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Case Report

Parallel Lives: A Case Series of Three Boys with Persistent Reactive Attachment Disorder

Nelson R[1*], Chadwick G[1], Bruce M[2] and Minnis H[1]

[1]University of Glasgow, Glasgow, UK

[2]NHS Lothian, Lothian, Scotland

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1. Abstract

Reactive Attachment Disorder (RAD), only diagnosed in the context of early abuse and neglect, is characterised by failure to seek and accept comfort. It involves lack of activation of the - developmentally essential - attachment system, hence has profound developmental disadvantages. RAD usually resolves quickly in the context of adequate care and has been assumed never to persist once the child is in a nurturing placement. We challenge this existing paradigm by presenting three cases of children whose RAD symptoms *have* persisted despite living in placements judged, by both social and child health services, to be of good quality. All three boys met DSM 5 criteria for RAD in late childhood/early adolescence and had had stable RAD symptoms since before age 5. In the absence of longitudinal data, except from unusual institutionalised samples, it has been impossible to evidence RAD beyond pre-school and virtually nothing is known about factors predicting its stability. This case series and systematic review provides the first opportunity to generate testable hypotheses about environmental circumstances and coexisting symptomatology that may influence RAD trajectories. As predicted more than a decade ago, persistence of RAD has had profoundly negative developmental implications for the children and an extremely detrimental effect on family life and relationships. Recognition of RAD symptoms is challenging because symptoms are classically internalising and therefore easy to miss. This case series will allow paediatricians to better recognise the subtle symptoms of RAD in order to improve their care of these children and their families.

2. Abbreviations

ADHD - Attention-Deficit/Hyperactivity Disorder

ASD - Autism Spectrum Disorder

CAMHS - Child and Adolescent Mental Health Services

DSED - Disinhibited Social Engagement Disorder

DSM-V - Diagnostic and Statistical Manual version 5

PTSD - Post-traumatic Stress Disorder

RAD - Reactive Attachment Disorder

WASI - Weschler Abbreviated Scale of Intelligence

WISC - Weschler Intelligence Scale for Children

WPPSI - Weschler Pre-school and Primary Scale of Intelligence

3. Introduction

Reactive Attachment Disorder (RAD) represents a closing down of the attachment system [1-3], therefore is associated with profound developmental disadvantages [1, 5, 6]. Because of its rarity, large scale longitudinal studies are impossible and persistence beyond early childhood has never been proven out-with institutionalised populations [7].

For rare diseases, the case series can provide the most robust methodology available [8]. We present three cases, following CARE guidelines [8], all of whom experienced severe maltreatment in early life and were then placed, between age three and five, with adoptive or foster parents who had undergone stringent background checks and had ongoing support. All boys and their families gave informed consent to the inclusion of their case reports. Names have been changed to protect identity.

***Corresponding Author (s):** Rebecca Nelson, ACE Centre, Academic CAMHS, Level 4 West Ambulatory Care Hospital, Glasgow, G3 8SJ, University of Glasgow, UK, E-mail: Rebecca.nelson@glasgow.ac.uk*

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4. Presentation, Diagnosis and Outcome

Diagnoses of RAD were made via multi-disciplinary assessment using a standardised observation protocol [9] and semi-structured interview for RAD [6, 10] and face to face clinical assessment (see **Table 1**). Additional standardised measures examined co-occurring psychiatric disorders [11] and cognition [12]. The Table provides an overview of the DSM-V criteria for RAD, cognitive functioning, co-occurring symptomatology and foster/adoptive history.

Table 1 : A case series of three boys with persistence of Reactive Attachment Disorder past the infant years and a systematic literature review.

Child	**DSM V Criteria for Reactive Attachment Disorder** Below are symptoms meeting Criteria A to D, at intial RDA Diagnosis. Criteria E (criteria not met for autism spectrum disorder); F (distrubance evident before age 5 years), G (child has developmental age of at least 9 months) and "persistence" criteria (symptoms present for more than 12 months) were met in all 3 cases.						**Foster/adoptive history**	**Co-ocuring Symptomatology**	**Cognitive functioning**
	A1 minimally seeks comfort when distressed	A2 minimally responds to comfort when distressed	B1 social/emotinal responsiveness	B2 Limited positive affect	B3 Episodes of unexplained irritability, sadness, or fearfulness	C-D the child has experienced a pattern of extremes of insufficient care presumed responsible for symptoms			
John	No comfort seeking e.g. failure to draw parents attention to severe tooth decay	No warmth or feedback when comfort offered	Usually sits alone at home looking tense and withdrawn Poor eye contact Abnormal reunion behaviour: e.g. would run up to his adoptive mother after school and stop just short of her	Severe anhedonia: Parents described him as having a "default setting' of misery"	Hypervigilance - constantly scans the environment in new places Frequent bouts of exterme upset	Exposed to extreme violence and neglect from age 0-4 years Found wandering outside alone aged 20 months Traumatic removal into foster care age 4 after police raid	Foster family 1: Age 4-5.5 years Foster family 2: 5.5 years present (adopted by same family aged 8) Near adoption breakdown age 11 - placed in foster care for two months then retruned to adoptive family	Past: Neonatal abstinence syndrome At assessment: Callous-unemotional traits; PSTD; Conduct Disorder Skin-picking Toileting problems Aggressive/bullying behaviour at school Gorging Poor concentration	WPPS Age of testing: 9 Full scale IQ = 109 Verbal Comprehension = 110 Perceptual Reasoning = 115 Processing Speed = 100 Working memory Index = 97
Brain	Does not seek comfort	Does not acceptthelp with tasks such as homework	His severe emotional withdrawal precipitated his intial referral to CAMHS at age 9 and refused to attend Poor eye contact	Seemed, to parents, to be anxious much of the time sudden outbursts o0f aggression and violence	Described by parents as "watchful and wary"	Concerns aboutneglect and domestic violence noted since 11 montyhs severe physical abuse and neglect	Briefly placed with Grandmother age 1 year Two foster placements from age 2 years Second foster placement became current adoptive from 3 years old	Past: Gorging Toileting problems until age 11 At assessment: Poor concentration and inpulsivity but not overactive Violence towards adoptive parents and destruction of the home	WASI Age of testing: 12 Full Scale IQ = 101 Verbal IQ = 90 Performance IQ = 108
Andrew	Does not seek comfort	Comfort not offered as never sought	Displays "no emotion" and e.g. laughed when close family member died Paid little attention tp foster family: adopted a 'loner' role	Fearful (afraid of bubble baths, the theatre and was terrified of soft play areas)	Frozen watchfulness - described by CAMHS worker at age 11 as being "like a rabbit in the headlights"	Did not receive any prenatal care Chronic neglect of toileting, diet, safety and health appointments	Placed in foster care as an "emergency placement" age 4 then not allowed home once social workers realised extent of neglect Now on " permanence order" ensuring care, until 18 years, with the same family he was placed with as an emergencyy	Past: Premature birth, mild hemiplegia Toileting problems Indiscriminate behaviors At assessment: Outbrusts of anger Adult "friends" online Lack of remorse Controlling and dominant with other children	WISC Age of testing: 15 Full scale = non-interpretable: high variance between domains Verbal comprehension = 89 Perceptual reasoning = 61 Processing speed = 68 Working memory = 97

Case 1, John

When assessed at age 9, RAD symptoms were noted since age 4. In addition, John was also diagnosed with Post-traumatic stress disorder (PTSD), Conduct Disorder and Attention deficit/Hyperactivity Disorder (ADHD). He had intensive relationship-focused psychotherapeutic treatment, with his adoptive mother, but symptoms persisted.

Case 2, Brian

When assessed at age 12, RAD symptoms were noted since age 3.At age thirteen, Brian was moved to a small residential unit where he continued to have problems seeking and accepting comfort from his keyworkers and his parents, who continued to visit. At age fifteen he continued to demonstrate RAD symptoms and difficult behaviour and was moved into a residential placement where he was the only child, and received intensive one to one care from a small team of staff. Involvement with his parents increased resulting in leading to weekend home passes. By age sixteen, when last assessed, Brian was no longer violent, no longer met criteria for RAD and was being investigated for possible ADHD.

Case 3, Andrew

When assessed at age 14, RAD symptoms were noted since age 4 years and the foster placement was in jeopardy. Ongoing health concerns relating to his premature birth included mild left-sided hemiplegia, talipes and partial sightedness. At first assessment, his foster carer had never considered whether or how Andrew had sought comfort as a young child, but described how he would stand silently by while physical care was provided to other foster children, all of whom had severe or profound physical and learning disabilities.

After two sessions of psychotherapy in which the therapist encouraged both Andrew and his foster mother to notice and respond to each other's signals, symptoms improved markedly.

None of the boys had symptoms of Autism Spectrum Disorder: during assessment, social communication was typical apart from emotional withdrawal and none had repetitive or stereotyped interests.

5. Family Perspective

Parents took part in qualitative interviews to explore the impact of RAD symptoms on the family. Transcripts were read independently by RN and GC and three common themes were extracted across families [13].

Family strain

High levels of stress characterised all aspects of family life. Burdensome child behaviours included lack of understanding of social cues, violence and eliciting negative attention. There was emotional separation between the child and the rest of the family. This was described, by John's mother, as living "parallel lives; one life with my husband, biological son and dog, and the other with John."

Frustration

A key frustration for families was lack of understanding of the child's emotions and the child's emotional unresponsiveness and abnormal interactions. Another frustration was the significant delay in identification of the problem and the associated lack of support, resulting in years of isolated suffering.

Resentment: Feelings of resentment were evident in all three families due to the significant strain that RAD had put on their family and their relationships. Both adoptive couples had experienced marital difficulties ascribed to the burden of their child's problems.

6. Discussion

Persistent RAD is rare: only a handful of cases were diagnosed by HM in over 20 years of clinical practice. All had associated developmental/neurodevelopmental problems, as previously described [5, 6, 14, 15]. All placements had been threatened with breakdown. John has persistent symptoms despite treatment, Andrew had rapid symptom resolution after psychotherapy in adolescence and Brian had symptom resolution after intensive one-to-one residential support. This provides the new insights that persistent RAD, while rare, threatens family life - but treatment is possible, even in adolescence.

Our systematic literature review found individual and contextual factors to be associated with RAD. Contextual factors include institutionalization [16], quality of care giving in the institution [16-19], harsh parenting, parental negativity [20], parental mental health problems [21, 22] and longer exposure to the maltreating pre-care environment [23]. Individual factors include male gender [22], reduced grey matter volume [24], lower cognitive ability [25], dopaminergic dysfunction [18, 26]; and genetic factors, particularly for males [21]. RAD is associated with depressive symptoms [18], social and emotional difficulties [18, 27], functional impairment [17, 18, 22], behavioural and conduct problems [22, 27, 28], hyperactivity [20, 28]; internalising symptoms [17, 28, 29] , externalising symptoms [28], stereotypies [17], help seeking from services [22], callous-unemotional traits [27] and symptoms of Disinhibited Social Engagement Disorder (DSED) [17, 18, 22, 27]. Studies were of moderate to high quality, but findings were not always consistent across studies, possibly due to a lack of consistent measures for RAD, confounding, differing sample types and modest sample sizes.

To conclude, in rare cases RAD can persist despite years of nurturing care. Symptoms are easy to miss but are associated with severe family stress and other developmental problems. Paediatricians should always consider RAD when assessing children with a maltreatment history.

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