孤独症儿童潜在早期识别标志

——发声异常及原因探析*

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摘要 孤独症谱系障碍(Autism Spectrum Disorder，ASD)儿童的早期识别及早期干预对其发育结果至关重要。发声是指儿童学会说话之前发出的声音。已有研究表明，非类言语和类言语发声异常是 ASD 儿童 2 岁前的潜在识别标志。ASD 儿童发声异常的理论解释主要有动机导向、神经运动导向、感知觉导向和社会反馈导向理论。未来研究可考虑探讨发声异常作为 ASD 儿童独特的早期识别标志的可能性，加强哭声在 ASD 儿童早期筛查中的研究，探索基于最具预测性声学参数集的自动学习分类模型，探究内在动机与社会动机对 ASD 儿童类言语发声的影响和进一步探究 ASD 儿童类言语发声异常的神经机制。从而为 ASD 儿童的早期识别及早期干预提供更加客观的理论依据。

关键词 孤独症谱系障碍，潜在早期识别标志，发声异常，原因探析

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1 引言

孤独症谱系障碍(Autism Spectrum Disorders，ASD)是一种神经发育性疾病，有两个核心症状：一是，社会沟通和社会互动存在持续性的缺陷；二是，表现出受限的和重复的行为模式、兴趣或活动(American Psychiatric Association, 2013)。ASD 有两种主要的发病模式：1 岁前发病的早发型(early onset)和 2 岁左右发病的倒退型(regression)(Boterberg et al., 2019)。倒

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退型 ASD 占比大约为 30%，主要以已习得的语言或社交技能的倒退或丧失为标志(Tan et al., 2021)。最新的数据显示全球 ASD 发病率略低于 1%(Lord et al., 2020)，我国 ASD 发病率为 1/142(Zhou et al., 2020)，美国 ASD 发病率为 1/54(Maenner et al., 2020)，由此可见 ASD 发病率较高。虽然目前没有治疗 ASD 核心症状的特效药(Goel et al., 2018)，但在早期尤其是 2 岁以前若能及时发现和干预，能有效改善 ASD 个体的语言、社交沟通等能力的发展轨迹并影响长期结果(Estes et al., 2015; Frazier et al., 2021; Sigafoos & Waddington, 2016; Wetherby et al., 2018)，甚至“预防”全面综合征的出现(Yirmiya & Charman, 2010)。

发声(vocalization)是指儿童学会说话之前发出的声音(Schoen Simmons, 2021)。发声包括非类言语发声(nonspeech-like vocalizations)和类言语发声(speech-like vocalizations)(Oller, 2000)。非类言语发声是与生物功能有联系的发声，包括有固定信号作用的声音(fixed signals)和植物性声音(vegetative sounds)(Garrido et al., 2017; Nathani et al., 2006)；类言语发声是指类似言语的声音，包括不同的类型并沿着一个连续体发展，在 1 岁左右变得更像言语(Nathani et al., 2006; Oller, 2000)。ASD 儿童的发声特征通常采用更具体的指标进行分析，其中非类言语发声常用分析指标为频率、比例、基频、停顿长度、持续时间等(e.g., Esposito et al., 2013; Plumb & Wetherby, 2013)，类言语发声常用分析指标为发声复杂性和沟通性发声(e.g., Mcdaniel et al., 2020; Yoder et al., 2016)。发声的具体类型及常用分析指标见表 1。

发声是早期正常发展的一个关键成就。正常发展(Typical Development，TD)儿童的发声在 0~1 岁之间表现出哭声和咕咕声频率逐步下降，元音和规范音节频率逐步上升的趋势(Cychosz et al., 2021; Nathani et al., 2006)。发声的非典型性发展是发育异常的指标，对早期发声的深入理解使得神经发育性疾病的早期发现和鉴别成为可能(Marschik et al., 2017)。研究表明，ASD 儿童的发声存在异常现象，与其核心的非典型沟通症状有关(Gabrielsen et al., 2015)，对发声异常的早期识别可能有利于改善 ASD 儿童平均在 5 岁时才能被诊断的现状

(van’t Hof et al., 2021)。然而，关于 ASD 发声特征已有研究存在分歧。例如，有研究支持 ASD 儿童比 TD 儿童有更多非类言语沮丧声的结论(Plumb & Wetherby, 2013)，也有研究发现两类儿童非类言语沮丧声无差异(Schoen et al., 2011)。有研究表明 ASD 儿童比 TD 儿童的类言语发声更少(Patten et al., 2014)，而另一研究却发现 ASD 儿童类言语发声过度(Werner & Dawson, 2005)。那么，ASD 儿童有哪些非典型的发声异常？这些发声异常作为早期识别标志的可能性如何？

本文将总结近年来 ASD 儿童早期发声的研究成果，概括 ASD 儿童发声异常的特征，分析 ASD 儿童发声异常作为早期识别标志的可能性，探析 ASD 儿童发声异常的原因，从而为 ASD 儿童的早期发现及早期干预提供一些新思路。

表 1 发声的类型、定义及常用分析指标

类型		定义	常用分析指标
非类言语发声	愉悦声(pleasure/delight)	表达愉悦情绪的声音，如笑声	非类言语发声的频次(非类言语发声的次数)、频率(单位时间内非类言语发声的频次)、比例(如沮丧声占总非类言语发声比例)等 基频(fundamental frequency, f0)是声带每秒振动的次数，反映了声音的音调 停顿长度(pause length)是哭声之间的停顿时间 持续时间(cry duration)是哭声的持续时间
	非典型发声(atypical)	一种非典型的声音，主要包括高音调的尖叫声、低音调的吼叫声和高强度的叫喊声	
	沮丧声(distress)	一种与消极情绪状态有关的发声，主要包括强烈的哭声、哀嚎声	
类言语发声	准共振元音(quasivowel)	一种微弱的、低音调的、有时伴有鼻音的呼噜声，产生时喉咙闭合、呼吸支持很少、声音很短(通常小于 100ms)、2000Hz 以上部分能量不足，很难转录为成人元音	发声复杂性(vocal complexity)是类言语发声的频次/频率、一致性和多样性。如非规范音节频率或频次(准共振元音、单个元音、单个辅音、边缘音节)，规范音节比例或频率，辅音类型等
	元音(vowel)	产生时喉咙开放、有清晰的共振峰	

边缘音节(marginal syllable)	也称边缘呀呀语(marginal babbling)，是指缓慢的 CV 序列，C 和 V 之间有较长的转换时间(通常大于 250ms)	沟通性发声(vocal communication)是指通过眼神、手势指向他人的明显为了传递信息的发声。如发声中用于沟通的发声比例，沟通性发声行为的数量
规范音节(canonical syllable)	主要包括单个的 CV 音节和由 2 个及以上 CV 序列构成的规范呀呀语(canonical babbling)。C 和 V 之间具有快速的转换(通常小于 250ms)。其中规范呀呀语包括重叠音节(reduplicated syllable，具有相同的 CV 组合序列)和非重叠音节(nonreduplicated syllable，具有变化的不同的 CV 组合序列)	
复杂音节(complex syllable)	主要包括不同于 CV 音节以外的 VC、CCV 等单音节，VCV、VCVC 等复杂双音节，VCVCV、VCVCCV 等有或无重音及语调的多音节串	

注: 非类言语发声分为固定信号声音和植物性声音，其中植物性声音是反射性发声，如咳嗽、打嗝、打喷嚏等发声，在发声研究中一般不对植物性声音进行分析，故此处只包括固定信号声音；C: 辅音，consonant；V: 元音，vowel。

2 发声异常与 ASD 早期识别

2.1 非类言语发声异常与 ASD 早期识别

对 ASD 儿童非类言语发声特征的研究主要包括两个方面：一是，非类言语发声频次、频率或比例；二是，哭声的基频、停顿长度和持续时间。虽然，已有非类言语发声频率、比例等研究结果不一致(e.g., Plumb & Wetherby, 2013; Schoen et al., 2011)。然而，关于沮丧声中哭声声学异常特征的结论相对一致，即 ASD 儿童的哭声基频较高，停顿长度和持续时间较短(e.g., Bornstein et al., 2016; Esposito et al., 2014; Esposito et al., 2015)。

2.1.1 较高的哭声基频是 ASD 潜在早期识别标志

基频作为最能说明哭声声学特征的参数(Lester & Gasse, 2020)，在 ASD 儿童哭声的研究中被研究者广泛使用。研究中 ASD 儿童哭声基频样本主要来自两方面：一是，熟悉或陌生

自然情境中的非疼痛哭声；二是，自然情境中的疼痛哭声。研究发现，ASD 儿童哭声基频高于同龄 TD 儿童。对已确诊的 ASD 儿童早期哭声的早期家庭视频分析表明，5~18 个月的 ASD 婴儿非疼痛哭声基频高于 TD 婴儿(Bornstein et al., 2016; Esposito et al., 2012, 2013; Esposito et al., 2015; Esposito & Venuti, 2010a, 2010b)。15 个月的 ASD 高风险婴儿和年龄稍大的 36~52 个月 ASD 幼儿陌生情境中较高的非疼痛哭声基频支持上述结果(Esposito et al., 2014; Ozturk et al., 2018)。前瞻性研究发现，熟悉的自然情境中 6 个月的 ASD 高风险婴儿疼痛哭声基频高于低风险婴儿，两组婴儿非疼痛哭声无显著差异，但后来被诊断为 ASD 的婴儿的两类哭声基频均最高(Sheinkopf et al., 2012)。以上证据显示，较高的哭声基频可能是 ASD 的早期识别标志，尤其较高的疼痛哭声基频可能是一个更敏感的指标。

2.1.2 较短的哭声停顿长度和持续时间是 ASD 潜在早期识别标志

哭声的停顿长度和持续时间反映了呼吸控制能力(LaGasse et al., 2005)，在 ASD 婴儿哭声研究中探究较多。已有研究表明，ASD 儿童哭声的停顿长度和持续时间短于同龄 TD 儿童。研究表明，12~13 个月的 ASD 婴儿哭声的停顿长度明显短于 TD 婴儿(Esposito et al., 2012, 2013; Esposito et al., 2015; Esposito & Venuti, 2009; Venuti et al., 2012)。前瞻性研究发现，ASD 高风险婴儿在 12 个月和 15 个月时哭声的持续时间短于 TD 婴儿，且在 2 岁或 3 岁被诊断为 ASD 的婴儿的哭声持续时间最短(Esposito et al., 2014; Unwin et al., 2017)。以上证据表明，较短的哭声停顿长度和持续时间可能是 ASD 的早期识别标志。

2.2 类言语发声异常与 ASD 早期识别

2.2.1 异常的发声复杂性是 ASD 潜在早期识别标志

有大量研究探讨了 ASD 儿童的发声复杂性，虽然有两项研究发现 ASD 儿童或 ASD 高风险儿童的发声复杂性与 TD 儿童无显著差异(Chericoni et al., 2016; Talbott et al., 2016)，但有更多的证据显示 ASD 儿童的类言语发声复杂性表现出异常现象，主要表现为类言语发声

复杂性明显低于或高于同龄 TD 儿童(e.g., Garrido et al., 2017; Heymann et al., 2018; Werner & Dawson, 2005)。

首先，多数研究证实 ASD 儿童类言语发声复杂性低于同龄 TD 儿童。前瞻性和回溯性研究表明，2 岁前 ASD 婴儿的元音、边缘音节和规范音节等类言语发声频率或比例显著低于 TD 婴儿且出现延迟现象(Garrido et al., 2017; Patten et al., 2014; Plumb & Wetherby, 2013; Wetherby et al., 2004)，表现出更少的辅音类型(Schoen et al., 2011; Wetherby et al., 2007)。ASD 高风险婴儿的元音、重叠音节、复杂音节等类言语发声频率或比例(Heymann et al., 2018; Paul et al., 2011; Winder et al., 2013)和辅音类型(Chenausky et al., 2017; Paul et al., 2011)均低于低风险婴儿。此外，研究显示类言语发声复杂性对 ASD 诊断具有良好的预测性。Patten 等人(2014)发现 9~12 个月时规范音节占总音节发声比率和音节发声频率对 ASD 诊断预测的准确率达 82%。Paul 等人(2011)发现 9 个月和 12 个月 ASD 高风险婴儿的辅音类型能预测 24 个月时 ASD 症状，准确率分别为 77%和 65%。以上证据表明，较低的类言语发声复杂性可能是 ASD 早期识别标志。

其次，少量倒退型 ASD 儿童类言语发声复杂性高于同龄 TD 儿童，出现过度发声现象。回溯性研究发现，15 名 12 个月大的倒退型 ASD 婴儿复杂呀呀语频率显著高于 TD 婴儿，而另外 21 名早发型 ASD 婴儿复杂呀呀语频率低于 TD 婴儿。进一步分析发现 46%倒退型 ASD 婴儿的复杂呀呀语频率高于 TD 婴儿的平均值，只有 6%早发型 ASD 婴儿表现出这种现象(Werner & Dawson, 2005)。这项研究还发现，随着年龄的增长，倒退型 ASD 儿童的复杂呀呀语逐步退化，在 24 个月时他们的复杂呀呀语与早发型 ASD 婴儿一样，都显著落后于 TD 婴儿。最近一项前瞻性研究发现，9 个月大 ASD 高风险婴儿比低风险婴儿有更多的类言语发声，这一结果主要体现在 12 名(31%)ASD 高风险婴儿的过度发声(Swanson et al., 2018)。以上结果表明，较高的发声复杂性可能是倒退型 ASD 儿童的早期识别标志。

2.2.2 较少的沟通性发声是 ASD 潜在早期识别标志

沟通性发声能够评价 ASD 儿童发声的社会性(Mcdaniel et al., 2020)。研究表明 ASD 儿童沟通性发声少于同龄 TD 儿童，2 岁前 ASD 婴儿有意图的发声沟通行为明显少于 TD 婴儿(Bacon et al., 2018; Dow et al., 2020; Dow et al., 2017; Plumb & Wetherby, 2013; Shumway & Wetherby, 2009)。ASD 高风险婴儿研究也支持上述结果，ASD 高风险婴儿产生更少的社交导向发声，随后被确诊的 ASD 婴儿的沟通性发声能力最低(Garrido et al., 2017; Sacrey et al., 2021; Winder et al., 2013)，且过度发声的 ASD 高风险婴儿的社交呀呀语也低于低风险婴儿(Swanson et al., 2018)。研究还发现，相比较单独使用发声复杂性，在结合考虑沟通性发声和总发声率后更有可能预测 ASD 的诊断(Garrido et al., 2017)。以上证据表明，沟通性发声与沟通复杂性的相互作用可能是预测 ASD 的更有效标志。

3 ASD 儿童发声异常的原因探析

综合已有研究结果，发现 ASD 儿童非类言语哭声基频较高，停顿长度和持续时间较短；类言语发声复杂性较低或较高，沟通性发声较少。这些发声异常不仅可能是 ASD 早期识别标志，也在一定程度上反映了 ASD 儿童社会沟通和社会互动方面的核心缺陷。以下结合动机导向、神经运动导向、感知觉导向和社会反馈导向等理论对现有结论进行分析与解释。

3.1 动机导向理论

ASD 儿童对特殊兴趣的强内在动机在一定程度上解释了他们类言语发声复杂性异常的问题。内在动机(intrinsic motivation)是指个体自发的好奇心和兴趣、主动练习技能和学习知识的倾向，受多巴胺能系统的支持，与注意力控制和自我参照认知的神经网络相关(Di Domenico & Ryan, 2017)。计算机建模研究证明，内在动机是婴儿类言语发声发展的一般机制(Acevedo-Valle et al., 2018; Moulin-Frier et al., 2014)，其关键思想是自我生成和目标选择。内在动机让婴儿自主构建自己的发声试验和制定自己的学习计划，解释了婴儿从最初忽略

周围语音环境的自我探索发声到受成人语音影响的模仿发声的自动转变，使婴儿的发声从最初的非类言语发声过渡到不清晰的准元音再到清晰的复杂规范音节。内在动机对 TD 婴儿类言语发声的作用也在行为研究中得到了验证，TD 婴儿的类言语发声大多是在独自探索的游戏中产生的(Oller et al., 2019)。当成人寻求与婴儿社会互动时，婴儿大多数发声(约 60%)似乎不是针对成人的；当成人与婴儿在一起但没有试图参与婴儿的活动时，这种探索性发声的优势更强(约 80%)(Long et al., 2020)。研究发现 ASD 个体比正常人群有更多刻板地从事特殊兴趣的内在动机(Grove et al., 2016)，且这一现象在 1 岁前的 ASD 婴儿中也很明显(Bacon et al., 2018)。ASD 婴儿可能更喜欢重复玩弄音节，而 TD 婴儿更倾向于探索语音的细微差别(Long et al., 2021)。那么可以推测，ASD 婴儿对特殊兴趣的强内在动机可能影响了他们类言语发声复杂性尤其是辅音多样性的习得，同时也可能导致倒退型 ASD 儿童类言语发声复杂性过高。值得注意的是，目前较缺乏内在动机对 ASD 儿童类言语发声影响的实证研究。近期 Long 等人(2021)尝试探究内在动机对 ASD 高风险和低风险婴儿规范音节发声的影响，结果并未显示内在动机对不同 ASD 风险婴儿的规范音节产生影响。未来对已确诊的 ASD 婴儿进行探究，也许能够提供内在动机影响类言语发声的新证据。

社会动机缺陷可能是 ASD 儿童沟通性发声较少的原因之一。社会动机(social motivation)是一套心理倾向和生物机制，偏向于个人优先适应社会世界，在社会互动中寻求和享受快乐，努力培养和维持社会纽带(Chevallier et al., 2012)。Chevallier 等人(2012)在总结已有证据基础上认为，社会动机机制的破坏可能构成了 ASD 的主要缺陷。ASD 社会动机缺陷与社会性奖励脑区功能失调有关，主要涉及尾状核和前扣带回功能降低，中脑腹侧被盖区和伏隔核之间的结构与功能连接性降低(Clements et al., 2018; Supekar et al., 2018)。ASD 儿童对面孔关注减少，更少参与到亲子互动中和发起共同注意互动行为也为社会动机缺陷假说提供了有力证据(Adamson et al., 2019; Reid et al., 2017)。在社会动机缺陷模型下，ASD 儿童很少主动采用

类言语发声的形式主动与他人沟通。此外，儿童的类言语发声发展涉及儿童与成人双主体的互动(Elmlinger et al., 2019; Oller et al., 2016)，ASD 儿童的社会动机缺陷使得他们难以与成人形成良好的社会互动。那么，社会动机也可能影响 ASD 儿童类言语发声复杂性的发展。然而，目前关于社会动机对 ASD 儿童类言语发声发展影响的实证研究极其有限，未来还需要开展该领域的研究。

3. 2 神经运动导向理论

类言语发声神经运动方面的不成熟或紊乱对 ASD 儿童类言语发声复杂性可能有特定影响。类言语发声神经运动机制可以从言语的神经运动方面进行分析，因为类言语发声或准言语(quasispeech)具备言语的语音结构但不带有言语的意义(Kent, 2015)，且有大量研究采用无意义的元音或规范音节作为材料对言语产生的神经系统进行研究(Kumar et al., 2016)。位于额叶初级运动皮质中的喉运动皮质(Laryngeal Motor Cortical, LMC)参与控制言语运动(Eichert et al., 2020)。对元音发声的功能核磁共振研究表明，初级运动皮质中喉部和下颌部肌肉控制区域有重叠，研究者认为这种皮质间的重叠可能支持了喉部发声和下颌振荡之间的耦合，从而产生音节结构(Brown et al., 2021)。此外，额叶皮质的认知功能也参与控制发声(Hage, 2018)。然而，ASD 儿童在 2 岁前前额叶和其它皮质过度生长(Bonnet-Brilhault et al., 2018; Hazlett et al., 2017; Hazlett et al., 2011; Kaushik & Zarbalis, 2016)，这可能会影响类言语发声运动的控制，从而导致类言语发声复杂性异常。不过，目前 ASD 儿童如何控制发声器官以协调产生类言语发声的神经运动机制尚不清晰，未来还需开展此方面的研究。

坐姿发育延迟可能是 ASD 儿童类言语发声复杂性较低的一个重要原因。坐姿为下颌活动提供了生物力学的支持，儿童类言语发声是通过打开和闭合下颌实现的(Green et al., 2002)。有研究比较了 6 个月时能独立坐和不能独立坐婴儿的音节发声，结果表明他们的音节发声频率存在差异，41%能独立坐的婴儿产生了音节发声，而只有 9%未独立坐的婴儿产生了音

节发声(Leezenbaum, 2016)。回溯性研究发现，ASD 儿童稳定坐立的平均年龄为 9 个月，延迟于 TD 儿童的 7.9 个月(Ucuz & Cicek, 2020)。前瞻性研究支持上述结论，ASD 高风险婴儿的坐姿与低风险婴儿不同(Kyvelidou et al., 2021)，6 个月时 ASD 高风险婴儿独立坐立的时间显著低于低风险婴儿(Leezenbaum & Iverson, 2019)。这些研究表明，坐姿习得较慢限制了 ASD 儿童坐立时探索新发声可能性的机会，从而影响了规范音节的习得(Iverson, 2018)。

迷走神经复合体损伤与基频的非典型模式有关，也反映了呼吸调节和哭声时间的模式(LaGasse et al., 2005)。人类声音基频的改变主要是通过肌肉运动改变声带的物理特性，如声带的长度、单位长度的质量、紧张度和硬度而实现的，而控制声带运动的喉内肌群主要由迷走神经支配(Dankbaar & Pameijer, 2014)。研究表明 ASD 儿童迷走神经表现出张力减弱的现象(Benevides & Lane, 2015)，这可能会导致支配哭声的声带控制不良与呼吸肌群运动协调不良(Sheinkopf et al., 2016)，从而导致哭声基频较高、哭声停顿长度和持续时间较短等异常的哭声模式。

3. 3 感知觉导向理论

ASD 儿童的听处理障碍和对言语声音刺激的非典型加工(Filipe et al., 2018; Soskey et al., 2017; Vlaskamp et al., 2017; Yau et al., 2016)可能影响他们的类言语发声复杂性。言语诱发听觉脑干反应的研究表明，ASD 儿童脑干水平语音听觉加工发育不成熟(Chen et al., 2019)。ASD 儿童对儿向言语(Infant-Directed Speech, IDS)表现出非典型的听处理过程，IDS 未引起 ASD 儿童神经增强反应(Chen et al., 2021)。ASD 儿童对自己名字的弱回应也印证了 ASD 儿童较弱的言语声音刺激加工能力。研究表明，9 个月 ASD 婴儿更不容易对自己名字产生反应，这一现象一直持续到 24 个月(Miller et al., 2017)和 36 个月(Hatch et al., 2021)。ASD 高风险婴儿的研究也发现，6 个月的 ASD 高风险婴儿对言语声音刺激的回应也低于低风险婴儿(Paterson et al., 2019)。以上证据可推测，ASD 婴儿较少受成人照顾者言语的影响，缺失了类

言语发声学习的机会，进而影响他们类言语发声复杂性的发展。近期的研究表明，听觉皮层和言语运动皮层之间存在相互作用，言语听觉感知影响言语产生运动皮层的反应(Cheung et al., 2016)，言语运动也会影响言语感知皮层的反应(Daliri & Max, 2016)。ASD 儿童言语听觉皮层和类言语发声运动皮层之间的相互作用机制还需要进一步研究。

3.4 社会反馈导向理论

社会反馈环路(social feedback loop)理论在一定程度上解释了 ASD 儿童类言语发声复杂性较低的现象。该理论强调社会环境中的成人回应对儿童类言语发声发展的作用，认为成人更有可能对儿童的类言语发声做出回应，在成人立即回应后儿童更有可能产生类言语发声(Warlaumont et al., 2014)。Warlaumont 等人(2014)使用一种有很好稳定性和效度的、主要针对 0~4 岁儿童与成人交互发声的语言环境分析(Language ENvironment Analysis，LENA)软件(Harbison et al., 2018; Jones et al., 2019; Richards et al., 2017; Woynaroski et al., 2017)，对自然环境中 TD 儿童与成人互动中的发声进行了微观分析并证实该观点。研究者将 ASD 儿童与 TD 儿童发声作对比，发现 ASD 儿童社会反馈环路的有效性降低。具体表现在两个方面：一是，ASD 儿童发出较少的类言语发声；二是，成人对 ASD 儿童的回应取决于类言语发声的程度降低。那么随着时间的推移，社会反馈环路迭代次数减少，导致 ASD 儿童类言语发声进一步减少。

4 研究展望

4.1 探讨发声异常作为 ASD 儿童独特的早期识别标志的可能性

从上文的综述可知，与 TD 儿童相比，ASD 儿童早期发声确实存在异常现象。然而，这些发声特征也可能是其他障碍儿童的特征。例如，研究发现 9~12 个月的脆性 X 综合征婴儿的规范音节少于同龄 TD 婴儿(Belardi et al., 2017)。此外，几项同时包含了 ASD 儿童和发育迟缓(Developmental Delay, DD)儿童的研究结论不一致。有研究发现 ASD 儿童比 DD 儿童表

现出更少的沟通性发声(Lee et al., 2018)，而另有研究却未发现两类儿童在沟通性发声方面存在显著差异(Dow et al., 2020; Wetherby et al., 2004)。关于非类言语发声中的非典型发声，Wetherby 等人(2004)发现 ASD 儿童和 DD 儿童无差异，而 Lee 等人(2018)却发现 ASD 儿童比 DD 儿童表现出更多的非典型发声。因此，非常有必要开展大规模的、纳入其他障碍类型儿童的前瞻性研究，以探讨发声异常作为 ASD 儿童独特的早期识别标志的可能性。

4.2 加强哭声在 ASD 儿童早期筛查中的研究

如上文所述，非典型的哭声可能是 ASD 儿童的早期识别标志。然而，目前缺乏哭声在 ASD 儿童早期筛查研究中的实证数据。近期研究也发现，非典型的哭声可能影响 ASD 儿童的预后(Esposito et al., 2017)，因为照顾者对 ASD 儿童非典型哭声难以理解会影响护理质量及长期结果(Bornstein et al., 2016)。此外，成人也能较好地区分 ASD 儿童和 TD 儿童的哭声。例如，一项研究发现 ASD 婴儿和 TD 婴儿的父母均认为 ASD 婴儿的哭声更痛苦和更不典型(English et al., 2019)。因此，未来有必要也有可能开展哭声在 ASD 儿童早期筛查中的研究。在筛查研究中，可同时采用疼痛型和非疼痛型哭声样本。非疼痛型哭声样本可以参照以往的研究，在婴儿自然状态下收集(e.g., Bornstein et al., 2016)。疼痛型哭声可能是早期更为敏感的指标，也更有可能在标准化的环境下获得。例如，可以收集婴儿接种疫苗时诱发出的疼痛型哭声。加强哭声在 ASD 儿童早期筛查中的研究，不仅有利于明确哭声在早期筛查中的价值，也有助于早期干预。

4.3 探索基于最具预测性声学参数集的自动学习分类模型

正如前文所综述的，发声常用的分析指标为类言语发声复杂性、类言语沟通性发声、非类言语哭声基频等。近期也有研究采用非类言语发声或类言语发声的声学参数集合对 ASD 儿童进行分类预测(e.g., Khozaei et al., 2020; Pokorny et al., 2017, August)。交叉验证也显示，在区分 ASD 儿童和 TD 儿童时声学特征显示出良好的分类准确性(Pokorny et al., 2017, August;

Santos et al., 2013, October)。然而，这些研究均没有报道最具有预测性的声学信息。例如，Santos 等人(2013)在前瞻性研究中对 18 个月 ASD 婴儿和 TD 婴儿的呀呀语、哭声等发声的声学特征进行分析，并基于基频、共振峰、音强等 20 多种声学参数采用支持向量机(linear kernel support vector machines)和概率神经网络分类(probabilistic neural network classifiers)模型对儿童进行分类，分类准确率达 97%。Pokorny 等人(2017)采用回溯性研究，对年龄更小的 10 个月 ASD 婴儿呀呀语的声学特征进行分析，采用了包括基频、振幅和频谱及它们内部更具体的 88 个参数的标准声学参数集(Eyben et al., 2016)，并使用支持向量机和单层双向长短期记忆神经网络(1-layer bidirectional long short-term memory neural network)模型正确识别出了 75%的 ASD 婴儿。可见，研究者探究了不同类型和数量的声学参数对 ASD 的预测准确性，但最优预测声学参数尚不明晰。

最近研究还发现，声学参数集合在性别上可能存在差异。Khozaei 等人采用音色和音强相关声学参数对 10 名男性 ASD 儿童和 10 名男性 TD 儿童的哭声进行分类模型训练，形成子集实例(subset instance)自动学习分类模型；然后采用该模型再对 28~53 个月语言能力处在 12 个月左右的 ASD 婴幼儿(21 名)和 18~51 个月的 TD 婴幼儿进行了分类，结果发现对男性儿童分类的准确率比女性儿童高 7%(Khozaei et al., 2020)。此外，声学参数的元分析显示声学特征是 ASD 儿童非常有希望的早期识别标志，自动学习模型中多个声学参数比单个声学参数判别的准确率更高(Fusaroli et al., 2017)。因此，未来的研究可以构建具有最优预测效果的声学参数集合并建立自动学习分类模型，为 ASD 儿童的早期筛查提供智能的方法，从而使得早期识别的可能性更高。

4.4 深入探究动机对 ASD 儿童类言语发声的影响

上文综述指出，类言语发声是在内在动机驱动下，与成人社会互动过程中逐步发展的。虽然，近期有研究者开始尝试探究内在动机和社会动机对 ASD 高风险婴儿类言语发声发展

的影响(Long et al., 2021)。但总体上来说，该领域的实证研究极其有限，尤其是缺乏已诊断ASD 儿童的动机与类言语发声关系的实证数据。首先，未来可深入探究内在动机对 ASD 儿童类言语发声复杂性的作用机制。例如，ASD 儿童对特殊兴趣的内在动机是如何影响他们的类言语发声复杂性的？计算机建模作为一种自动化且高效的方法在 TD 婴儿类言语发声发展中得到应用，未来可以尝试采用通过计算机建模探究内在动机对 ASD 儿童类言语发声复杂性的影响。其次，探究社会动机如何对 ASD 儿童类言语发声复杂性产生影响。社会动机使得儿童注意社会信号、得到社会回报进而维持社会关系(Chevallier et al., 2012)，从而使儿童在社会互动中习得更接近言语的发声。ASD 儿童的社会动机缺陷以何种机制影响他们的类言语发声复杂性的呢？未来有必要继续开展相关研究。

4.5 进一步探究 ASD 儿童类言语发声异常的神经机制

如前文所述，神经运动系统不成熟及其与言语听觉感知皮层的相互影响，可能是 ASD 儿童类言语发声复杂性异常的重要原因。然而，ASD 儿童类言语发声的神经机制尚不清晰。未来 ASD 儿童类言语发声的神经机制可尝试从两个方面进行研究。一方面，可尝试从言语失用角度探究 ASD 儿童类言语发声异常的神经机制。失用症是一种影响言语所需动作协调能力的运动障碍(Akhtar et al., 2016)。研究表明言语失用(apraxia of speech)在 24~55 个月大 ASD 儿童中的患病率可能高达 60%(Tierney et al., 2015)。因此，最近有研究尝试将提示伴有言语失用的平均年龄为 56 个月 ASD 儿童和年龄匹配的言语失用儿童神经系统进行比较。结果发现言语失用儿童的对言语产生起重要作用的大脑结构存在异常，主要表现为顶叶(缘上回)和额叶(中央旁、三角部)皮质体积增加，而 ASD 儿童无这种现象(Conti et al., 2020)。但该研究中的 ASD 儿童具备了一定的口语能力，这可能会影响研究结果。未来可纳入无口语或口语较少的 ASD 儿童继续开展相关研究。另一方面，开展 ASD 儿童言语听觉皮层与类言语发声运动皮层之间相互作用的机制的研究。对 TD 婴儿的研究表明，类言语发声运动和听

觉感知之间的映射在婴儿学会说话之前已存在，3 个月婴儿的口部运动抑制对规范音节的听觉辨别有影响(Choi et al., 2021)。对 ASD 儿童该领域的深入探究可能能提供支持他们类言语发声发展神经机制的新证据。

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Potential early identification markers for children with autism spectrum disorder—Unusual vocalizations and theoretical explanations

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Abstract: Early identification and early intervention of children with autism spectrum disorder (ASD) are critical to their developmental outcomes. Vocalizations are sounds produced by children before they learn to talk. Studies suggested that unusual nonspeech-like and speech-like vocalizations are potential early identification makers for children with ASD before the age of 2 years. The theoretical explanations for unusual vocalizations mainly include the motivation orientation theories, the neuromotor orientation theories, the perceptual orientation theory, and the social feedback orientation theory. Future research may consider in (1) exploring the possibility of unusual vocalizations as unique early identification markers for children with ASD, (2) strengthening the study of crying in early screening of children with ASD, (3) constructing an automatic learning classification model based on the strongest predictive acoustic parameters, (4) analyzing the influence of intrinsic and social motivation on speech-like vocalizations in children with ASD, and (5) investigating the neural mechanisms of unusual speech-like vocalizations. These evidence may be helpful for early identification and intervention of children with ASD.

Keywords: autism spectrum disorder (ASD), potential early identification markers, unusual vocalizations, theoretical explanations