Gaps Exist in the Comprehensive Care of Children with Inflammatory Bowel Diseases

Hilary K. Michel, MD[1,2,*], Sandra C. Kim, MD[1], Nalyn Siripong, PhD[3], and Robert B. Noll, PhD[4]

Objectives To describe patterns of primary and specialty care delivery in pediatric patients with inflammatory bowel diseases (IBD), delineate which members of the healthcare team provided services, and identify gaps in care.
Study design Cross-sectional survey of parents of children (2-17 years) with IBD and adolescents with IBD (13-17 years) at a free-standing, quaternary children's hospital regarding healthcare receipt.
Results There were 161 parents and 84 adolescents who responded to the survey (75% and 60% response, respectively). The mean patient age was 14 ± 3 years, 51% were male, 80% had Crohn's disease, 16% ulcerative colitis, and 4% IBD-unspecified. Most parents were white (94%), living in a suburban setting (57%). Sixty-nine percent of households had ≥1 parent with a bachelor's degree or higher. Most had private insurance (43%) or private primary with public secondary insurance (34%). Most patients received annual check-ups (70%), vaccinations (78%), and care for minor illnesses (74%) from their primary care provider. Check-ups for gastrointestinal symptoms, IBD monitoring, and changes in type/dosing of IBD treatment were provided by their gastroenterology provider (77%, 93%, and 86% of patients, respectively). Discussions about family/peer relationships, school/extracurricular activities, and mood were not addressed in 30%-40% of participants. Adolescents frequently reported that no one had talked to them about substance use (40%), sexual health (50%), or body image (60%); 75% of adolescents and 76% of their parents reported that no one had discussed transitioning to an adult provider.
Conclusions There were gaps in the psychosocial care of pediatric patients with IBD. Coordinated, comprehensive care delivery models are needed. *(J Pediatr 2020;224:94-101)*.

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Inflammatory bowel diseases (IBD), including Crohn's disease, ulcerative colitis, and IBD-unspecified, are chronic, relapsing, remitting inflammatory conditions of the gastrointestinal tract. Up to 20% of patients with IBD are diagnosed at <20 years of age, making it a relevant pediatric and adolescent health problem.[1,2]

The goal of care for patients with IBD is to develop and maintain a proactive partnership between patients, families, primary care, and gastroenterology (GI) providers. This process theoretically would allow patients with IBD to receive both lifelong, disease-specific treatment, as well as routine primary healthcare services. Guidelines summarizing these services have been published for pediatric and adult patients, and provide roadmaps for healthcare providers.[3-11] However, most publications lack an indication of which providers are responsible for which services, and how they should best communicate and collaborate with the rest of the healthcare team, risking both duplications and gaps in care. The concern for variable, fragmented care is supported by work in adults with IBD suggesting preventive care use rates are significantly lower than those of peers without chronic illness, and in other pediatric chronic diseases, demonstrating significant unmet primary healthcare needs, especially in the realm of psychosocial care.[12-17] Gaps in psychosocial care in patients with IBD have been associated with increased healthcare use, poor treatment adherence, and decreased quality of life.[18-22]

The aims of this study were to describe the delivery of specific healthcare services to pediatric patients with IBD, delineate which members of the healthcare team provided services, and identify gaps in care. Understanding the current status of care receipt is an important first step in identifying the unmet needs of pediatric patients with IBD and developing comprehensive and coordinated models of care.

GI	Gastroenterology
IBD	Inflammatory bowel diseases
PCP	Primary care provider

From the [1]Division of Pediatric Gastroenterology, Hepatology, and Nutrition, University of Pittsburgh Medical Center (UPMC) Children's Hospital of Pittsburgh, Pittsburgh, PA; [2]Division of Pediatric Gastroenterology, Hepatology, and Nutrition, Nationwide Children's Hospital and The Ohio State University College of Medicine, Columbus, OH; and the [3]Clinical Translational Science Institute, and [4]Department of Pediatrics, University of Pittsburgh, Pittsburgh, PA

*Current Institution: Nationwide Children's Hospital, Columbus, OH.

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Methods

This cross-sectional, single-site, anonymous survey was conducted at a large freestanding children's hospital.

Participants were parents of pediatric and adolescent patients with IBD and adolescent patients. Inclusion criteria for parents included being a parent or legal guardian (referred to as parent[s] from this point forward) of a patient 2-17 years old diagnosed with IBD ≥12 months before recruitment. Inclusion criteria for adolescents included being 13-17 years of age, carrying one of the diagnoses as discussed elsewhere in this article, and having a parent who agreed to participate in the study. Exclusion criteria for both groups included the inability or unwillingness to consent/assent and respond to the survey in English. If a parent had >1 eligible child, they completed the survey in reference to the child who was diagnosed first.

We used a preexisting institutional pediatric GI research registry to identify participants. All parents of patients who met the inclusion criteria and were enrolled in our GI research registry as of March 2019 were recruited by phone, mail, or in person at the time of a routine clinic visit. After permission was obtained from a parent, eligible adolescents were invited to participate in a separate adolescent-specific survey. Parents of adolescents were not excluded if their child did not provide assent, but any adolescent who assented needed a parent to consent as well. There was no incentive for study participation.

After verbal consent/assent was obtained, participants completed the survey on a tablet computer or via a secure email link to the survey. A small number of participants completed paper versions of the survey and responses were transcribed by study staff. Study data were collected, managed, and stored anonymously using REDCap tools hosted at the University of Pittsburgh, Pittsburgh, Pennsylvania.[23,24]

Survey Data

Parent surveys assessed patient demographics and medical history, and details of primary and IBD-specific healthcare use in the past 12 months. Specifically, we queried participants about 18 different healthcare services and inquired who provided each service within the past 12 months. Response options included primary care provider (PCP), GI provider, both, other healthcare provider, or no one. Parental demographics were also collected.

Adolescent surveys also investigated details of primary and IBD-specific care received in the past 12 months with a focus on areas specific to teens (eg, discussions about body image, alcohol and drug use, mood, and sexual activity). The adolescent survey aimed to ensure capture of the full extent of services delivered to adolescent patients, including those without parent awareness. Survey questions were assessed for face validity by pediatric clinicians and researchers with expertise in IBD and primary/specialty care interactions. Readability was assessed at a fourth-grade level by the Flesh-Kincaid readability test.[25]

Statistical Analyses

Participant data were included in analysis if they responded to ≥10% of survey questions. Descriptive statistics were used to describe patient and parent characteristics, patterns of IBD and primary care, and specific care received. Continuous variables were described with means and SDs. Categorical variables were reported as frequencies with percentages. Differences between categorical variables were assessed using Pearson χ^2 tests or Fisher exact tests as appropriate.

Ethical Considerations

University of Pittsburgh Institutional Review Board approval and a waiver of written informed consent was obtained before study initiation: PRO17100303.

Results

Survey Response

There were 217 parents who met the inclusion criteria, were enrolled in the GI research registry, and were invited to participate in the study; 214 provided consent to receive the survey, 3 declined, and 161 responded to the survey for a parent response rate of 75% (161/214). Among 147 adolescents approached to participate in study, 140 provided assent to receive the survey, 7 declined, and 84 responded to the survey for an adolescent response rate of 60% (84/140).

Patient and Parent Demographics and Patient IBD Characteristics

Patient demographics and IBD characteristics are reported in Table I. (At the time of the study, our center was seeing approximately 500 patients with IBD in the 2- to 17-year age range.) Our cohort was 56% male; 76% had Crohn's

Table I. Patient demographics and IBD characteristics (n = 161)

Characteristics	
Current age, y	14 ± 3
Male sex	82 (51)
Disease type	
Crohn's disease	128 (80)
Ulcerative colitis	25 (16)
IBD-unspecified	7 (4)
Don't know	1 (1)
Duration of disease, y	4 ± 3
Current IBD treatment	
Aminosalicylates (5-aminosalycilate, sulfasalazine)	30 (19)
Immunomodulators (thiopurine, methotrexate)	61 (38)
Biologics	125 (78)
IBD-nutritional therapy (exclusive or partial enteral nutrition)	9 (6)
Steroids (budesonide or prednisone)	10 (6)
Antibiotics	1 (1)
No treatment	1 (1)
Patients with ≥1 other chronic condition*	47 (30)

Values are mean ± SD or number (%).
*n = 159; total N varied for different questions as no survey responses were required.

Table II. Parent demographics*

Characteristics	No. (%)
Female caregiver completing survey	133 (86)
Ethnic background	
Hispanic/Latino	2 (1)
African American/black	2 (1)
Asian/Pacific Islander	3 (2)
White	145 (94)
Mixed/multiple race/ethnicity	2 (1)
Living setting	
Rural	48 (31)
Urban	17 (11)
Suburban	88 (57)
Health insurance type	
Private only	66 (43)
Public only	32 (21)
Private primary with public secondary†	52 (34)
Prefer not say	4 (3)
Two-parent household	122 (79)
Highest household parental educational attainment‡	
Less than high school degree	0 (0)
High school degree or equivalent	11 (7)
Some college, no degree	15 (10)
Associate degree	22 (14)
Bachelor's degree	49 (32)
Graduate/professional degree	56 (37)

*n = 154 unless otherwise stated. Total number varied for different questions because no survey responses were required.
†In Pennsylvania, children with chronic illnesses like IBD are eligible for secondary public insurance.
‡n = 153, highest educational level completed by any parent in patient's household.

disease, 20% had ulcerative colitis, and 4% had IBD-unspecified. Parent demographics are presented in Table II.

Components of Medical Care Received and from Whom

All parents reported that their child had seen their GI provider at least once in the last 12 months, and the majority (141/161 [88%]) had seen their PCP in this time period. Figure 1 shows the proportion of patients who received a variety of healthcare services and from whom over the prior 12 months, per parent report. Figure 2 shows the responses from adolescents about receipt of adolescent-specific healthcare services over the same time period.

Some services seemed to more clearly fall into the jurisdiction of the PCP. For example, parents reported that their child's PCP performed annual checkups, administered routine vaccinations, and cared for minor illnesses 70%, 78%, and 74% of the time, respectively. In contrast, services like check-ups for GI symptoms, monitoring IBD activity, and changing type or dosing of IBD treatment appeared to be relegated to the responsibility of the GI provider (77%, 93%, and 86% of the time, respectively). Some topics, like puberty, were most frequently discussed by both the PCP and GI provider according to 34% of all parents, 38% of parents of children ages 8-15, and 47% of surveyed adolescents ages 13-15 (age ranges chosen as those in which conversations about puberty would seem relevant).

A final subset of services was frequently reported as not having been completed by any provider in the past 12 months. Some of these omissions may have been because the service was not indicated or required for the patient. For example, 41% of parents reported that their children were neither seen for another chronic condition nor referred to another physician in the past 12 months. Similarly, 18% of adolescents and 17% of their parents reported that no one had discussed puberty with them or their child in the past 12 months. However, conversations about puberty are likely only relevant in children approaching or progressing through puberty, and not those who have completed the process. Because we did not directly assess the degree of pubertal development in this study, we used patient age as a proxy. When we focused on parents of children 8-15 years old (n = 106) and adolescents ages 13-15 years old (n = 45), only 16% and 11% reported puberty had not been addressed, respectively. Other findings, however, are more concerning: 75% of adolescents and 76% of their parents reported that no medical provider had discussed the process of transitioning to an adult provider, a regularly accepted component of care for patients with a chronic disease. Despite nearly all parents (96%) having children who were school age or older, >40%reported that their child had not discussed family relationships, peer relationships, academic performance, extracurricular activities, or mood with a medical provider in the last 12 months. Because mood may be assessed differently in younger children compared with adolescents, we compared the reports of parents of 6- to 12-year-olds with those of 13- to 17-year-olds, but found similar percentages reporting gaps in care (43% vs 40%, respectively).

Adolescent participants also reported gaps in care; >60% said no one had discussed body image with them; >40% had not discussed family or peer relationships, sex and sexuality, or drug and alcohol use. More than 30% had not discussed academics, extracurricular activities, or mood with any medical provider in the prior 12 months.

Although some services differed between parent and adolescent surveys, a subset of topics was addressed by both parent and adolescent surveys. Of the 80 parents and adolescents who responded to these questions, complete agreement on which provider addressed topics occurred with the following frequencies: transition to adult care 74%, peer relationships 56%, school/grades 55%, mood 49%, family relationships 49%, extracurricular activities 48%, and puberty 41%. Parental and adolescent reports of gaps in service delivery were within 5% of each other, except for discussions about extracurricular activities, where 30% of adolescents reported this gap compared with 41% of parents.

Gaps in Psychosocial Care by Parental Education and Insurance Type

For services reported as missing by ≥30% of parents or adolescents, we assessed for differences in care delivery by parental education (highest household educational attainment of bachelor's degree or more vs less than a bachelor's degree) and insurance type (public only vs private or multiple

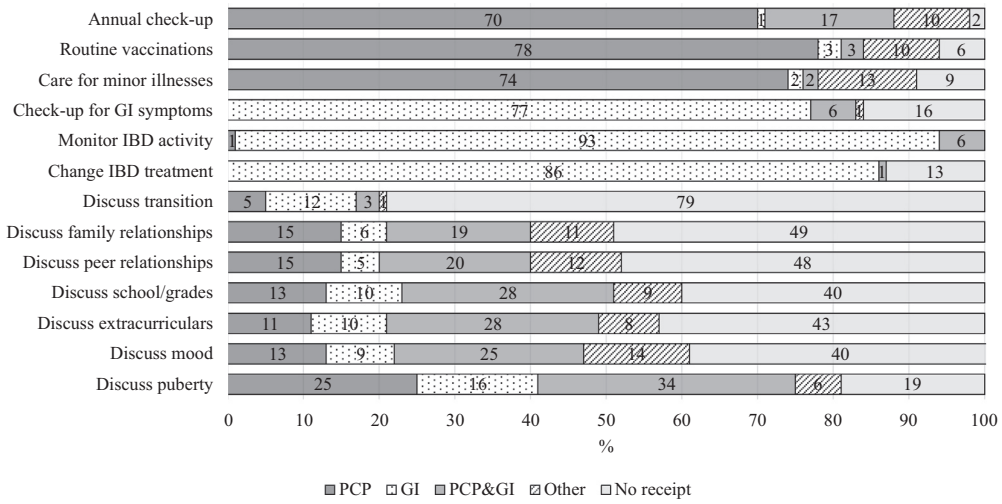

Figure 1. Services received in last 12 months per parent report (total n = 159).

types [includes private and private primary with public secondary insurance]). There were no significant differences in parent report of discussions about family or peer relationships, school/grades, extracurriculars, mood, or transition to adult care by parental education or insurance type. A significantly smaller percentage of adolescents with public insurance reported discussing peer relationships (39% vs 71%; $P < .05$) compared with those with private or multiple types of insurance. The same pattern was seen regarding discussions about family relationships (52% vs 79%; $P < .10$), although this did not reach statistical significance. Otherwise, there were no significant differences in discussions about school/grades, extracurriculars, mood, body image, sex, alcohol/drug use, or transition between groups.

Discussion

We describe the receipt of primary and IBD-specific care in pediatric and adolescent patients with IBD and demonstrate that certain healthcare services, including annual check-ups, provision of routine vaccinations, and care for minor illnesses, were almost always performed by patients' PCPs. Other services, including check-ups for GI symptoms,

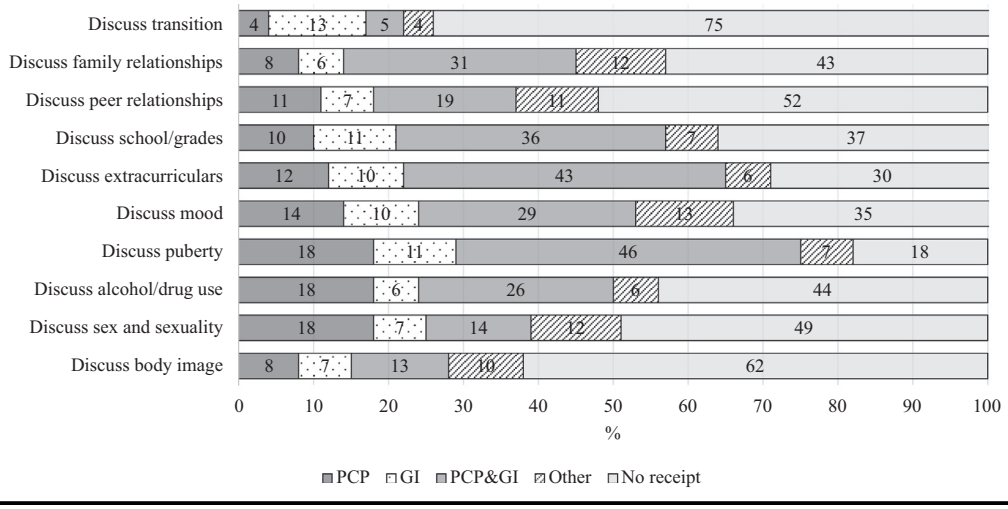

Figure 2. Services received last 12 months per adolescent report (total n = 84).

monitoring of IBD activity, and changing IBD therapies, were almost always performed by patients' GI providers. A small number of topics, including pubertal development and extracurricular activities, were discussed by both the PCP and GI provider. However, a large portion of the services we inquired about, many related to psychosocial care, were frequently left unaddressed. In addition to being essential for holistic, comprehensive care, addressing psychosocial needs in patients with chronic diseases can improve a variety of health outcomes, including treatment adherence and quality of life.[26-29]

In pediatric patients with IBD, gaps in addressing psychosocial topics are worrisome given the wide-reaching effects of the disease and its therapies. For example, children with IBD are at an increased risk for missing school owing to medical appointments and symptoms, and those with greater disease activity are more significantly affected.[30] The data are mixed on the impact of IBD on academic performance, with some studies supporting equal or greater academic achievement than controls, and others suggesting poorer performance.[31-35] Our data emphasize the importance of screening for potential academic issues, especially in sicker patients.

A pediatric IBD diagnosis can also cause significant stress in the home. In 1 study, 25% of families of children with IBD endorsed difficulties across family functioning dimensions, such as problem solving and communication.[36] Additionally, children in families where parental distress was greater report decreased health-related quality of life.[37] A healthy family environment and good social support system can be a protective psychosocial factor for IBD patients, even in the setting of increased disease activity.[38] Assessing family function, potentially with brief screening instruments like the Psychosocial Assessment Tool or the distress thermometer for parents would allow us to identify opportunities for intervention in the form of support services and behavioral healthcare, potentially optimizing both medical and psychosocial outcomes.[39,40]

Peer relationships and social functioning can be affected in children and adolescents with IBD as well. Youth with IBD have reported lower social functioning compared with youth without chronic illness by various measures, and the impacts of IBD on social functioning may be even greater in patients diagnosed in adolescence compared with those diagnosed in childhood.[41,42] This may be partially explained by the fact that IBD has the ability to significantly impact patients' physical appearances during vulnerable developmental phases. This is even more concerning because nearly 20% of adolescents and their parents reported that no one had talked about pubertal development with them in the last 12 months, a topic that is considered standard of care in both general pediatrics and IBD care.[4] Associations between IBD and eating disorders have also been described.[43] Of the adolescents surveyed in this study, 52% had not discussed peer relationships and 62% had not discussed body image with a provider in the last 12 months. Developing sensitive, efficient screening tools to assess social functioning and body image is important, because several interventions such as cognitive behavioral therapy, peer support groups, and IBD camp attendance have been shown to increase social functioning and quality of life, respectively.[26]

The impact of IBD on social functioning may also be related to an increased risk for anxiety, depression, and chronic pain, during both disease quiescence and flare.[44,45] Recent studies have suggested that those diagnosed with IBD at <6 years of age are at greater risk for behavioral health disorders than those diagnosed later, and have also reported an association between IBD diagnosis and increased risk for suicide attempt.[46,47] However, more than one-third of patients and parents in our study reported that no one had talked with them or their child about mood in the last 12 months. When we examined only the responses of parents of adolescent patients (compared with parents of patients <13 years of age), this percentage remained unchanged, despite the US Preventive Services Task Force recommending annual depression screening for all children 12-18 years of age.[48] It is notable that a similar percentage of adolescents and their parents reported gaps in discussing mood, despite it being a confidential topic. Building systems to confidentially screen for anxiety and depression, and securely share findings between primary and specialty care teams will be essential for appropriate referral and evidence-based treatment of children with psychosocial needs.

Nearly 50% of adolescents reported that no healthcare provider had discussed sexuality and sexual activity with them in last 12 months. Patients with IBD are frequently on immunosuppressive therapies, some which also are teratogenic (ie, methotrexate). A well-informed decision to start any medication should include thorough conversations about the risks and benefits of the therapy, and in the case of teratogenic medications, address sexual activity and contraception. Additionally, IBD and its treatments can affect sexual function, satisfaction, and fertility in both males and females.[49,50] As we prepare teenage patients for self-management as adults, discussions about sexual health, especially in those on teratogenic medications, should be routine.

Similarly, 44% of our adolescent participants reported that alcohol and drug use had not been addressed in the last 12 months. Several IBD therapies can interact with alcohol, yet teens with IBD have exhibited poor knowledge of these effects.[51,52] Moreover, alcohol use has been associated with treatment nonadherence.[53] Marijuana use is also common in pediatric and adolescent patients with IBD, with a recent study finding 32% had ever used marijuana and 9% used it daily.[54] Although no definitive data exist to prove the efficacy of cannabis in healing inflammation in IBD, inquiries about its benefits are unlikely to slow, because studies have demonstrated improved symptoms and quality of life in adults with IBD who use marijuana.[55]

Finally, 75% of adolescents and 76% of their parents reported they had not discussed the transition process with a provider in the preceding 12 months. These findings are similar to those recently published from a nationally representative sample of youth with special healthcare needs; in this study, only 17% of >5000 youth received transition planning support in the form of discussions about transition, education, and skills building around self-care, and the opportunity to meet alone with a healthcare provider.[56] A coordinated transition process, where adolescents gradually gain the skills and knowledge needed to manage their healthcare independently can lead to improved physical and emotional health outcomes.[56,57]

The lack of comprehensive care for children and adolescents with chronic disease is not a new finding. Carroll et al conducted a survey of adolescents with chronic disease (including IBD) and found that, of the 56% of patients who reported psychosocial problems attributable to their chronic disease, only one-half had spoken with their physician about their concerns.[14] In our study, the services that are more clearly primary care or GI related seem to be addressed with more regularity, whereas most psychosocial topics remain largely unaddressed. Perhaps this is evidence of failed patient comanagement; if providers do not know the tasks for which they are responsible, a lack of ownership may result. Fragmented care and inadequate communication between providers may also contribute to the problem. Several studies in adults suggest that PCP-specialist comanaged care is associated with improved outcomes compared with either provider acting alone.[58,59] For this reason, perhaps a typology of specialists' clinical roles would improve the way providers set expectations for all involved parties, improving comprehensiveness of care and decreasing unneeded redundancy.[60] A lack of resources and time to address psychosocial topics may also be contributing, as documentation in the electronic medical record takes up more and more provider time.[61] A better understanding of the current roles played by nurses, dieticians, social workers, behavioral health specialists, peer and community support groups, and investigating inventive ways to incorporate and clearly communicate their expertise will also be key. Reassuringly, gaps in care were not significantly associated with proxies for socioeconomic status such as insurance type or parental education, although we realize our population was quite homogenous.

As with all survey research, results were susceptible to ascertainment and selection biases. Although study participant sex and IBD type seemed to be reflective of our overall IBD population, we were unable to compare other variables between the overall population and our sample, such as IBD therapies and socioeconomic variables. Our results may have been affected by recall bias, although we attempted to mitigate this by restricting recall of care to specific venues in the preceding 12 months. This was a single-site study in a large, quaternary care pediatric hospital, and participants were primarily white and highly educated, which may limit generalizability. Clearly, similar work needs to be done in other settings with more diverse populations. ■

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Reprint requests: Hilary K. Michel, MD, Nationwide Children's Hospital, 700 Children's Dr, Columbus, OH 43205. E-mail: hilary.michel@nationwidechildrens.org

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50 Years Ago in *The Journal of Pediatrics*

Congenital Cytomegalovirus Infection: Can It Recur in a Sibling?

Embil JA, Ozere RJ, Haldane EV. Congenital cytomegalovirus infection in two siblings from consecutive pregnancies. J Pediatr 1970;77:417-21.

Embil et al reported congenital cytomegalovirus (CCMV) infection in subsequent infants of a young mother. They challenged the widely accepted notion at that time that CCMV resulted from viral transmission when mothers had primary infection during pregnancy.

Fifty years later, the factors affecting in utero transmission of CMV are still not well-elucidated. CCMV can occur even if the mother has CMV antibodies during pregnancy owing to past infection (nonprimary or recurrent infection). However, the rate of transmission in utero is much higher for primary infections.[1] Nonprimary maternal infections can occur either owing to reinfection by immunologically variant (envelope glycoprotein epitopes) strain or reactivation of latent infection. There is uncertainty as to what triggers the reactivation and transfer of virus in a previously immune mother, so most such CCMV infections are considered to be due to reinfection. Primary and recurrent maternal infections are differentiated based on a lack of CMV IgM antibody and presence of high-avidity IgG antibody (indicating maturity), but the reliability of this is questionable.[2] In CCMV owing to recurrent infection, the placenta is thought to induce local immunosuppression in the uterus, triggering reactivation of latent virus in macrophages ($CD14^+$ monocytes), which then transmits to the fetus despite persistent maternal immunity.[3]

The complex nature of protective response to CMV (involving both innate and humoral immunity), capability of CMV to evade host immune responses, widely prevalent immunologically variant strains, and a high possibility of reactivation of latent infection pose crucial challenges in the development of an effective CMV vaccine. Prenatal screening using maternal serology for CMV is not routinely recommended owing to the lack of approved therapeutic prenatal interventions for prevention or treatment of CCMV after primary maternal infection. Nonprimary maternal CMV infections still pose a diagnostic challenge owing to the lack of specific virologic or immunologic markers, and preventive modalities targeting the same are still a work in progress.[1]

Pinky Meena, MD
Piyush Gupta, MD, FAMS
Department of Pediatrics
University College of Medical Sciences
Delhi, India

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