Atypical trajectories of number development: a neuroconstructivist perspective

Daniel Ansari and Annette Karmiloff-Smith

Despite the fact that number deficits are as prevalent as literacy deficits, research on basic numerical skills lags seriously behind the successful studies identifying low-level deficits in dyslexia. We review current debates on number, discussing how the competing theories pertain to mathematical disabilities in normal children and numeracy deficits in genetic disorders. We stress the need to consider these issues within the framework of a developing system rather than from the neuropsychological perspective of focal damage. The earlier the exploration of atypical trajectories in very basic numerical skills, the better we will be able to chart their developmental impact on subsequent, higher-level arithmetic abilities.

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Problems with number come in a variety of forms. Sometimes children with otherwise normal intelligence show particular problems with arithmetic. This is usually called 'mathematical disability' or 'dyscalculia' and contrasts with number problems in children with genetic disorders where number is impaired alongside low IQ and other cognitive problems. We will call the latter 'numeracy deficits'. Despite much progress on normal number development, dyscalculia and numeracy deficits turn out to be surprisingly neglected areas of the cognitive sciences, despite their importance in schooling, everyday life, and employment [1]. Paradoxically, although current estimates suggest that dyscalculia is more prevalent than dyslexia [2–5], and frequently co-occurs with the latter, it is only the field of reading impairment that has witnessed major advances in identifying the processes underlying the deficit [6,7]. Like dyslexia, dyscalculia can be found as a relatively isolated problem in school children whose other abilities fall within the normal range. But recent research indicates that subgroups of children suffering from both dyscalculia and dyslexia versus those with seemingly pure dyscalculia differ in significant ways (see Box 1) [4].

Numeracy deficits are very prevalent in genetic disorders. They have been reported, although not thoroughly investigated, in a variety of syndromes such as Velocardiofacial syndrome [8], Turner syndrome [9,10], Fragile X syndrome [11], Down's syndrome [12], and Williams syndrome [13]. Number scores are often worse than reading scores in clinical groups, suggesting that number is a particularly vulnerable cognitive domain in the atypically developing brain. In this review, we examine current theoretical frameworks used to explain normal number abilities and how these might pertain to dyscalculia in normal children and numeracy deficits in disorders of known genetic origin. We believe that, rather than using the neuropsychological perspective of focal brain damage in adults, it is vital to take a truly developmental approach to dyscalculia and numeracy deficits. In particular, we argue for the need to understand very basic processes underlying numerically-relevant computations and how these can go awry very early in development.

Numerical cognition in the normal brain

Although the cognitive neurosciences have neglected the study of number in cognitively impaired children, the study of numerical cognition in the normal brain has greatly advanced [14,15]. Evidence for the existence of phylogenetic and ontogenetic continuity in a very basic system of numerical skills has been extensively discussed [14–18]. Furthermore, since the advent of functional neuroimaging, researchers have started to characterize the cerebral circuits involved in the representation and processing of numerical stimuli [19,20]. One theory claims that number is an innately specified module, dedicated to the processing of quantity and composed of abilities such as detecting changes in numerosity and ordering amounts by size [15]. Recent findings from brain-imaging and behavioral studies have led to another influential claim that the fundamental organization of numerical cognition is based on two systems of representation: an approximate, analog, language-independent system, and an exact, language- and culture-dependent system [20]. In the analog representation the variability of the signal is proportional to the size of the represented magnitude. By contrast, the exact system is discrete and represented by the integer-list representation of number in natural languages. These two systems are claimed to recruit separate circuits in normal brains and can be differentially damaged in adult patients [21].

The developmental foundations of numerical cognition

A neuroconstructivist perspective on disorders of numerical cognition necessitates the study of the foundations of numerical cognition and how numerically-relevant representations are structured and change over developmental time. We review the

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Box 1. How specific is dyscalculia?

In the DSM-IV [a] diagnostic criteria and through much of the scientific literature on the topic, dyscalculia is frequently defined as an isolated problem due to number-specific underlying deficits. There are, however, reports of a high degree of co-occurrence of dyscalculia and dyslexia [b,c]. The co-morbidity of impairments in both domains suggests at the very least that dyscalculia is not specific in all cases. Moreover, recent research indicates that subgroups of children suffering from both dyscalculia and dyslexia versus those with seemingly pure dyscalculia differ in significant ways [d,e]. Such findings obviously cast doubt on investigations in which children diagnosed with dyscalculia are treated as a homogenous group. In addition, a large-scale study demonstrated that not only do 17% of children with dyscalculia also have dyslexia, but that an additional 26% of such children suffer concurrently from Attention Deficit Hyperactivity Disorder [f]. If there are indeed children with specific impairments in numerical cognition, then future research should separate these children from groups who also suffer from other forms of learning disability.

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numerical competence of typically developing infants and young children and show how the study of such competencies in children with dyscalculia and numeracy deficits might elucidate these conditions.

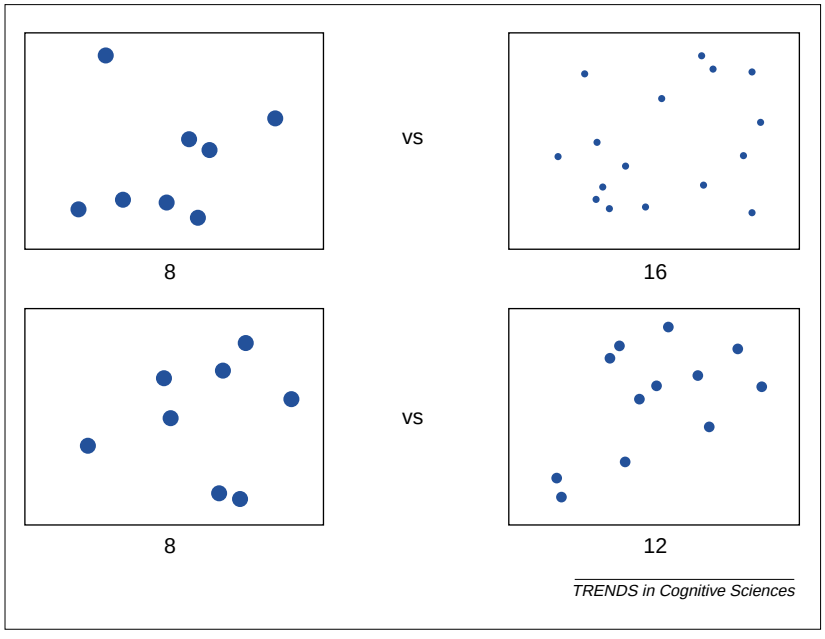

Fig. 1. Representation of numerosity in infants. Six-month old infants were habituated to a display of dots, either 8 or 16 dots (a), or 8 or 12 dots (b). To ensure that infants were discriminating numerosity, the stimuli were designed such that variables continuous with numerosity were controlled for, including density, area occupied by the dots and the spatial extent of the displays. In addition, the size of the dots varied between the habituation displays for each numerosity to ensure that the average size of the dots was equated between numerosities. During the test phase they were presented with both the numerosity to which they had been habituated and the novel numerosity. The results showed that infants looked significantly longer at novel displays in the 8 vs. 16 dots condition but not in the 8 vs. 12 dots condition. Hence, six-month olds showed no ability to discriminate between 8 and 12 dots, but could discriminate 8 vs. 16. These findings were the first to show that infants can discriminate between large numerosities. Moreover these results show that infants' number discrimination abilities are related to the relative size (ratio) of the numerosities. (Adapted from Ref. 31.)

Several experiments have revealed that young infants [22,23], and even newborns [24], can discriminate between 2 versus 3 dots or objects. Furthermore, it has been shown that 5-month olds are able to track simple numerical transformations of object arrays, such as addition and subtraction [25]. Although infants can make such discriminations with respect to small numbers, they fail at numerosities greater than 3. Thus, they are not successful in discriminating between 4 versus 6, despite their ability to differentiate 2 versus 3. Such findings have led to the contention that, rather than relying on an analog representation, infants are 'subitizing'. This well-known phenomenon is argued to be a visual process [26,27], based on findings that adults can make fast judgments (without enumeration) about quantities that range from 1–4, a process that starts to break down as sets get larger. Note, however, that this explanation only holds for infant's ability to discriminate numerosities presented in parallel in the visual modality and cannot explain their ability to enumerate sequentially presented puppet jumps [28] and discriminate between numerosities in the auditory domain [29]. In addition, whereas subitizing offers an explanation of why infants fail at numerosities larger than 3, it does not characterize the actual structure of small number representations.

The 'Object-File' model of infant numerical representation

How do infants represent small numerosities? A recent model suggests that they represent each of the objects in a set by a separate (non-numerical) symbol. Each additional object is represented by the opening of a separate file in the brain's representational system. This account is known as the *Object-File* theory [30–32]. Representation by object files cannot exceed 3 or 4 objects at a time. Evidence consistent with the object-file model, has shown that infants base their choice of quantity on total surface area rather than number, and that their performance breaks down with larger numbers even if the ratio difference between numerosities is held constant [33]. Despite these non-numerical choices, the representation is still numerical because there is one file per object and numerical equivalence can be established through one-to-one correspondence between object files. In this framework, the ontogenetic pathway to number is via object-based attention. Object files provide the representational basis from which infants develop the ability to disentangle perceptual variables such as surface area from numerosity. The opening of new object-files would lend itself directly to a mapping onto the subsequent process of constructing a symbolic integer-list representation of natural numbers, with discrete intervals and a successor function (+1). As the opening of new object files is analogous to the successor function, it might serve as a bootstrapping device for the development of discrete, symbolic

Box 2. The neural basis for mathematical disability: evidence from children born preterm

Brain damaged adults with dyscalculia have been found to have lesions in the inferior parietal lobes [a]. In particular, the left angular gyrus has been related to calculation [b]. Is this region of the brain also affected in children who develop problems with calculation?

Difficulties with numeracy are frequent in children born preterm, despite intelligence and reading scores in the normal range [c]. In a recent study using voxel-based morphometry [d], it was found that children with dyscalculia born preterm have less grey matter in an area of the left parietal lobe than those without number difficulties (see Fig. I), despite being matched on all other variables (gestational age, birthweight, etc.).

Similar investigations in non-preterm children with mathematical disability are worth undertaking, differentiating those who have concurrent dyslexia and those who do not. Structural anomalies of this kind are likely to be present from birth and to subsequently constrain the acquisition of basic number skills, with downstream effects on the development of higher-level skills, such as arithmetic. Note, that gray matter abnormalities in the inferior parietal lobes have also been shown in children with Velocardiofacial syndrome [e].

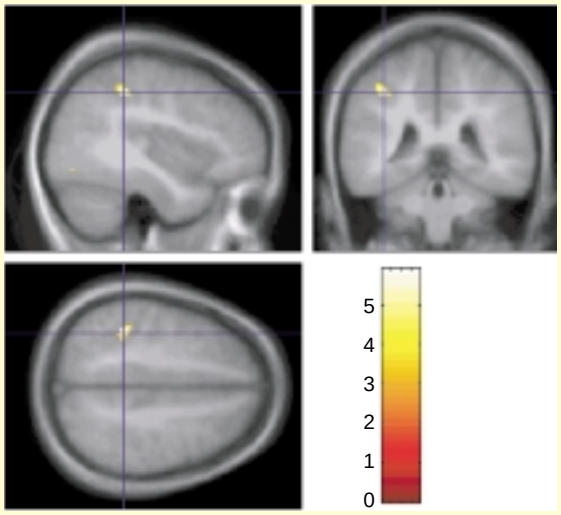

Fig I. Statistical parametric maps showing a region of grey matter in the left inferior parietal lobe that is significantly reduced in size in children with very low birthweight who have calculation difficulties. Such children obtained scores on a standardized test of calculation ability significantly below the scores predicted by their overall IQ, relative to children also born preterm but without a calculation deficit (children whose calculation ability scores were consistent with their IQ).

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number representation. It is for this reason that object files have been considered a more likely ontogenetic basis for arithmetic than the analog system of number representation.

The analog model of infant number representation

Recent evidence challenges the object-file view. Indeed, by 6 months infants can discriminate 8 versus 16 dots [34], but interestingly, they fail at 8 versus 12 dots (Fig. 1). These findings are consistent with the analog model of number representation which postulates that discriminability decreases as the ratio between numerosities increases. Variables of the stimuli continuous with numerosity (e.g. density, surface area etc.) were carefully controlled, a caution that had not been in place in earlier studies of infant numerosity [35,36]. The demonstration of the infant capacity to distinguish 8 from16 dots is difficult to reconcile with subitizing or with the sole availability of a limited object-file system of representation. Furthermore, recent data [37] show that 11-month olds, but not 9-month olds, appreciate the ordinal relationship between 4, 8 and 16 dots. This provides further evidence for infants' ability to represent numerosities greater than 4. This finding also highlights the importance of taking a developmental perspective on infants' numerical representations and the ontogenetic changes within them. Together with the evidence for phylogenetic continuity, the evidence currently lends support to the argument that it is the analog representation of number that forms the basis of subsequent integer processing and exact calculation [17,18]. But it does not explain the early set size limitation found in most studies. It is thus possible that both forms of representation play a role in early development [16,31].

From typical to atypical number development

The commonalties between the models reviewed above have crucial implications for the study of dyscalculia in normal children and of numeracy deficits in genetic disorders. Both the object-file and analog systems of representation are fundamental representational precursors to number development. Subtle impairments in these foundational, numerical representations and the integration of differentially impaired systems of numerosity representation could have cascading downstream effects over developmental time on higher-level numerical competencies like addition and multiplication [38].

Dyscalculia in otherwise normal children

Dyscalculia is considered to exist when an individual's scores on standardized tests of mathematical ability are substantially below that expected given their chronological age, measured intelligence and age-appropriate education [39]. As mentioned, it is rather prevalent in school-aged children, and turns out to be a particular problem for children born preterm (see Box 2). Yet, the contrast

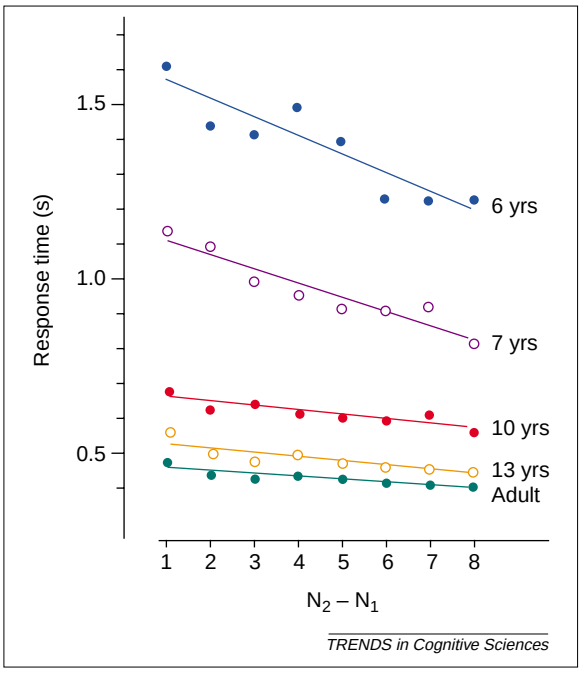

Fig. 2. Mean response time to decide which of two digits is numerically larger, plotted as the difference between two numbers, N_2–N_1. Data is shown for children of four different ages, and adults, and illustrates the robust effect of numerical magnitude on reaction times (the bigger the numerical difference the faster individuals decide which is the greater number). Note that the effect of distance decreases over developmental time, suggesting that the features of magnitude representations become less subjectively different from one another and thus more discriminable.

between the flourishing research on the normal development of number representation and the meager literature on dyscalculia is striking.

For the normal case, the cognitive neuroscience approach has shifted from culturally-dependent aspects of numerical cognition, such as arithmetic, to an analysis of its representational primitives. By contrast, the vast majority of studies of dyscalculia remain focused on higher-level, school-like concepts such as addition and multiplication. Analyses of the disability have been inspired by adult neuropsychology [40], with few questions raised as to the developmental trajectory of the disorder. Where research has turned to lower-level processes, it has tended to focus on more domain-general cognitive competencies that affect all tasks including numerical ones. These general problems include immature development of problem-solving strategies [41], poor working memory span leading to computational errors [42], deficits in the long-term retrieval of arithmetic facts [43,44], slow speed of processing [45] as well as disturbances of visuo-spatial functioning [46]. All of these have been found to correlate with dyscalculia. Yet, such domain-general deficits are unlikely to impact on numerical processing alone. There is thus an urgent need to explore low-level, number-relevant causes and how these may go awry over developmental time.

Adult models of acquired dyscalculia have also been used to account for findings from single case studies of children with developmental dyscalculia [47–49]. Double dissociations have been claimed between different domains of numerical processing, such as number processing and calculation abilities [50–52]. Although the comparison between developmental and acquired dyscalculia focuses on impairments specific to the domain of number rather than merely domain-general competencies, it ignores the complex developmental trajectory from the infant starting state to the adult endstate [38,53]. Such studies simply assume that data from adult neuropsychological patients can be taken as evidence for how the brain of the normal child starts out developing number processing systems.

Methodological considerations in dyscalculia research

From an empirical perspective, the tasks that are used to diagnose selective deficits in developmental dyscalculia are often directly derived from test batteries designed for individuals with brain lesions [49]. These batteries use a pencil and paper approach and cannot yield an in-depth analysis of underlying processing. A different and, we consider, more fruitful approach derives from psychophysical paradigms that systematically manipulate the numerical stimuli and measure both reaction time and accuracy. Substantial progress has been made in describing and investigating the fundamental, psychophysical properties of normal number representation. For example, the smaller the numerical difference between two numbers, the longer it takes to judge which of them is the larger (distance effect). Furthermore, reaction time is positively related to the relative size of magnitudes (size effect). These effects have been found in infants [34], children [54,55], adults [56], and animals [17]. Figure 2 illustrates the ontogenetic continuity of the distance effect: the effect is shown to be present in both children and adults, progressively decreasing with age.

The study of atypical number development requires an account of how subtle impairments in basic number representations and their developmental trajectory lead to the formation of deviant end-state representational systems [38,57]. Qualitative and quantitative differences in the distance and size effects may predict and explain disorders of higher-level numerical processing.

The key issue here is to separate the actual processes of calculation from the resultant product of calculation. Only with more sophisticated experimental approaches will it be possible to detect differences in the way in which children represent and process numerical information as well as the effect this may have on their subsequent performance on tests of arithmetic. Even when evidence shows that children with dyscalculia suffer from arithmetic fact retrieval difficulties, this does not preclude the possibility that impoverished number representations actually contribute to the retrieval problems. Indeed, recent studies have shown that arithmetic fact retrieval is not a straightforward retrieval process but involves the activation of numerical magnitudes [58].

Box 3. Numerosity discrimination in atypically developing infants

How does the development of number go awry in children with genetic disorders? In one experiment, based on well-established techniques used with normal infants [a,b], a population of 65 infants was divided into four groups [c]. Two groups were clinical, Williams syndrome [d] and Down's syndrome [e], who were matched on both chronological (CA) and mental age (MA). Two were normal controls, one matched on MA to control for general level of intelligence, and the other matched on CA to control for length of experience. Infants were familiarized with pairs of stimuli containing arrays of changing sets of two objects in different configurations, colours and sizes. After familiarization to the pairs, they were tested on a pair with one stimulus containing two new objects, and the other containing three objects, a novel numerosity. If infants were merely sensitive to changes in the types of object, they would show no preferential looking in the test phase when new objects in new positions were displayed but numerosity changed. However, if during familiarization they had become sensitive to the constant numerosity (two) of each of the displays, they should look significantly longer when, during the test phase, one of the pair displayed a novel numerosity. The looking-time results for the four groups as a function of familiar versus novel numerosity (Fig. I) showed that infants with Williams syndrome behaved like both MA and CA controls in their sensitivity to novel numerosities. By contrast, infants with DS looked equally long at both test displays, pointing to a significant impairment in their early numerosity discrimination.

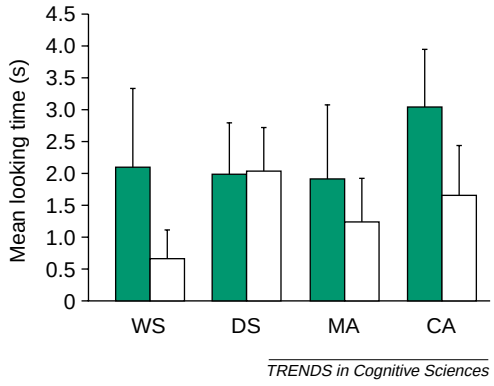

Fig I. Infants with Williams (WS) and Down's syndrome (DS) as well as controls matched for mental age (MA) and chronological age (CA) were tested for their ability to discriminate between 2 and 3 dots. During familiarization infants were presented with one of the two numerosities until they looked away for a specified amount of time. Subsequently infants were presented with alternating displays of both numerosities (familiar, green bars; novel, white bars) and their looking time was measured. Infants with WS, and MA and CA controls looked significantly longer at the numerosity they had not previously seen, but infants with DS did not.

Interestingly, the WS proficiency with small numbers in infancy co-occurs with the fact that adults with WS show more serious impairments on a wide variety of number tasks than adults with DS [f]. If development were linear, the pattern in the adult end state would have predicted that infants with WS would be as impaired as (or even more impaired than) infants with DS. In fact, the opposite obtains. So, in the case where a low-level capacity such as the object-file system seems unimpaired, other interacting systems, such as a defective analog system, lead to impairments of number in the adult end state. Thus, disorders of numerical cognition in the end state in different syndromes might arise from impairments to different low-level components of the numerical system. Such an approach stresses the need to follow the entire atypical pathways between the initial infant state and adult end state.

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Numeracy deficits in disorders with known genetic bases

Children and adults with a variety of genetic disorders suffer from numeracy deficits [8–13]. However, the volume of papers that has addressed this question both empirically and theoretically turns out to be even sparser than that for dyscalculia.

From the few studies that have been conducted on children with genetic disorders, the approach has not differed substantially from that used in the study of dyscalculia, focusing on higher-level arithmetic skills.

Little is known about whether the roots of numeracy deficits in genetic disorders lie in impairments to basic number-relevant processes or to impaired domain-general processes. In our view, the time is ripe for considering numeracy deficits in developmental disorders via a more basic approach, in the hope of mirroring the increasingly successful identification of the underlying causes of dyslexia. In the latter domain, the discovery early in life of low-level timing and phonological deficits in spoken language have been shown to impact on later written language development [6,7]. For number, differences in the way in which pre-verbal infants discriminate quantities or later process and compare magnitudes, may hold the key to identifying the basic precursors of numeracy deficits in developmental disorders (see Box 3).

Outstanding questions

- Do analog and symbolic systems of number develop independently or become progressively integrated or separated over developmental time?
- What developmental changes explain why young children do not have a full grasp of the meaning of counting, despite the surprising numerical abilities of preverbal infants?
- What factors explain the co-morbidity of dyslexia and dyscalculia? Are common brain/cognitive systems affected, are both particularly vulnerable to atypical development or is number even more at risk than reading?
- In children with dyscalculia, are their other abilities really normal or can we detect subtle representational impairments?
- Which aspects of numeracy deficits in genetic disorders are due to syndrome-specific causes and which to syndrome-general impairments?

Concluding comments

The wealth of knowledge pertaining to the phylogenetic and ontogenetic bases of numerical cognition in the normal brain has yet to be fully exploited to gain insights into the neurocognitive bases of dyscalculia in normal children and numeracy deficits in children with disorders of known genetic origin. We argue that to understand impairments of numerical cognition in general, it is necessary to investigate whether differences in very basic number representations underlie these conditions. Instead of recourse to adult models of numerical cognition, it is crucial to understand the atypical developmental trajectory from subtle deviations in the systems of numerical cognition that represent the ontogenetic foundations of number representation through progressively to impairments in arithmetic. This approach, we predict, will enable us to delineate predictors of numerical problems and hence diagnose, study and remediate these conditions much earlier.

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