Rapid onset functional tic-like behaviors in young females during the COVID-19 pandemic

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This article has been accepted for publication and undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process which may lead to differences between this version and the Version of Record. Please cite this article as doi: 10.1002/mds.28778

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Running title: Rapid onset functional tic-like behaviours

Keywords: tics, Tourette syndrome, functional movement disorders, COVID-19 pandemic

Financial disclosure related to research covered in this article: The authors of this article have no conflicts of interest to report in relation to research covered in this article.

Funding sources for study: None
Background

Since the beginning of the COVID-19 pandemic, our group of colleagues working at eight different Tourette syndrome clinics from across the world, have all witnessed a parallel pandemic of young people 12 to 25 years of age (almost exclusively female) presenting with the rapid onset of complex motor and vocal tic-like behaviors (1). In most cases, these behavioral patterns are consistent with a functional neurological disorder. There have been striking commonalities in the phenomenology of these tic-like behaviours seen across our centres in Canada, the United States, the United Kingdom, Germany and Australia. Our goal with this Viewpoint is to help clinicians recognize patients with this disorder and distinguish them from cases of Tourette syndrome (TS). We begin by describing the clinical phenomenology and demographic characteristics of youth with rapid onset functional tic-like behaviours using illustrative data from the Tic Disorders Clinical Registry at the Calgary Tourette and Pediatric Movement Disorders Clinic. We then discuss our shared experiences across our eight centres and provide preliminary viewpoints on the pathophysiology and treatment of this complex disorder.

The Calgary Tic Disorders Clinical Registry

This registry enrols participants at their first clinic visit into a prospective cohort study assessing long-term outcomes in youth with tics. The registry is approved by the Calgary Health Research Ethics Board and all participants provide informed consent. Baseline data elements includes age, sex, age at tic onset, current medication use, tic disorder diagnosis, Yale Global Tic Severity Scale (YGTSS) score, presence of comorbid Attention Deficit Hyperactivity Disorder (ADHD) and symptom severity on the Conners 3, Obsessive Compulsive Disorder (OCD) and symptom severity on the Children’s Yale Brown Obsessive Compulsive Scale (CYBOCS), Anxiety Disorder (including Generalized Anxiety Disorder,
Social Anxiety Disorder, or Panic Disorder) and symptom severity on the Multidimensional Anxiety Scale for Children version 2 (MASC2), Major Depressive Disorder and symptom severity on the Child Depression Inventory version 2 (CDI2), and Autism. Possible tic disorder diagnoses recorded in the registry included TS, Persistent Motor Tic Disorder (PMTD), Persistent Vocal Tic Disorder (PVTD), or Provisional Tic Disorder (PTD). The diagnosis of functional tic-like behaviours was added to the registry in 2020. Using this registry data, we contrasted clinical features present at the first clinical visit of participants diagnosed with primary tic disorders and those diagnosed with functional tic-like behaviours. Children categorized with primary tic disorders met DSM-V criteria for TS, PMTD, PVTD, or PTD. Children categorized with functional tic-like behaviours had the rapid onset of complex tic-like behaviours with escalation to peak severity within hours to days. All diagnoses were made by movement disorders specialists with expertise in tic disorders. Continuous variables were compared between groups using a two-sample t-test, and categorical variables were compared using the chi-squared test.

Data from 290 registry participants collected between 2012 and June 30th, 2021 were analysed, including 270 with a primary tic disorder (215 TS, 28 PMTD, 4 PVTD and 23 PTD), and 20 with functional tic-like behaviours. Of the 20 patients with functional tic-like behaviours, 17 had no history of previous tics, while 3 had mild simple tics earlier in childhood which never came to clinical attention. Rapid onset of functional tic-like behaviours occurred in all participants during the pandemic period (after March 1, 2020), and all endorsed exposure to influencers on social media (mainly TikTok) with tics or TS. With respect to the phenomenology of tic-like behaviours, 18 of 20 had complex vocalizations consisting of the repetition of random words or phrases (e.g. knock knock, woo hoo, beans) 11 of 20 engaged in the repetition of curse words, or obscene, offensive or derogatory
statements, 13 of 20 had complex arm/hand movements (clapping, pointing, sign language, or throwing objects) and 14 of 20 had complex behaviours in which they would hit or bang part of their body, other people (typically parents) or objects.

< Insert Table 1 here >

Table 1 summarizes the demographic and clinical features of registry participants. Participants with functional tic-like behaviours were more likely to be female, were older at first visit, were older at symptom onset, had higher YGTSS total tic and impairment scores, were more likely to have an anxiety disorder or major depressive disorder diagnosis, and had significantly higher total symptom scores on the MASC2 and CDI2 (all p<0.0001). Logistic regression controlling for age and sex demonstrated a significant association between the diagnosis of functional tic-like behaviours and the diagnosis of an anxiety disorder (OR 4.42, 95% CI 1.22, 16.00, p=0.02), or major depressive disorder (OR 4.92, 95% CI 1.29, 18.83, p=0.02). Linear regression controlling for age and sex demonstrated a significant relationship between the diagnosis of functional tic-like behaviours and total tic severity on the YGTSS, with a coefficient of 10.60, 95% CI 5.89, 15.30, p<0.0001.

**Viewpoint**

While functional tic-like behaviours have certainly been described by others in the past (2-4), until now these cases have represented a small fraction of referrals to TS/tic disorder clinics(2, 3, 5). Table 2 provides estimates on the percentage of new referrals for which functional tics were the primary problem both before the pandemic, and in the first half of 2021, and the average annual number of referrals for tics or movement disorders, across five of our centres. Although after the pandemic started referral volumes increased in three
centres, remained the same in one centre and decreased in one centre, all centres experienced a dramatic increase in the proportion of referrals for functional tic like behaviours. While in the past we have all managed children with TS with functional tics in addition to tics related to TS, and see a small number of functional tic cases each year as the primary diagnosis, it is the unprecedented increase in new referrals of young females with the rapid onset of tic-like behaviours since the pandemic started that has been so unusual. This has allowed us to make new observations and gather insights into this specific presentation. Many of these rapid onset cases have no definite history of previous tics. They experience the rapid onset of complex tic-like behaviors which escalate in frequency and severity over a period of hours to days, prompting emergency department visits, and even hospital admission. Their presentation is notable for complex motor tic-like behaviors and vocalizations, with a relative lack of classic simple motor and/or phonic tics and the absence of the expected rostrocaudal progression at onset (6), characteristic of primary tic disorders. Common manifestations include large amplitude arm movements, hitting objects, hitting/punching self or family members, clicking, whistling, repeating a wide range of random and/or bizarre words or phrases, and blurting out obscenities or offensive statements. In many cases, a premonitory urge prior to these tic-like behaviours is endorsed, as are distractibility and suggestibility. However, suppressibility of tic-like behaviours is more limited and variable between individuals. The magnitude of functional disability and level of parental distress caused by the tic-like behaviours is extreme. Family functioning is often dramatically affected and disrupted. Moreover, many of these young people can no longer attend school or work due to symptom manifestation but are able to perform some activities of daily living (e.g., utilization of smartphones, computers, creative projects, etc.).

< Insert Table 2 here >
The phenomenology of these rapid onset cases represents a noticeable departure from the usual demographic and natural history of TS (see Table 3). Tic onset in TS typically occurs between four and seven years of age. Boys are disproportionately affected, by a ratio of over three to one (7). Tics typically begin insidiously, with young children usually having a few different tics at a time that wax and wane and evolve in character. In early years, tics are mostly simple, for example, eye blinking, nose wrinkling, facial grimacing, sniffing, throat clearing, or coughing. Complex tics may emerge later, over a period of months to years, but typically after simple tics have been present for some time. Tics often worsen in preadolescence (age 10-12) and improve in late adolescence (8). Other typical characteristics of tic disorders, such as the report of premonitory sensations or urges to perform tics, subsequent relief of urges after the tic, suggestibility, and distractibility can be present in association with both tics (more so in adolescents than in children (9, 10)) and functional tic-like behaviors and may therefore be less useful in differentiating these two groups of patients. At difference, an ability to suppress or postpone tics at least briefly is usually demonstrated in older children with ‘typical’ tics, whereas suppressibility of functional tic-like behaviors appears to be less efficient. The associated psychiatric comorbidity pattern in these rapid onset cases also differs from TS. The most common comorbid disorders in children diagnosed with TS are Attention Deficit/Hyperactivity Disorder (ADHD) and Obsessive-Compulsive Disorder (OCD)(11). In the rapid onset cases, there is a higher representation of anxiety disorders and major depression.

Although most young people with this rapid onset of tic-like behaviors have not reported any history of previous tics, we have witnessed several young patients with a history of mild
simple tics who reported an explosive onset of complex tic-like behaviours during the same period. Age, sex distribution, phenomenology and type of onset in this less represented subgroup are similar to the majority of youth with rapid onset of complex tic-like behaviors without previous history of tics. This similarity intriguingly suggests the possibility of shared predisposing factors in these two subgroups. This presentation differs substantially also from other acute syndromes in which tics or tic-like movements are predominant. In particular, we did not notice any association with recent upper respiratory/pharyngeal infections or acute obsessive-compulsive spectrum symptoms (e.g. those observed in pediatric acute onset neuropsychiatric syndromes or PANS), and the phenomenology was not consistent with acute drug-induced movement disorders.

Another relevant characteristic of this new clinical presentation is the association with specific psychosocial stressors, the exposure to which may have increased substantially during the COVID-19 pandemic in this age group. A proportion of these patients reported family-related emotional distress linked to tensions between parents or other family members, which may have been exacerbated by the lockdown. Other patients have described a temporal association between symptom onset and increased stress levels related to “virtual schooling”, meeting academic expectations, and navigating school/home transitions that are accompanied by several academic challenges.

What could be at the origin of this specific, explosive presentation of tic-like behaviors, and why is it occurring now? Recently, there has been a growth in online video material of youth manifesting tic disorders, shared on social networks. In some cases, these videos were pooled under thematic hashtags focused on TS and have yielded exponentially rising popularity at the beginning of 2021. Interestingly, we and others (1, 12, 13) have noticed a
phenomenological similarity between the tics or tic-like behaviors shown on social media and
the tic-like behaviors of this group of patients. In some cases, the patients specifically
identified a link between these media exposures and the onset of symptoms, although, with
some of the younger children, the social media use was only disclosed after careful
questioning. The COVID-19 pandemic has been a major source of stress and anxiety for
people around the world, resulting in increased mental health symptoms and demand for
mental health services (14-18). Increased social isolation and the widespread utilization of
social media may have contributed as precipitating factors in a relevant proportion of these
patients. External factors like watching popular social media personalities’ videos portraying
tics or tic-like behaviours may have instilled a belief that ‘tics’ may catalyse peer acceptance
or even popularity. This exposure to tics or tic-like behaviors is a plausible trigger for the
behaviors observed in at least some of these patients, based on a disease modelling
mechanism. However, this specific social media exposure to tic-related videos, while
reported in all patients in the Calgary series, was not reported in every patient seen at all the
other centres, suggesting that it cannot be considered a pre-requisite or necessary causative
factor. There is a need for systematic investigation of the relationship between symptom
onset, severity and amount of social media exposure. The explosive behavioral pattern
exhibited by these young people could also share pathophysiological mechanisms with the
general population of people with functional tic-like behaviors, as proposed in greater detail
in Figure 1.

A comprehensive interview of patients, families and relevant informants is a first, necessary
step to understand the antecedents and triggering factors involved, which will allow deeper
understanding of this clinical picture and guide personalized management decisions.
Comparisons to historical precedents of similar outbreaks at a more local level are also
useful. For example, a regional outbreak of tic-like behaviors was documented in adolescent girls in 2012 in Le Roy, New York, which was attributed to a combination of conversion disorder with mass psychogenic illness (19).

As our familiarity with this behavioral pattern increases through clinical experience, we need to explore in depth the psychopathological profile of these patients, as well as identify recurrent predisposing family- and peer-related stressors. It would also be relevant to investigate social and adaptive functioning as well as social cognition domains, particularly the processing of socially salient stimuli, their perception and integration of reward mechanisms related to social cues. Finally, a striking characteristic of this behavioral pattern is its ‘epidemic’ diffusion over a relatively short time, which differs from the slower pace of referral to specialists’ attention of functional tic-like behaviors and indicates the involvement of suggestibility and behavioral modelling. In this respect, it would be useful to explore whether abnormalities of sense of agency and action monitoring, similar to those observed in people with other functional neurological disorders, are a consistent trait also in these patients or whether performance on these domains is more variable.

We wish to bring neurologists’ attention to this emerging disorder and highlight the important phenotypic differences these cases have from typical cases of TS. A prompt diagnosis and expert review to clarify the phenomenology when necessary is recommended. We also acknowledge that diagnostic labelling may be difficult when childhood onset simple tics and the more complex types of rapid-onset tic functional tic-like behaviors co-exist in the same patient (20). Our initial, anecdotal experience is that these patients do not respond typically to conventional pharmacotherapies for tics, either showing dramatic improvement within hours or days of starting an alpha agonist (suggestive of a placebo response) or having no response
whatsoever to antipsychotic medications with demonstrated high efficacy for tics (21).

Behavioral treatment approaches including personalized psychoeducation seem more appropriate to initiate a therapeutic process. Intuitively, function-based therapeutic strategies (22-24), including mitigating potential triggering exposures, particularly social media content associated with tics, initiating stress management interventions related to other identifiable psychosocial stressors, reducing social reactions to symptom expression, and addressing comorbid anxiety and depression could be confirmed as high yield strategies by future observations. Our prediction is that cognitive behavioral therapies, particularly when including components of the Comprehensive Behavioural Intervention for Tics (CBIT) (25), might have a considerable chance of success to treat this type of repetitive behavior.

Figure 1 Legend: Possible pathophysiological mechanisms for the functional tic-like behaviours exhibited by this group of patients. As recently proposed in the context of functional tic-like behaviors (19), a combination of predisposing traits (encompassing, among others, genetic and epigenetic factors and previous life events), predisposing states (such as raised anxiety levels and related low mood) and environmental precipitating factors (increase in media exposure to tic-like behaviors, different stressors driven by the pandemic) may prompt an excess of behavioral alterations, such as recurrent tic-like behaviors. In specific groups of people like the one that this viewpoint is focusing on, the environment might be providing the individual with over-abundant external stimuli that may be discerned as highly salient (i.e., attractive and ‘popular’ tics or tic-like behaviours). Such behaviours will be selected and reinforced, and the individual will, particularly at an initial learning stage, allocate an excess of attention to them, thereby enhancing their probability of recurrence reinforcement.

Table 1 Legend: Calgary Tic Disorders Clinical Registry Comparison of Clinical and Demographic Features of Primary Tic Disorder cases with Rapid Onset Functional Tic Like Behaviours

Table 2 Legend: Estimated Proportion of Referrals for Functional Tic Like Behaviours and Average Annual New Patient Referrals for Tics/Movement Disorders, Pre and Post COVID-19 Pandemic
Table 3 Legend: Side-by-side Comparison of Phenomenological Presentation of tics and rapid onset functional tic-like behaviors.
Documentation of Author Roles

1. Research project: A. Conception, B. Organization, C. Execution


3. Manuscript Preparation: A. Writing of the first draft, B. Review and Critique

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Full financial disclosure for the previous 12 months

Tamara Pringsheim has received grant funding from the Public Health Agency of Canada and Alberta Health.

Christos Ganos holds a research grant from the VolkswagenStiftung (Freigeist Fellowship) and has received honoraria from the Movement Disorders Society for educational activities.

Joseph F. McGuire reports receiving research support from the Tourette Association of American (TAA), the American Academy of Neurology (AAN), the American Psychological Foundation (APF), and the Hilda and Davis Preston Foundation. He has served as a
consultant to Bracket Global, Syneos Health, and Luminopia, and also received royalties from Elsevier.

Tammy Hedderly has no financial disclosures.

Douglas Woods has received book royalties from Oxford University Press, Guilford Press, and Springer Press and has received speakers fees from the Tourette Association of America.

Donald L. Gilbert has received honoraria and/or travel support from the Tourette Association of America/ Centers for Disease Control and Prevention, the Child Neurology Society, and the American Academy of Neurology. He has received compensation for expert testimony for the U.S. National Vaccine Injury Compensation Program, through the Department of Health and Human Services. He has received payment for medical expert opinions through Advanced Medical/Teladoc. He has served as a consultant for Applied Therapeutics and Eumentics Therapeutics. He has received research support from the NIH (NIMH) and the DOD. He has received salary compensation through Cincinnati Children’s for work as a clinical trial site investigator from Emalex (clinical trial, Tourette Syndrome) and EryDel (clinical trial, Ataxia Telangiectasia). He has received book/publication royalties from Elsevier, Wolters Kluwer, and the Massachusetts Medical Society.

John Piacentini has no financial disclosures.

Russell C. Dale has National health and Medical research Council Investigator fellowship and Cerebral Palsy Alliance funding.

Davide Martino No conflicts of interest to report. Compensation for consultancies for Sunovion, honoraria from Dystonia Medical Research Foundation Canada, royalties from Springer-Verlag, research support from Ipsen Corporate, funding grants from: Dystonia Medical Research Foundation Canada, Parkinson Canada, The Owerko Foundation, and the Michael P Smith Family.
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12. Hull M, Parnes M. Tics and TikTok: Functional tics spread through social media. Movement Disorders Clinical Practice. n/a(n/a).
Table 1 Calgary Tic Disorders Registry Comparison of Clinical and Demographic Features

<table>
<thead>
<tr>
<th>Variable</th>
<th>Primary Tic Disorder N=270</th>
<th>Rapid Onset Functional Tic-Like Behaviours N=20</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Female Sex, proportion</td>
<td>58 (21%)</td>
<td>19 (95%)</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>Age at first clinical visit (mean and 95% CI)</td>
<td>10.5 years (10.1, 10.9)</td>
<td>14.3 years (13.5, 15.0)</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>Age at tic onset (mean and 95% CI)</td>
<td>6.4 years (6.1, 6.8)</td>
<td>13.9 years (13.1, 14.7)</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>YGTSS Total Tic Score</td>
<td>18.4 (17.4, 19.5)</td>
<td>33.3 (28.7, 38.0)</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>YGTSS Impairment Score</td>
<td>15.8 (14.2, 17.3)</td>
<td>28.6 (23.1, 34.1)</td>
<td>0.0001</td>
</tr>
<tr>
<td>ADHD diagnosis, proportion</td>
<td>120 (44%)</td>
<td>5 (25%)</td>
<td>0.09</td>
</tr>
<tr>
<td>Conners 3 Inattention Subscale T score</td>
<td>65.2 (63.3, 67.1)</td>
<td>68.9 (61.1, 76.8)</td>
<td>0.16</td>
</tr>
<tr>
<td>Conners 3 Hyperactivity Subscale T score</td>
<td>67.9 (66.0, 69.9)</td>
<td>64.8 (57.3, 72.3)</td>
<td>0.21</td>
</tr>
<tr>
<td>OCD diagnosis, proportion</td>
<td>51 (19%)</td>
<td>1 (5%)</td>
<td>0.12</td>
</tr>
<tr>
<td>CYBOCS score</td>
<td>5.1 (4.1, 6.1)</td>
<td>2.7 (0.9, 13.1)</td>
<td>0.22</td>
</tr>
<tr>
<td>Anxiety disorder diagnosis, proportion</td>
<td>51 (19%)</td>
<td>15 (75%)</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>MASC2 Total T Score</td>
<td>57.4 (55.3, 59.5)</td>
<td>71.0 (64.6, 77.4)</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>Depression diagnosis, proportion</td>
<td>11 (4%)</td>
<td>11 (55%)</td>
<td>&lt;0.0001</td>
</tr>
</tbody>
</table>
Table 2 Estimated Proportion of Referrals for Functional Tic Like Behaviours and Average Annual New Patient Referrals for Tics/Movement Disorders, Pre and Post COVID-19 Pandemic

<table>
<thead>
<tr>
<th>Centre</th>
<th>Pre-pandemic Estimated percentage of referrals for FTLBs as the primary problem</th>
<th>January to June 2021 Estimated percentage of referrals with FTLBs as the primary problem</th>
<th>Pre-pandemic Average number of referrals received per year for tics/movement disorders</th>
<th>2020-2021 Average number of referrals received per year for tics/movement disorders</th>
</tr>
</thead>
<tbody>
<tr>
<td>Calgary Alberta Children’s Hospital Tourette Clinic</td>
<td>1-2%</td>
<td>30%</td>
<td>186</td>
<td>290</td>
</tr>
<tr>
<td>Institution</td>
<td>2-5%</td>
<td>35%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>-------------------------------------------------</td>
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</tr>
<tr>
<td>Sydney Children’s Hospital at Westmead Tic Clinic</td>
<td></td>
<td></td>
<td>82</td>
<td>116</td>
</tr>
<tr>
<td>TANDeM Evelina London Childrens Hospital GSTT</td>
<td>2%</td>
<td>30%</td>
<td>300</td>
<td>600</td>
</tr>
<tr>
<td>Cincinnati Children’s Movement Disorders Clinic</td>
<td>1%</td>
<td>20%</td>
<td>600</td>
<td>600</td>
</tr>
<tr>
<td>UCLA Child OCD, Anxiety and Tic Disorders Program</td>
<td>2%</td>
<td>20%</td>
<td>92</td>
<td>71</td>
</tr>
</tbody>
</table>

FTLBs = functional tic-like behaviours

Table 3. Side-by-side Comparison of Phenomenological Presentation of tics and rapid onset functional tic-like behaviors.

<table>
<thead>
<tr>
<th></th>
<th>Typical TS Tics</th>
<th>Rapid Onset Functional Tic-like Behaviors</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age of Onset</td>
<td>Childhood</td>
<td>Adolescence or early adulthood</td>
</tr>
<tr>
<td>Symptom Onset</td>
<td>Gradual</td>
<td>Abrupt/Acute</td>
</tr>
<tr>
<td>Initial Type of Tic</td>
<td>Simple Motor</td>
<td>Complex Motor or Complex Vocal</td>
</tr>
<tr>
<td>---------------------</td>
<td>--------------</td>
<td>--------------------------------</td>
</tr>
<tr>
<td>Sex</td>
<td>Male predominance</td>
<td>Female predominance</td>
</tr>
<tr>
<td>Most common tics</td>
<td>Eye blinking, Head movements, Sniffing, Throat clearing</td>
<td>Large amplitude arm movements, Self-injurious movements (e.g., hitting self or family members), Wide range of odd words or phrases, Obscene words or phrases</td>
</tr>
<tr>
<td>Most common comorbidities</td>
<td>Attention Deficit/Hyperactivity Disorder, Obsessive-Compulsive Disorder</td>
<td>Anxiety Disorders, Depressive Disorders</td>
</tr>
<tr>
<td>First-line treatment approach</td>
<td>Comprehensive Behavioral Intervention for Tics (CBIT), Exposure and Response Prevention (ERP), Alpha adrenergic agonists</td>
<td>Psychoeducation, Cognitive behavioral therapy, CBIT, with particular emphasis on the functional interventions - identification and management of antecedents and consequences of functional tic-like behaviours</td>
</tr>
</tbody>
</table>
Figure 1

- **PREDISPOSING TRAITS**
  - Genetic factors
  - Epigenetic factors
  - Interceptive awareness
  - Social cognition traits
  - Previous life events

- **PREDISPOSING STATES**
  - Anxiety levels
  - Depressed mood

- **PRECIPITATING FACTORS**
  - Psychosocial stressors (e.g., social isolation, online schooling, other)
  - Physical stressors
  - Exposure to tics and tic-like behaviours through social media or other sources

- **EXPLOSIVE-ONSET TIC-LIKE BEHAVIOURS**
- Attention focus on novel behaviours
- Behavioural reinforcement
- Action selection based on salience/social reward
- Perceived intensity of external stimuli

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