

典型的なトゥレット障害患者は社会的であり周囲へ気を使う傾向が強く、気持ちの表現も上手なことが多く、自閉症の3主徴に含まれる対人的相互反応の質的障害およびコミュニケーションの質的障害に該当する特徴は認め難い。しかし、残る一つの自閉症の主徴である行動、興味および活動の限定され、反復的で常同様な様式については、トゥレット障害とPDDとで共通性が高いと思われる。すなわち、どちらも、やっつけられないと思えば思うほどやっつけまうという衝動性の高さで特徴付けられる強迫様症状を有している(金生, 2008)。

自験例で自閉症またはアスペルガー障害が確定できず知的な遅れを伴わないトゥレット障害患者44名について詳細な評価を行ったところ、最終的に4名がPDDであると判明した。トゥレット障害患者にPDDを併存する頻度は9.1%となった。この4名の患者と同時期に外来通院中であり、知的な遅れはないものの当初から自閉症またはアスペルガー障害と診断されていたため対象から除外された患者が6名いたため、それらを加えて、トゥレット障害とPDDとの併存患者10名について検討した。PDDを伴わないトゥレット障害患者40名と比較して、チックの種類、AD/HD症状や自傷行為の頻度などに大差はなかった。しかし、OCDを併存する割合は、PDDを伴う場合に60%であるのに対して、PDDを伴わない場合に20%であり、トゥレット障害とPDDとの併存患者で有意に高率であった。しかもOCDと診断された患者全員が、強迫症状が過剰である、または不合理であると認識したことがあり、自閉症のこだわりとは明らかに異なる典型的な強迫症状を有していた。トゥレット障害とPDDが強迫性を共有すると改めて確認されたと言えよう。また、詳細な評価の後にPDDの併存が確定した4名は、チックが比較的重症であり、OCDまたは強迫症状を併存して

おり、さらに、意欲減退、注意散漫、睡眠障害、不登校などの行動上の問題を伴っていた。薬物療法などによってチックが軽減して活動性も高まるにつれて、相手の気持ちや場に合わせた言動がとれないことが明らかとなり、不適応の基盤にPDDが存在することが浮き上がってきたと思われる。

このトゥレット障害とPDDとの併存患者10名は、チックの発症年齢の平均が約7歳であり、トゥレット障害としては標準的であった。チックのための最初の医療機関受診年齢の平均が約10歳であると同時に、PDDに気づかれた年齢の平均も約10歳であり、チックに加えて対人関係などの問題が目立ってきてついに受診になった可能性があると思われた。不登校は60%と高率であり、トゥレット障害とPDDとの併存が学校での適応に与える影響の大きさがうかがえた。同時に、50%が通級学級や適応指導教室の利用を含めた特別な教育的支援を受けていた。

Ⅶ. 治療から見た併存症

治療のためにはトゥレット障害患者を総合的に評価することが重要である。トゥレット障害の重症度としては、チックの頻度、強さ、複雑さ、行動や発語への影響などからなるチック自体の重症度、自己評価や社会的機能に対するチックの直接的な悪影響の重症度を把握する必要がある。同時に、これまで述べてきたような併存症の有無および重症度を考慮する必要がある。併存症のない場合もあるが、複数の併存症を有する場合も少なくないだろう。チックとすべての併存症を見渡して治療の優先順位をつけていくことになる。

治療の優先順位を考える上では、患者本人および周囲の受けとめ方を把握することも重要である。例えば、患者は極めて頻回の瞬目チックがうっとうしくて気になっているのに対して、

家族や学校は他児とのトラブルの改善を求めているというように、患者と周囲とで問題にしている点が異なることはしばしばである。一見したところ患者がチックも併存症も認識していないようで、丁寧に聞いてみると意外と本人なりに気にしているが、そのことに家族は気づいていない場合もある。また、患者の年齢や発達水準、不安の強さなどによって受けとめ方は異なってくるだろう。いずれにしても、チックや併存症のどれからアプローチしたら患者の自己肯定感や社会的機能を保って生活しやすくなるのかとの観点で検討することが必要である。

VIII. おわりに

ジル・ド・ラ・トゥレットによる報告で重要視されたコプロラリアとエコラリアは現在のトゥレット障害の診断基準に含まれていないが、やってはいけないと思えば思うほどやってしまうという強迫性と衝動性で特徴づけられる複雑音声チックである(金生, 2003)。トゥレット障害はチック障害であると同時に強迫性と衝動性が問題になると考えると、ここで取り上げた併存症はまさにトゥレット障害に併存するべくして併存していると言えよう。それも考慮に入れてつつトゥレット障害の本態の検討を深めて、より適切な治療につながる事が望まれる。

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◇質疑応答◇

第1セッション「小児のトゥレット障害」

質問：杉山登志郎先生

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①トゥレット障害の特に汚言症(Coprolalia)がフラッシュバックによる強迫症状とよく似ていることに気づいた。チックとフラッシュバックの関連性について教えていただきたい。

②怒り発作とトラウマとは関連があるのか。

回答：金生由紀子

チックとトラウマとの関連性に関する研究はないのではないかと、自分自身の経験の中ではチック、トゥレット障害の全てがトラウマを抱えているわけではなかった。

また、汚言症も幅があって、トラウマ(いやな記憶)と結びついているために相手を傷つけてしまっているのではないかと自覚している人と、自動的に(典型的なチック症状として)汚言が出てしまう人があるように思われる。

回答：星加明徳

小児科領域では怒り発作は少ないので、関連性を説明するのは難しい。自分自身の経験ではこれまでに300人以上の患児の中で、怒り発作と思われる症例は1例しかない。

回答：金生由紀子

強度行動障害の研究の中で、トゥレット障害を併存した自閉症、またはPDDがいるという報

告があった。強迫性、衝動性を持つという点で共通する部分があるので、それぞれの幅をどこまで広げるか、どこで区切るかは難しいと思う。チックは明確な運動症状があることで区切り、その上でスペクトラムと考えるといいと思う。今後の検討を要する。

質問：平野浩一先生(浜松医科大学小児科)

①全身性チックの「ミオクロースス様」の動きはミオクローススとは別なのか。

②ミオクローススの鑑別において器質的疾患が除外されると「心因性ミオクロースス」との診断が教科書に記載されているが、チックと同じものなのか。

回答：星加明徳

①ミオクロースス様の動きはチック症状の経過の一部で出現する。トゥレット障害のチックの動きには多様性があり、ジストニー様とかミオクロニー様と表現されているものがある。基本的には神経疾患のミオクローススと同じものとする。

②私自身は「心因性ミオクロニー」といわれるものはないと思っている。ゆくゆくは神経学的な機序が見つかると思うが、現在は仮に「突発性」「心因性」と言わざるを得ない。今回提示したミオクロニー様の動きはトゥレット障害の経過中に出現する神経症状(あるいはチック症状)と考えている。

* * *

Short Communication

Pervasive developmental disorder with attention deficit hyperactivity disorder-like symptoms and mismatch negativity

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The present study examined the correlation between the attention deficit hyperactivity disorder (ADHD) Rating Scale-IV Japanese version (ADHD RS-IV-J) score and mismatch negativity (MMN), in 10 pervasive developmental disorder (PDD) children with ADHD-like symptoms, and examined whether MMN become the objective measure to assess the severity of ADHD-like symptoms in PDD children. Consequently, score of ADHD RS-IV-J had a positive

correlative tendency with MMN latency and had a significant strong negative correlation with MMN amplitude. Therefore, MMN may become an objective measure to assess the severity of ADHD-like symptoms in PDD children.

Key words: attention deficit hyperactivity disorder, event-related potential, mismatch negativity, pervasive developmental disorder.

EVENT-RELATED POTENTIALS (ERP) contribute to the research in cognitive function as physiological measures that can be easily measured and which are non-intrusive. Therefore, ERP are useful measures to examine cognitive disturbance of children with developmental disorder. Mismatch negativity (MMN), one of the ERP, reflects an automatic cerebral discrimination process, not under attentive control.¹ We previously indicated that MMN might be an objective measure of the severity of attention-deficit-hyperactivity disorder (ADHD) symptoms in ADHD patients.²

In the DSM-IV, comorbidity of pervasive developmental disorder (PDD) and ADHD is not recognized and also PDD is an exclusion criterion of ADHD.³ In recent years, however, an increasing number of cases have involved difficulty in distinguishing PDD from

ADHD. Therefore, an objective measure of ADHD-like symptoms in PDD patients is needed. In the present study we examine the correlation between the ADHD Rating Scale-IV Japanese Version (ADHD RS-IV-J)⁴ score and MMN, in PDD children with ADHD-like symptoms.

METHODS

Subjects

Ten children diagnosed with PDD based on DSM-IV,³ participated in the present study. They also had ADHD-like symptoms. All subjects were male and right-handed. Their mean age was 8.60 ± 1.65 years. Their mean Wechsler Intelligence Scale for Children-Third Edition Full/Verbal/Performance Intelligence Quotient scores were 101 ± 15.7 , 107 ± 21.8 , and 93.2 ± 10.5 , respectively. The mean ADHD RS-IV-J score was 25.4 ± 5.95 .

All subjects and/or their parents gave informed consent to participate in the study. In addition, this study was approved by Institutional Review Board of Nara Medical University Hospital.

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MMN measurement

MMN was measured using auditory odd-ball tasks. An NEC Multi Stim was used as the auditory stimulus system (NEC, Tokyo, Japan). Standard stimuli ($P=0.9$) were tone bursts at 1000 Hz and deviant stimuli ($P=0.1$) were tones at 1100 Hz, with all stimuli lasting 50 ms at 500-ms intervals and an intensity of 80 dB. These infrequent and frequent stimuli were given in random order through headphones. MMN was measured while the subjects were reading books of their choice, without paying particular attention to the auditory stimuli given, as instructed (READ condition).

MMN recording

MMN was recorded with an NEC Synax 1200 (NEC, Tokyo, Japan). Electroencephalograms were obtained at Fz, Cz and Pz on the scalp using disk electrodes, with both ear lobes as the reference electrode sites. MMN was analyzed during the period between 100 ms pre-stimulus and 400 ms post-stimulus. The amplitude was measured with the potential of 0 ms latency as the baseline.

MMN analysis

One hundred responses to infrequent deviant stimuli and 900 responses to frequent standard stimuli were averaged separately. The waveform of the latter responses was subtracted from that of the former. MMN was identified as negativity with the peak latency at 100–250 ms based on the subtraction waveform.

Statistical analysis

The correlation between the ADHD RS-IV-J score and MMN was examined using the Pearson coefficient of correlation (r). Data are expressed as the mean \pm SD. Data analyses were conducted using SPSS version 11.0 for Windows (SPSS, Tokyo, Japan).

RESULTS

As shown in Table 1, score of ADHD RS-IV-J had a significant strong negative correlation with MMN amplitude recorded at Cz and Pz and had a positive correlative tendency with MMN latency recorded at Fz and Cz.

Table 1. ADHD RS-IV-J score and MMN measures

Pearson correlation Coefficient	r	Significance
MMN latency		
Fz	0.57	*
Cz	0.56	*
Pz	0.54	NS
MMN amplitude		
Fz	-0.54	NS
Cz	-0.81	**
Pz	-0.80	**

* $P < 0.1$, ** $P < 0.01$. NS, not significant.

ADHD RS-IV-J, Attention-Deficit-Hyperactivity Disorder Rating Scale-IV Japanese version; MMN, mismatch negativity.

DISCUSSION

We previously reported that MMN amplitudes of the ADHD children were smaller than those of the healthy control children.⁵ But neither MMN amplitude nor latency has been reported to differ significantly between the adult ADHD group and the adult healthy control group.^{6,7} Therefore, in the ADHD group, MMN dysfunction might be able to improve with maturation.^{6,7} In addition, Achenbach *et al.* reported that symptoms of hyperactivity/impulsiveness were ameliorated with maturation and that symptoms of attention deficit remained in a large number of the ADHD combined-type children.⁸ Therefore, MMN has been indicated to be associated with hyperactivity/impulsiveness of ADHD.

We also reported that ADHD RS-IV-J score had a significant strong positive correlation with MMN latency and had a significant strong negative correlation with MMN amplitude in the ADHD combined-type group.² Therefore, MMN might be an objective measure of severity of ADHD symptoms in the ADHD group.

The present study indicates that there may be a correlation between ADHD RS-IV-J score and MMN in the PDD group with ADHD-like symptoms as well as in the ADHD group. Therefore, MMN may be an objective measure of the severity of ADHD-like symptoms in the PDD group with ADHD-like symptoms. We indicate a strong possibility that hyperactivity/impulsiveness and attention deficit associated with PDD are not completely different from those associated with ADHD when considered

in the context of ERP. Further studies are needed to determine whether there are PDD patients with comorbid ADHD.

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注意欠陥/多動性障害とチック障害を 併存した兄弟例*

WISC-ⅢとADHD RS-Ⅳ-Jの検討

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はじめに

注意欠陥/多動性障害(以下、AD/HD)は、不注意、多動、衝動性を主症状とする行動の障害であり、児童精神医学領域ではもっとも有病率の高い障害の一つである。現時点で、神経生物学的な背景と強い遺伝的な素因が関与し、さらにその他の要因が複雑にからむ多因子性の病因が考えられている。

一方、チックは幼児期の後半から児童期に生じやすい運動症状で、それを主症状とする症候群がチック障害である。以前は、一過性のチック障害

は心因性、慢性のチック障害は脳器質性と別々に考えられていたが、現在ではチック障害は連続しており、遺伝的要因と環境要因との絡み合いが関与するとされている。

AD/HDでは、一般よりもチックの併存の頻度は高く、チックの頻度が、一般の小児では6%であるのに対して、AD/HDでは34%であったとの報告⁸⁾がある。一方、チック障害の併存症の中でもAD/HDはその頻度が高く、50%以上に及ぶという報告⁶⁾もある。

これらの報告から、AD/HDとチック障害の病態、病因に共通または密接な関係があると考えられており、近年の脳画像研究からは、両者に共通して特定の皮質-線条体-視床-皮質回路(CSTC回路)の関与が想定されている⁵⁾。AD/HDとチック障害との遺伝的関係への一定の結論は得られていないが、近年分子遺伝学的研究では、両者にドーパミンD₄受容体遺伝子の関連性^{2,3)}があることが示唆されている。疫学的な報告では、AD/HDとチック障害の併存した症例の同胞もまたそれらの併存があることはいわれているが、各同胞間で詳細に比較検討した報告はない。

そこで今回我々は、主訴が多動であったAD/HDと一過性チック障害の併存した次男と、主訴が音声チックであったAD/HDとトゥレット障害の併存した長男という兄弟例を経験したので報告し、病歴、家族背景、臨床所見、WISC-Ⅲと

* The Siblings with Attention-Deficit/Hyperactivity Disorder and Tic Disorder: Comparison of the results of assessments using WISC-Ⅲ and ADHD RS-Ⅳ-J

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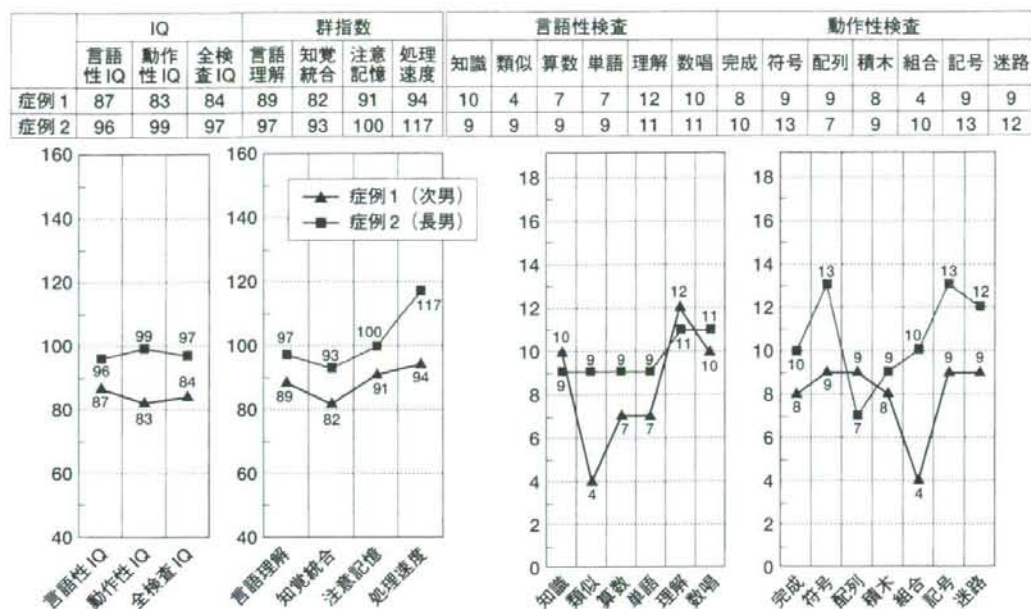


図1 兄弟のWISC-IIIの結果

ADHD Rating Scale-IV 日本語版(以下, ADHD RS-IV-J)の比較を中心に考察した。

症例

〈症例1(次男)〉 初診時年齢6歳(小学校1年生)。

主訴 学校で落ち着きがない。人の話を聞けない。

既往歴 特記事項なし。

家族歴 父は38歳で、会社員。書類に印鑑を押し忘れたり、重要な会議の予定を忘れていたり、と不注意なところがある。母は37歳で、専業主婦。小学校の時、目をパチパチする運動性チックがみられたが自然に消失した。

現病歴 同胞2名中第2子として出生。在胎39週、出生時体重3,180gであった。1歳2か月に独歩がみられ、歩くようになるとよく動き、常に大人が手をつないでいないとならなかった。始語は1歳4か月頃、2語文は2歳にみられた。乳幼児健康診査では何も指摘されなかった。3歳で、かかりつけの小児科医に「多動だね」と言わ

れ、幼稚園に通園し始めた4歳頃から順番を待てないことや、友達をたたくことが目立つようになった。就学前には、マンションの階段の上から排尿する、石や物を投げる、友達を蹴るという問題行動が目立ち、注意されるといったんやめるが繰り返した。X年3月から、喉を鳴らす音声チックと目をパチパチする運動性チックがみられるようになった。X年4月、小学校入学。授業中勝手に話したり、立ち歩いたりすることを担任より聞き、授業参観で周囲をキョロキョロ見て絶えず体をゆすっている本児の様子を母が目当たりしたこと、で両親が心配になり、X年6月15日当科受診となった。

初診時現症 簡単な質問に、視線も合意的に答えられる。馴れ馴れしい印象を受ける。診察室でも飛んだり跳ねたりと常に動く。絵を描いていたかと思うと突然「お兄ちゃん」と呼び部屋を出て行く。

検査結果

①WISC-III(図1) 全検査IQ 84, 言語性IQ 87, 動作性IQ 83。

②ADHD RS-IV-J 家庭版 15 pt (不注意 6 pt, 多動/衝動性 9 pt), 学校版 20 pt (不注意 11 pt, 多動/衝動性 9 pt)。

初診後経過 DSM-IVに基づき, AD/HD と一過性チック障害と診断し, 両親および担任に, AD/HD とチック障害に関する心理教育を行った。X 年 7 月には授業中に立ち歩くことはなくなり, チックもほぼなくなった。その後, 大人が本児とかかわるうえで手をやくようなことは徐々になくなっていった。X 年 12 月, 一方的に話をすることはあるが, チックや行動上の問題はなくなっていた。引き続き, 経過を追っている。

(症例 2(長男)) 初診時年齢 10 歳(小学校 4 年生)。

主訴 目をパチパチするチックがある。喉をクックと鳴らす。

既往歴 特記事項なし。

現病歴 同胞 2 名中第 1 子として出生。在胎 37 週, 出生時体重 2,750 g であった。1 歳 3 か月に独歩がみられ, 始語は 1 歳頃, 2 語文は 2 歳 2 か月にみられた。乳幼児健康診査では何も指摘されなかった。弟が生まれた 3 歳半頃から突発的に首を振る運動がみられたが, 両親はあまり気にかけなかった。幼稚園でよく友達をたたいてしまう時期があった。小学校入学後, 持ち物を玄関に置いていても忘れるような不注意が目立ちだした。学年が上がるにつれて, 大人に反抗することが多くなったが, 両親は「男の子はこんなものか」とあまり心配しなかった。X-1 年 11 月頃から「クッ」と喉を鳴らすようになり, X 年 4 月からは, 目をパチパチとさせたり, 喉を鳴らすことが多くなった。弟が当科を受診したことを契機に, X 年 8 月 10 日当科受診となった。

初診時現症 少しソワソワした様子があるが, 座って質問に的確に答える。時折, パチパチと瞬く運動性チックがみられる。

検査結果

①WISC-III (図 1) 全検査 IQ 97, 言語性 IQ 96, 動作性 IQ 99。

②ADHD RS-IV-J 家庭版 9 pt (不注意 7 pt,

多動/衝動性 2 pt), 学校版 8 pt (不注意 5 pt, 多動/衝動性 3 pt)。

初診後経過 DSM-IVに基づき, AD/HD と診断した。また, 3 歳半頃から首を振る運動性チックが慢性的にみられ, X-1 年 11 月以降は「クッ」と喉を鳴らす音声チックやパチパチと瞬く運動性チックがさらにみられるようになったことから, トウレット障害と診断した。両親に AD/HD とトウレット障害に関する心理教育を行うとともに, トウレット障害に対する薬物療法を始めた。X 年 8 月 10 日, haloperidol 0.75 mg/日を就寝前に服薬とした。その後, もともとあった「クッ」と喉を鳴らす音声チックの頻度が増加したため同薬を 1.5 mg/日に増量したが, 症状の軽減はなく, X 年 10 月 15 日に同薬 3 mg を就寝前のみの服薬とした。X 年 10 月 20 日, 徐々に増強してきた眠気と嘔気の出現のため haloperidol を中止した。約 2 週間の休薬期間を設け, X 年 11 月 4 日より risperidone に変更し, 1 mg/日を就寝前に服薬とした(なお, 薬物療法に際して, 両薬剤が本邦においては適応外使用であること, 副作用などを両親に十分に説明したうえでインフォームド・コンセントを得た)。運動性チック, 音声チックともに徐々に気にならなくなり, 眠気など副作用も認めなかった。同量で経過をみる中, 衝動性による問題があるため服薬方法は変えず, risperidone 2 mg に増量した。その後, 衝動性の低下を認めたため同量を継続し, 経過を追っている。

考察

AD/HD とチック障害の関連性が示唆される中, この兄弟例は両者とも AD/HD とチック障害を併存しているが, 次男は AD/HD 症状が前景にたち, 長男はチックが前景にたっており非常に興味深い。以下に, WISC-III, ADHD RS-IV-J の順に比較検討を行うこととする。

1. WISC-III の比較検討(図 1)

AD/HD では言語性 IQ が動作性 IQ よりも高い^{1,4)}といわれるが, 今回の両者とも, 言語性 IQ

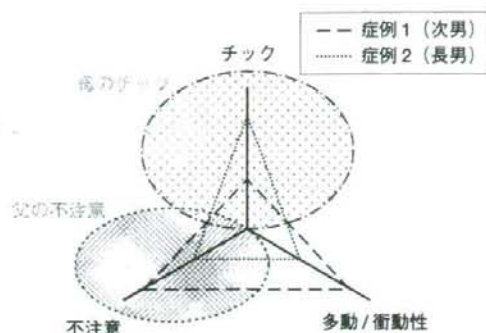


図2 兄弟の臨床所見と家族背景

と動作性 IQ には明らかな差はないといえる。また、集中力と注意力に障害がある AD/HD では群指数の処理速度が低くなるといわれる^{4,7)}が、次男は 94、長男は 117 であり両者とも低くはない。しかし、それらには 23 の差があり、この差は両者の不注意の程度の差とも考えられる。群指数すべてでは、両者に得点の違いはあるが、知覚統合と処理速度に乖離があることや、注意記憶と処理速度の得点点その他の群指数の得点より高いことは類似している。つまり、両者とも空間的な処理能力が弱く、推論することを苦手とし、逆にパターンの、機械的な処理は得意であるといえる。

2. ADHD RS-IV-J の比較検討

AD/HD を疑うべき指標として、ADHD RS-IV-J の家庭版の合計 14~16 pt、学校版の合計 11~21 pt がいわれ⁹⁾、今回の次男では家庭版 15 pt、学校版 20 pt であり、この指標を上回った。長男では、家庭版・学校版ともに AD/HD を疑うべき指標を下回った。しかし、健常児に対する ADHD RS-IV-J の報告⁹⁾によると、9~11 歳の男児の ADHD RS-IV-J 家庭版の平均は合計 6.74 pt、不注意スコア 4.69 pt であり、長男の ADHD RS-IV-J 家庭版の合計 9 pt と不注意スコア 7 pt はそれらを上回っており、長男の不注意の程度が健常児より高いことがうかがわれる。

以上「1.」、「2.」を踏まえチック、不注意、

多動/衝動性という 3 つの観点から両者の特徴をまとめると図 2 のようになる。

チックについては、長男は診断がトゥレット障害であり複数のチックが慢性にみられたのに対して次男は一過性であったように、程度の差がみられた。不注意については、前述した WISC-III や ADHD RS-IV-J から、両者に不注意が存在するが、長男は次男より不注意の程度が低いといえる。多動/衝動性については、両親が述べる主訴からもわかるように、次男については落ち着きのなさが目立つが、長男についてはそれほど目立っていない。また、ADHD RS-IV-J の多動/衝動性スコアからも、次男は長男より多動/衝動性の程度が高いといえる。

また、家族背景も考えると、母の運動性チックの既往と父の不注意がある。臨床的には、AD/HD 患者の家族内に不注意や衝動性を持つ者が存在することがあり、その特徴は社会的、職業的機能への著しい影響はないことをよく経験する。今回の父にも、社会的、職業的機能への著しい影響はないが、不注意がある。この家族背景を図 2 に示したが、母のチックの影響は次男より長男に強く、父の不注意の影響は長男より次男に強く及んだとも考えられる。

おわりに

AD/HD とチック障害を併存した兄弟例の症例提示とともに、両者の臨床所見、WISC-III と ADHD RS-IV-J の比較を中心に考察した。

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Brief Note

**Japan-China Comparative Research Related to
Early Detection of Children With Autism:
Development of Early Autism Diagnostic
and Identification Systems
Adapted to China**

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and Satoru TAKAHASHI****

The present paper elucidates characteristics of early detection of Chinese children with autism compared to comparable Japanese children. Data were obtained from questionnaires completed by the children's parents. In addition, the status of early diagnosis and identification of autism in China is reviewed. Differences between the two countries in relation to the early detection of children with autism provided data useful for developing a system for early diagnosis and detection that was adapted to China. The results of the review revealed significant differences between the situation in Japan and China, especially in terms of 14 of the items: delayed speech and other speech problems, clumsiness, delayed overall motor development, delayed control of neck, not good at forming interpersonal relationships, inability to fit in communal life, unusual habits or patterns; abnormal food preferences, pica and/or refusal to eat; bedwetting, enuresis, or encopresis; self-injurious behavior; violence and aggression; and night crying and night terrors. All of these items were significantly higher in China. Even if autism has a biological basis, its course and expression are affected by the care and educational systems relating to the children who have autism, as well as by the national welfare level. As a consequence, it appears that symptoms of Chinese children with autism are more varied and intense than those of Japanese children with autism. Chinese children have, in particular, more social behavior problems and adjustment disabilities.

Key Words: early detection, early diagnosis and identification of autism, Japan-China comparative research, children with autism

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Introduction

According to the Autism Society Japan (2004), the incidence of autism is about three per thousand. If children with pervasive developmental disorders (PDD) and autistic spectrum disorder (ASD) are included, the incidence rises to one per hundred. Proportionally, there would be more than 13 million children with autistic spectrum disorder, including more than 3.9 million children with autism, among China's 1.3 billion children (National Bureau of Statistics of China). Although when considered in this way, China has a huge number of people with autism, practically speaking, there are no administrative policies or medical, child-care, education, or welfare systems to accommodate them (Lv & Takahashi, 2005).

Only some two decades have passed since the first case report of autism in China (Tao, 1982). During the 1980s and 1990s, Chinese specialists in child psychiatry and special education were insufficiently aware of autism. For that reason, autism was, at that time, categorized as "mental retardation" or as a psychiatric disease of children. Recently, however, the establishment of the Beijing City Autism Society, and the Beijing Star and Rain Educational Institute for Children with Autism, the first private care institutions for children with autism in China, has raised the interest in treatment, research, and education for children with autism. However, as yet, autism has not been publicly accredited as a disability in China, and thus it remains at the margin of policies related to education, medicine, and welfare (Lv, 2006; Lv & Takahashi, 2005).

The first problem in care for children with autism is the medical diagnosis of autism. The diagnosis often determines the direction of subsequent treatment or care education. To discover that children have this disability and intervene as early as possible engenders the reduction of activity constraints caused by the disability. However, little previous research describing early identification of, and care-education for children with autism exists in China; consequently, conventional responses such as diagnosis, treatment, and care-education are largely under-developed.

During 1986-2001, for example, Beijing University Mental Health Research Center statistically analyzed their early detection time of 1,180 cases meeting the autism diagnostic standard in DSM-IV (American Psychiatric Association, 1994), who had asked for diagnosis and treatment throughout China (Liu, Yang, Guo, Liu, Meixiang, & Xiaolv, 2004). They found that the age of onset ranged from 6 months to 6 years (average 22.8 months). Patients who were diagnosed before 3 years old accounted for 24%, and those after 3 years old, 79%. The average time from onset to consultation was 35 months. Those data suggest that many children with autism are not being diagnosed or treated early enough.

On the other hand, investigation of the time of identification of the children's disabilities in Japan, specifically in Tokyo and Shizuoka prefectures, revealed that the earliest time that professionals were consulted about the children's disabilities was

when the children were between 1 year 6 months and 1 year 11 months old, and that the average age when the children's disability was identified was at around two years of age (Nagai & Hayashi, 2004). Japan has one of the best infant medical examination systems in the world. With this system, early detection of developmental disabilities such as autism is possible. Through early detection of disabilities, early treatment-education, and schools' and other educational institutions' or social welfare institutions' assistance, symptoms can be reduced.

Compared to this, Chinese children with autism cannot be given proper diagnosis at an early age, and therefore miss an important period for early care-education.

Recently, research about the biological foundation of autism has advanced rapidly, but its pathologic mechanisms remain unknown. Therefore, for the present, early evidence as expressed by behavior before three years of age is important for early diagnosis of autism. Autism is currently defined by behavioral characteristics, which vary depending on the child's age and development, so early diagnosis is difficult. Furthermore, according to the present diagnostic standards, no symptoms of autism can be clarified before age two, so further research on early evidence must be pushed ahead (Kurita, 2002). In order to treat autism when the child is at an early age, it is necessary to discern evidence that will lead to an early diagnosis.

However, in China, few studies of early evidence and detection of autism have been conducted; in addition, countermeasures by medical institutions and consultation care-education agencies remain largely undeveloped. Meanwhile, although autism is thought to be a disorder of the central nervous system, comparative research on daytime autistic symptoms has revealed that variation in the basic behavioral symptoms is affected by social and cultural influences (Yu & Ohta, 2006).

Based on the problems described above, the present paper is intended to elucidate characteristics of early evidence of Chinese children with autism, through having Chinese parents of children with autism complete questionnaires and comparing those data with data on early evidence in Japanese children with autism, enabling an examination of the early diagnosis and identification of children with autism in China. By revealing differences between Japan and China in the early evidence of autism, and referring to the superior system of early detection and the early treatment-education programs in Japan, basic data will be provided that should assist in the development of a system for early diagnosis and detection adapted to the circumstances in China.

Participants

A sample of 162 children with autistic spectrum disorders (2.6-8.9 years old, average 4.3 years; 147 boys, 15 girls) was employed in the present study. Among the participants, there were 125 children with autism (77%), 28 children with Pervasive Developmental Disorders, Not Otherwise Specified (PDDNOS; 17%), and nine children with Asperger's syndrome (6%). The children were diagnosed at the Sixth Affiliated Hospital of Beijing University (75%), Nanjing Brain Hospital (9%), Beijing

Children's Hospital (7%), the Third Affiliated Hospital of Sun-Yat-Sen University (6%), and other hospitals (4%). The diagnoses of these children were made according to the criteria of DSM-IV (American Psychiatric Association, 1994).

The children were selected from private care-education institutions for children with autism in Beijing and Qingdao in China (the Beijing Star and Rain Educational Institute for Children with Autism, and the Yilin Training Center for Children with Autism, respectively). They came from all over China (16 provinces, 4 direct controlled areas, and 1 autonomous area), suggesting that these data mirror the early evidentiary features of Chinese children with autism.

We also examined data from 141 Japanese children with autism (2-12 years old; 117 boys, 24 girls) by Ohta and Nagai (1987), using the same questionnaire that was filled in by the parents of those children at their first visit.

The children were selected from the Neuropsychiatric Outpatient Clinic of Tokyo University, the Kanagawa Day Treatment and Guidance Center for Children, and two other outpatient clinics. These children were outpatients at the same facilities mentioned above. All of them received treatment during the same time period (January 1981 to December 1982).

Procedure

We produced a 25-item questionnaire, based on items used in the Ohta and Nagai's (1987) study. Items related to behavioral symptoms of children with autism, including early developmental history; the child-care environment; the time when the parents discovered their child's disability; symptoms; visitations and consultations with experts; and early care-education. We distributed 200 questionnaires to parents of children with autism who were being given care-education at the above two institutes. The collection rate was 81%; usable data were obtained from 162 families. The parents of the children (146 mothers, 12 fathers, 4 others) filled in the questionnaire. The present research was executed in August 2005 and November 2006.

The items relating to the symptoms by which the parents had discovered their child's disability for the first time were classified into four groups, following the symptom categories in Ohta and Nagai (1987). Items 1 and 2 were in Group I (delayed or abnormal mental functioning), items 3-7 in Group II (delayed or abnormal gross motor functioning), items 8-23 in Group III (abnormal behavior), and items 24 and 25, Group IV (physical disorders). Additionally, Group III (abnormal behavior) was further subdivided into III A (social behavioral disorders, items 8-14) and III B (adaptive behavioral disorder, items 15-23).

Results

Early Development History

The children of the 162 parents completing the questionnaires include 139 children without disabilities (86%), 12 premature babies (7%), and 11 children with some problems during the neonatal stage (7%). The birth weights were 2,500-5,200 g.

There were no low-birth-weight babies, but 32 heavy babies (20%). The age when they started to walk was 10-20 months. Walking began before 18 months of age in 143 of the children (88%).

Furthermore, 138 of the children (85%) showed speech retardation. Replying to the questions about abnormalities related to the pregnancy and delivery, 28 of the mothers (17%) reported having had strong insecurity or mental stress during pregnancy or at delivery, and 31 mothers (19%) had pregnancy complications such as high blood pressure. The Caesarean section rate was 27%.

Symptoms That Led the Parents to Discover That Their Child had a Disability

In the present study, the first evidence of autism in children with autism in China was regarded as symptoms that were apparent from the perspective of their parents. The age of the children when the parents first realized their child had a disability ranged from 4 months to 4 years 5 months (average 2 years 3 months); the peak period for identification ranged from 1.5 to 2.5 years (51%), supporting the result of 1.5 to 2.5 years (57%) reported by Ohta and Nagai (1987). In all, 98 of the children (60%) had been diagnosed as having autism before they were three years old. Of the parents, 154 (95%) said that their child was diagnosed at a hospital. In many cases, it was the mother (N=89, 55%) who discovered their child's disability first.

Table 1 summarizes the symptoms reported by the parents of children with autism in China. The item "delayed speech and other speech problems" was answered affirmatively by 88%, followed by "behaves as if deaf" (85%), "poor relations in peer group situations" (58%), "not good at forming interpersonal relationships" (56%), "delayed overall motor development" (38%), "retarded-like behavior" (35%), and "abnormal food preference, pica, and/or refusal to eat" (30%).

The most commonly selected items in Japan (Ohta & Nagai, 1987) were also "delayed speech and other speech problems" (84%), followed by "poor response to others" (55%), "restless and hyperactive" (45%), "behaves as if deaf" (32%), and "not good at forming interpersonal relationships" (26%).

χ^2 tests comparing the early symptoms of the Japanese and Chinese children revealed significant differences between the Japanese and Chinese cases in terms of 14 items: "retarded-like behavior", "clumsiness", "delayed overall motor development", "delayed walking with no support", "delayed control of neck", "not good at forming interpersonal relationships", "poor relations in peer group/situations", "behaves as if deaf", "reluctant to attend elementary school or kindergarten", "unusual habits or patterns", "abnormal food preferences, pica, and/or refusal to eat", "bedwetting, enuresis, or encopresis", "self-injurious behavior", "violence and aggression," and "night crying and night terrors". Every item was significantly higher in the reports by the Chinese parents.

Visits or Consultations to a Health Center or Medical Institution

Parents were asked if they had taken their children to a health center for checkup, what diagnosis had been given to their child, and whether they had been

TABLE 1 Japan-China Comparison of the Symptoms by Which Parents First Discovered Their Children's Abnormality

Symptoms	Items	Nation	N	Presence	%	P
Mental development retardation and abnormality	1. Delayed speech and other speech problems	C J	162 141	142 119	87.6 84.4	NS
	2. Retarded-like behavior	C J	162 141	56 12	34.6 8.5	***
Motor development retardation and abnormality	3. Clumsiness	C J	162 141	43 12	26.5 8.5	***
	4. Delayed overall motor development	C J	162 141	61 12	37.7 8.5	***
	5. Delayed walking with no support	C J	162 141	9 12	5.3 8.5	NS
	6. Always sleeping	C J	162 141	7 5	4.3 3.5	NS
	7. Delayed control of neck	C J	162 141	26 3	16.0 2.1	***
Behavioral abnormality	8. not good at forming interpersonal relationships	C J	162 141	91 36	56.2 25.5	***
	9. Poor response to others	C J	162 141	107 77	66.0 54.6	NS
	10. Poor relations in peer group situations	C J	162 141	94 27	58.0 19.1	***
	11. Behaves as if deaf	C J	162 141	138 45	85.2 31.9	***
	12. Mother-infant separation is bad	C J	162 141	20 10	12.3 7.1	NS
	13. Reluctant to attend elementary school or kindergarten	C J	162 141	29 3	17.9 2.1	***
	14. Not entrust the body to a person who carries it	C J	162 141	21 10	13.0 7.1	NS
	15. Restless and hyperactive	C J	162 141	82 63	50.6 44.7	NS
	16. Unusual habits or patterns	C J	162 141	45 21	27.8 14.9	**
	17. abnormal food preferences, pica behavior, and/or refusal to eat	C J	162 141	49 18	30.2 12.8	***
	18. Nervous and hypersensitive	C J	162 141	17 21	10.5 14.9	NS
	19. Bedwetting, enuresis, or encopresis	C J	162 141	18 4	11.1 2.8	**
	20. Self-injurious behavior	C J	162 141	15 1	9.3 0.7	***
	21. Violence and aggression	C J	162 141	29 4	17.9 2.8	**
	22. Night crying and night terrors	C J	162 141	38 8	23.5 5.7	***
	23. Visible nervous tic	C J	162 141	5 1	3.1 0.7	NS
Physical abnormality	24. Convulsions or spasms	C J	162 141	2 3	1.2 2.1	NS
	25. Paralysis of limbs	C J	162 141	1 0	0.6 0	NS

** $p < .01$, *** $p < .001$

referred to a care-education facility. Of the parents, 151 (93%) answered that their children underwent a medical checkup at a health center, at which time, 85 of the children (53%) were noted to have some disability or the possibility of a disability. Among those 85 children, 43 (51%) had been referred to other medical or consultation institutions, 11 (13%) were not referred to a specific alternative place, but simply told to go to some other institution, 15 (18%) were not particularly advised to go to any other institution, and 16 (19%) were told to keep track of the child's further development.

Next, when parents were asked what medical or consultation institution they took their children to for the first time after identifying their child's disability, 153 parents (96%) answered that they took their child to a hospital, 7 parents took their child to a health center, and 2 did not take their child anywhere else. The departments that they visited first were pediatrics ($N=103$, 64%), psychiatry ($N=19$, 12%), a department of neurology or internal medicine ($N=27$, 17%), and other ($N=14$). In response to the question about what diagnosis was given by the medical or consultative institutions, 93 parents (58%) answered "some disease or disorder is suspected," 28 parents (18%), "disease," 20 parents (13%), "keep track of the child's further development," and 19 parents (12%), "none particularly."

Describing the referrals to other medical or consultation institutions, 66 parents (41%) were "referred to other medical or consultation institutions," 34 (23%) were "not referred to a specific alternative place, but simply told to go to some other institution," and 60 patients (37.5%), "not particularly advised to go to any other institution." The parents visited and consulted with doctors or medical institutions a maximum of 15 times, with a mean of 4.8 times.

When the parents were asked if their child had ever been to another medical institution or facility for consultation or care-education aside from the medical institution or facility that they visit now, 112 parents (69%) answered "yes." The mean number of visits a month was 22.3, and the mean number of minutes per day of training was 263 (that is, 4 hours 23 minutes). Some of the parents ($N=38$; 34%) stayed near the child's institution of residence in order to visit with their child. It took 59 patients (53%) less than 1 hour to get to the institution where their child was residing; 10 parents (9%) reported more than one hour but less than 2 hours commuting time.

Discussion

The parents who participated in the present study complained about their child's speech or general motor development retardation, social maladjustment such as difficulty with interpersonal relationships and other abnormal behavior, as late as when their child was 3 years old (85% of parents).

The following symptoms were checked more frequently by the parents in China: delayed speech and other speech problems; behaves as if deaf; poor response to others; poor relations in peer group situations; not good at forming interpersonal

relationships; restless and hyperactive; delayed overall motor development; retarded-like behavior; and abnormal food preferences, pica, and/or refusal to eat.

Compared to the early symptoms of the Japanese children with autism, the symptoms checked by the Chinese parents coincide almost exactly with the results obtained by Ohta and Nagai (1987). These symptoms were classified by Ohta and Nagai (1987) into Group I (delayed or abnormal mental functioning), Group II (delayed or abnormal gross motor functioning), Group III A (abnormal behavior: social behavioral disorder), and Group III B (abnormal behavior: adaptive behavior disorder).

We confirmed anew that the same basic early symptoms of autism appear, irrespective of the parents' social situation. Even if the early symptoms of Chinese children with autism were not so different from the research results by Ohta and Nagai (1987) and others (Ohta & Nagai, 1992; Zhuang & Du, 2003), the Chinese results showed tendencies by which the expression of the symptoms was more intensive, and more social behavior and adaptation disorders were visible. It is suggested that these results are closely related to the child care support system. In the actual condition of the Chinese system, discovery and dealing with children with autism at an early age are not developed at all. It seems that mothers with children with autism occurring in such situations have high physical and mental stress, and that cultural factors such as parent's expectations for their child, have some impact on the results of the study.

In China, which has no national policy relating to developmental disabilities such as autism, infant health care is limited to detection of newborn congenital abnormalities, examination of height, weight, and nutritional state, immunization, and so on, and few screening tests for development disorders such as autism are conducted. Moreover, only a very few medical institutions are able to diagnose and treat autism. Although the Chinese Classification of Mental Disorders, Second Edition (CCMD-2R, Chinese Medical Association, 1995), according to ICD-10, classifies autism as a pervasive developmental disability, no doctor in China who is in charge of autism has sufficient knowledge of the child development disability area, so autism is misunderstood or incorrectly treated in many cases (Yu, Ohta, & Wu, 2007).

Additionally, no national policy exists in China for early care; education and care-education of children with autism are conducted only in 70 private care-education facilities for children with autism (as of April 2007), with problems reported such as the high cost burden and low expertise of staff (Lv, 2006; Lv & Takahashi, 2005).

In this way, the near-total absence of medical and care-educational administrative policy and an early identification system makes it difficult to diagnose autism at an early age, with the result that children with autism and their families face very harsh conditions. It is thought that parents of children with autism who are isolated feel helpless from being in such a situation. According to Lv and Takahashi (2004), a major cause of stress for mothers of Chinese children with autism is that there are

few places such as treatment facilities and administrative institutions.

Each family in China is supposed to be composed of one child and six adults, according to the one-child policy. Parents dote on their child, and, at the same time, expect their child to be very successful. That may be why the parents of the Chinese children with autism were greatly concerned about the problem of their child's social behavior and adaptation. Consequently, although the early symptoms of Chinese children with autism are similar to those of Japanese children, the disability might express itself in more various and intensive social behavior problems or adjustment disorders in China.

In addition, in the present study, the children diagnosed with autism before they were three years old accounted for 60% of the children in the sample, which was higher than expected. These results are inferred to be critically influenced by the social class of the parents of the children with autism who visit the two private care-educational facilities for children with autism (Beijing Star and Rain Educational Institute for Children with Autism and the Yilin Training Center for Children with Autism). In the present study, although the parents were not asked about their educational background or annual income, research by Lv and Takahashi (2006) at the same facilities suggests that the parents of children with autism who visit those facilities belong to socially wealthy classes, and that the mothers have very high educational backgrounds compared to the general situation of Chinese women. Because parents who belong to a wealthy class tend to be well informed about child management and think much about their child's development, these parents might detect their child's disability at an earlier age.

Even if they were to identify their child's disability and take the child to a hospital, however, the hospital would not be able to give a definitive diagnosis in many cases. Furthermore, the hospitals' responses, such as referrals to other medical and consultation institutions, were insufficient, so most of the Chinese parents had searched all over the country for doctors, spending enormous funds simply for a diagnosis.

Conclusions

From an analysis of answers to a questionnaire sent to parents of children with autism, including a comparison of the obtained data with those on early symptoms of Japanese children with autism, the present study has revealed some of the characteristics of the early symptoms of children with autism in China.

Significant differences were found between the Japanese and Chinese cases, especially on 14 items: "retarded-like behavior", "clumsiness", "delayed overall motor development", "delayed walking with no support", "delayed control of neck", "not good at forming interpersonal relationships", "poor relationships in peer group situations", "behaves as if deaf", "reluctant to attend elementary school or kindergarten", "unusual habits or patterns", "abnormal food preferences, pica and/or refusal to eat", "bedwetting, enuresis, or encopresis", "self-injurious behavior", "violence