period. The possible effect puberty might have on the course of the reduction of her stereotypies was compared to the response of other adolescent autistic clients treated in the same clinic with similar procedures and by case reports presented in the literature. Being aware of this possible regression effect at puberty is important for occupational therapists who make decisions regarding appropriate candidates for therapy and make plans for maintaining the gains upon discharge.

Theoretical Framework of Therapy

Unlike the usual stimulus-response paradigm often used to reduce self-stimulation (5-7), or the approach that employed the principle of over-correction, and the incompatibility of engaging in two different but simultaneous activities (8-12), the rationale for reducing stereotypies in the study client held that the brain is naturally inclined to direct purposeful somatomotor activity and to receive sensory input as a result of that activity. However, the neurophysiology of autism greatly reduces the capacity for engaging in purposeful goal-directed action. Lacking the capacity for more complex interaction with the physical environment, and, further, lack-

ing normal somatosensory processing, the autistic person's drive toward action and sensory input may be expressed in stereotyped actions that produce somatosensory input normally received through interface with the physical world.

If this view is tenable, then improving the autistic person's somatosensory processing and capacity to register, to orient, and to interact purposefully with the world would result in such interaction and reduce the need for the typical autistic patterns of self-stimulation. The adaptive responses would, in themselves, facilitate sensory registration, thus enabling further purposeful activity. To the extent that therapy based on this premise was successful, the autistic person would automatically reduce self-stimulation without the need for constant external reinforcement. However, for permanent changes in central nervous system function, the therapeutic process should occur early in life while that system is more plastic.

The proposed theoretical framework is consistent with Rincover's (13) view that self-stimulation may reflect an autistic individual's attempt at receiving necessary sensory input, and with Ornitz's (14) proposal that spontaneous abnormal motility may be a way of making sense out of sensations through kinesthetic feedback or a means of compensating for physiological sensory deprivation. The idea that providing meaningful sensory input to improve sensory registration may be beneficial in the reduction of self-stimulation is supported by Thelen's (15) findings of reduced stereotypies in infants who receive vestibular stimulation and by those of Bonadonna (16) who reduced stereotypic rocking for 1 hour in three nonautistic severely retarded per-

sons with 4 minutes of linear movement in three different planes. McLean and Baumeister (17) elaborate on this proposal by suggesting that vestibular stimulation improves motor development and that stereotypies decrease as motor development improves. Furthermore, Pribram and McGuinness (18) suggest it is control of the somatomotor system, bringing about a response and providing feedback, that constructs the neuronal models that enable organization of behavior, a deficit area in autistic persons. If lack of control of the somatomotor system results in diminished ability to organize purposeful activity and, consequently, in engaging in non-purposeful stereotyped behavior, then improved somatomotor function might reduce stereotypies. The use of this approach in an attempt to reduce stereotypies was appropriate to the study client since she generally demonstrated hypo-responsivity to sensory input, with the exception of occasional hyper-responsiveness to gravity that suggested poor modulation of input from the vestibular system. In addition, no elicited vestibular postrotary nystagmus was clinically observable. Initially, there was no voluntary participation in purposeful activity when in therapy.

Method

The amount and types of self-stimulation in which the client engaged when placed in an empty room were recorded by videotape the day before starting a course of occupational therapy, and recorded approximately every 6 weeks thereafter for a period of about 2 years. To observe a long-term, rather than the immediate, effect of therapy, taping was done consistently on Monday mornings, which were always several days after the last treatment session.

Had it been anticipated that the client's record would become a formal study, additional baseline data would have been gathered. Regrettably, they were not, and this places a limitation on the interpretation of the data. Types of self-stimulation in which the client engaged were fine finger manipulation and staring, stiffening of an extremity and maintaining that position, masturbation, total body motion (lying on back and shaking all four extremities), and flapping and shaking one or more extremities. Since flapping and shaking varied in intensity, the speed of the motions was judged as mild (one-to-two motions per second), moderate (three-to-five motions per second), or extreme (more than five motions per second). The number of units of videotape (one unit equaled about 3 seconds) in which the abnormal motility was manifested was noted for the first 307 units of tape. To place more unfavorable value on the greater intensity of motility, the number of units of moderate intensity flapping and shaking was multiplied by 2 and the number of units of extreme intensity flapping and shaking was multiplied by 3. The total of unweighted plus weighted units of self-stimulation was considered the "amount" of self-stimulation for a given date. Interrater reliability of the total amount of self-stimulation on 1 day's tape observation was computed at 83 percent, determined by dividing the lesser amount obtained by one author by the greater amount obtained by the other author, and multiplying by 100. All reported measurements of the amount of self-stimulation were made by the same observer.

Specific therapeutic procedures included extensive initial use of a Woodson oscillator (19) to stimulate the macular (gravity and linear

motion) receptors of the vestibular system. The oscillator provided back and forth motion at three cycles per second with excursions of either 0.467 or 0.794 centimeters. Tactile stimulation was provided through brushing and proprioceptive input through joint traction and compression. As the client began to demonstrate the ability and desire to engage in purposeful activity, a net hammock with both ends suspended from a common overhead point served as a swing, which was low enough to keep her feet on the ground in order to ameliorate a feeling of gravitational insecurity. Later she advanced to lying prone in the net and swinging herself for both linear and rotary axial movement designed to stimulate the vestibular system. As more complex environmental action developed, she voluntarily swung prone in an inner tube hanging vertically, climbed a jungle gym, rolled over bolsters, and swung with a trapeze.

The client received occupational therapy for two 50-minute sessions per week over a 111-week time span. Absences of an approximate total of 5 or 6 weeks occurred each of the 2 years, and an additional 10-week absence for surgery for scoliosis. For 26 weeks following surgery and while the client was in a full body cast, therapeutic activities were restricted to some passive stimulation of the tactile and vestibular systems' sensory receptors while lying on a nontilting platform-type swing. Strong vestibular stimulation was avoided because its effect on the client's seizurelike episodes was unknown.

During the school year, including summer school, the client attended a special education class appropriate to her condition. She was absent from school 1 month for surgery.

Results

The amount of self-stimulation in which the client engaged during the time samples and the events that, in addition to therapy, possibly influenced the amount of motility are shown in Figure 1. Both variables are plotted relative to the number of weeks from the start of therapy. The quantity of abnormal activity decreased steadily to the last videotaping before surgery, then rose sharply after return to therapy. Subsequent samples of behavior showed variable amounts of self-stimulation that never decreased to the level achieved before the interruption in treatment for surgery.

During the 10th week of therapy, the client had an involuntary motor event consisting of gritting the teeth, simple nonlanguage vocalization, and strong arm and leg extension with some hip flexion. The episode lasted about 3 minutes, followed by a period of quiet, deep breathing, and the appearance of sleepiness. The client's parents reported a previous occurrence of similar behavior and that the client had demonstrated these symptoms on the way to the clinic the day it was first observed during therapy. The parents considered the behavior to be reactions to frustration rather than seizures. Electroencephalographic results were negative. Medication (Depakene) reduced their occurrence, but was discontinued because of the side effects. These manifestations of a neurophysiological event occurred occasionally throughout the rest of the time the client received therapy.

Around the 20th week of therapy the client began to demonstrate the ability and desire to use the therapeutic equipment, all of which was designed to stimulate the somatosensory system and elicit adaptive motor responses. The advent of an

Figure 1

Amount of self-stimulation throughout an interrupted course of occupational therapy

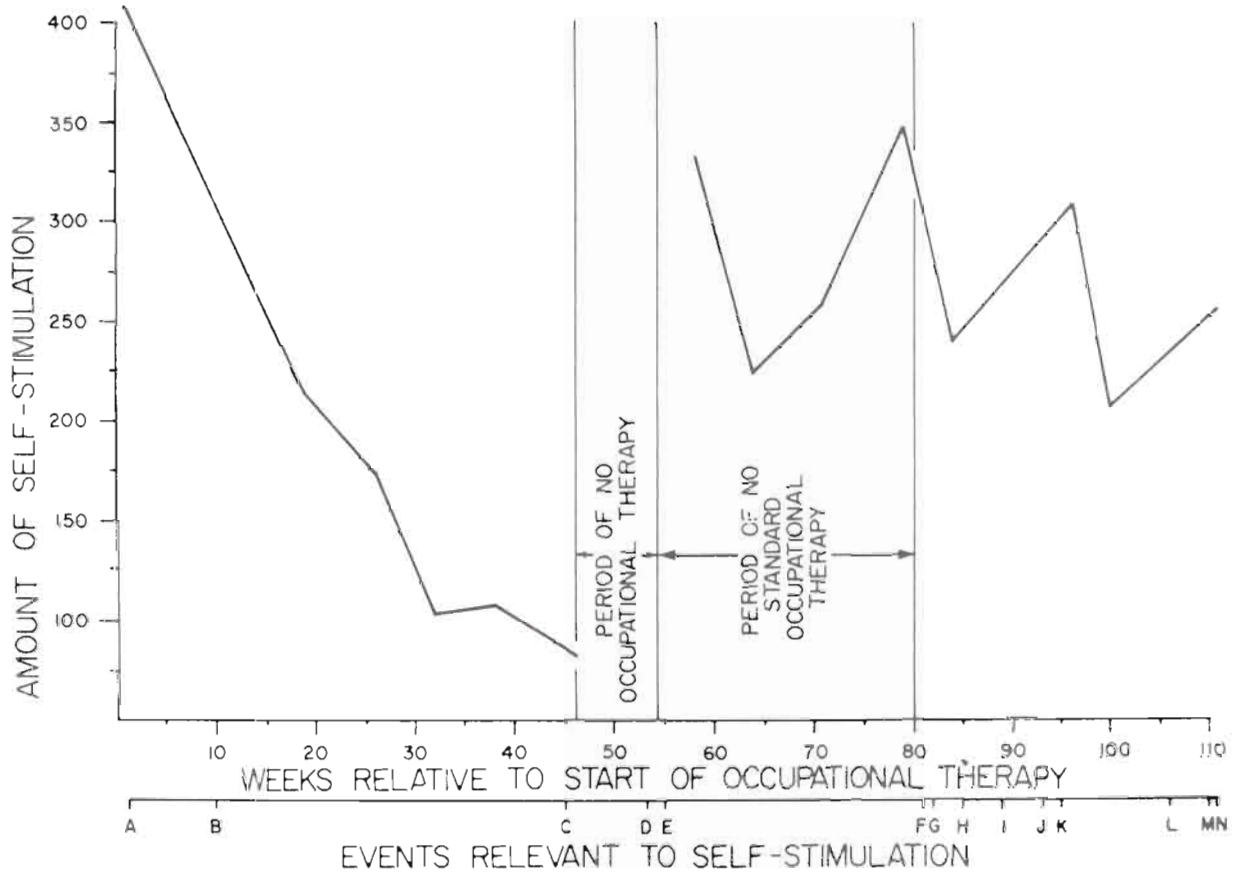


Figure 1 Event Legend

Event	Week	Event
A	1	Started occupational therapy
B	10	Seizurelike motor episodes noted
C	45	Last treatment before surgery
D	53	Surgery for scoliosis
E	55	Return to occupational therapy with full body cast
F	81	Cast removed; no further activity restriction
G	82	First menstrual period
H	85	Started Depakene for seizurelike episodes
I	89	Second menstrual period
J	93	Stopped Depakene because of side effects
K	95	Third menstrual period
L	106	Started Dilantin for seizurelike episodes
M	110	Last occupational therapy session
N	111	Fourth menstrual period

inclination to engage in purposeful activity during therapy was accompanied by a corresponding decrease in stereotypies both in the clinic and at home. On the 32nd week, it was observed on videotape that the client appeared to look for an opportunity to engage in purposeful activity, but, finding no appropriate equipment in the videotaping situation, began to self-stimulate. After 45 weeks of occupational therapy the client took a 9-week leave of absence for a vacation, followed by surgery for scoliosis.

On the 55th week after first starting therapy, the client returned to therapy with a full body cast. During this period the client was noticeably less interested in both sensory

input and in engaging in purposeful activity than she was before surgery. She often arrived at the clinic in an agitated state or appeared disorganized. The body cast was removed at the 81st week and menarche occurred 1 week later. Activity was no longer restricted and attempts were made to help the client regain the level of environmental interaction previously achieved and a reduction of the self-stimulation. Although the skill necessary to engage in adaptive activity did not seem reduced, the inclination and willingness to do so was less.

Discussion

The course of self-stimulation was initially consistent with the theoretical formulation. As adaptive responses emerged, the quantity of self-stimulation began to diminish. Following the interruption in therapy and the application of non-standard procedures, both increased self-stimulation and loss of willingness to interact with the physical environment occurred. In conjecturing about the reason for the loss of previously attained gains toward more normal behavior, one must consider a number of events that centered about her absence from therapy, the surgery, possible seizures and medication for them, puberty, and the interaction of all of these conditions.

Gains from educational and therapeutic programs are more easily lost by the autistic than the nonautistic child if effort and experience are not maintained. Such a loss might account for the return of the stereotypies following absence from therapy. Wearing a cast for a prolonged period, which did not interfere with self-stimulation, but did interfere with sensorimotor activity, would also favor a return to pre-

vious, less adaptive motor patterns. However, it cannot be argued convincingly that the surgical procedure and accompanying break in standard therapy in themselves left the client with a much different capacity for response to the therapeutic procedures from that with which she first entered therapy. Yet the amount of self-stimulation recorded at 28 weeks after resumption of standard therapeutic procedures following surgery was far greater than that recorded at 26 weeks after first starting therapy.

The introduction of medication for seizures confounds interpretation of the data, yet the frequency with which seizures start at puberty in autistic children must be considered one of the possible pubertal changes in autism. The effect the anesthetic used for surgery could possibly have contributed to the client's change in stereotyped behavior.

Related Case Reports. Support for the probability that puberty and the various conditions associated with its onset was the major factor that interfered with the client's return to earlier levels of self-stimulation is found in the responses to therapy of the only other autistic individuals who had received similar therapy by the authors at a time when puberty onset effects could be observed. Younger autistic clients had been treated with varying results (20). Brief reports on the other medically diagnosed adolescent autistic clients treated follow.

A girl who was treated for a period of 19 months between ages 10 years, 8 months and 12 years, 3 months showed good gains in the ability to organize her behavior in relation to the physical environment, in reduced self-stimulation, in more appropriate verbalization, and in the ability to interact with

others, including more eye contact with the therapist. Originally, she had little motivation to engage in activities demanding simple somatomotor adaptive responses. She expressed a repeated preference for lying down and going to sleep. In time she developed good task orientation, a willingness to participate, and the ability to attend to a task such as traversing a five-piece obstacle course. Head rocking was reduced from a high frequency to a low frequency, with none at all seen during one session in the ninth month of treatment. Social interaction in the clinic developed first to parallel play, then to cooperation, with taking turns.

At the end of a year of treatment the client took a 3-week vacation and, on her return, was initially very disorganized and had reverted to frequent self-stimulation and task avoidance. None of the skill in purposeful sensorimotor activity was lost and some of the former interest in it returned, but self-stimulation did not subside with further treatment after the vacation. Emotional lability, previously not a problem, occasionally interfered with therapeutic participation and caused considerably trouble at home. The client's mother reported that she refused to do what was asked of her and had "violent" temper tantrums that lasted 20 minutes and required physical restraints. A year earlier, her minor tantrums were handled at home by having her sit in a corner for a few minutes. At the time of discharge, menarche had not occurred, but secondary sex characteristics (breasts, pubic hair) were beginning to appear. It may be that, in this client and in the study client, aggravation of abnormal symptoms was facilitated by an interruption in therapy concurrent with the onset of puberty.

A severely retarded autistic boy without a history of seizures or behavior problems was treated between the ages of 12 years, 10 months and 15 years, 6 months with a 3-month absence after the first year of treatment. The client demonstrated modest gains in environmental awareness and interaction that plateaued during the last months of therapy. Six weeks before discharge, the client had a seizure and a month before that, his school teacher reported regression in task performance. No regression was seen at home or in occupational therapy.

Another 12-year-old autistic boy was referred to occupational therapy because he had reached a plateau and his behavior was regressing. After 18 months of therapy his treatment was terminated because response was minimal. In contrast to this client, satisfactory gains in language, cognition, and social response were made throughout 19 months of therapy with another autistic boy treated between the ages of 14 years, 10 months and 16 years, 6 months. This client demonstrated hyper-responsivity to tactile, auditory, and olfactory sensory input. Autistic persons with hyper-responsivity to sensory input may be more apt to respond to sensory integration procedures than those who are hyporesponsive (1).

Conclusion

The few cases reported here show results that are consistent with Gillberg and Schaumann's (4) observation of deterioration about the time of puberty in about a third of autistic children. The regression was most noticeable in self-stimulation and in social-emotional behavior. Gains in environmental awareness and ability to interact with the physical world plateaued but did not appear to be lost; however, the

inclination toward such interaction diminished. These changes are not unlike those observed among the nonautistic, but brain-dysfunctioning, clients who received therapy employing sensory integration procedures. The theory underlying the therapeutic use of sensory integration procedures is based partly on the capacity to enter into the neurobiological development of the child during the early critical period for maturation of sensory integrative mechanisms. That period is past by the onset of puberty. In all the five cases described, therapy was initiated after the 10th birthday.

Occupational therapy must continually evaluate the benefits of various treatment approaches within different dysfunctional groups. The results of this study suggest that attempts to improve the autistic person's interface with the environment may not be optimal when therapy is initiated at adolescence. Further, the tendency for autistic persons to lose what has been gained may need to be countered by a permanent home program. Continued research is needed in order to enhance therapists' ability to determine which autistic children are most likely to exhibit regression at puberty and which intervention strategies are most successful in minimizing this occurrence.

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