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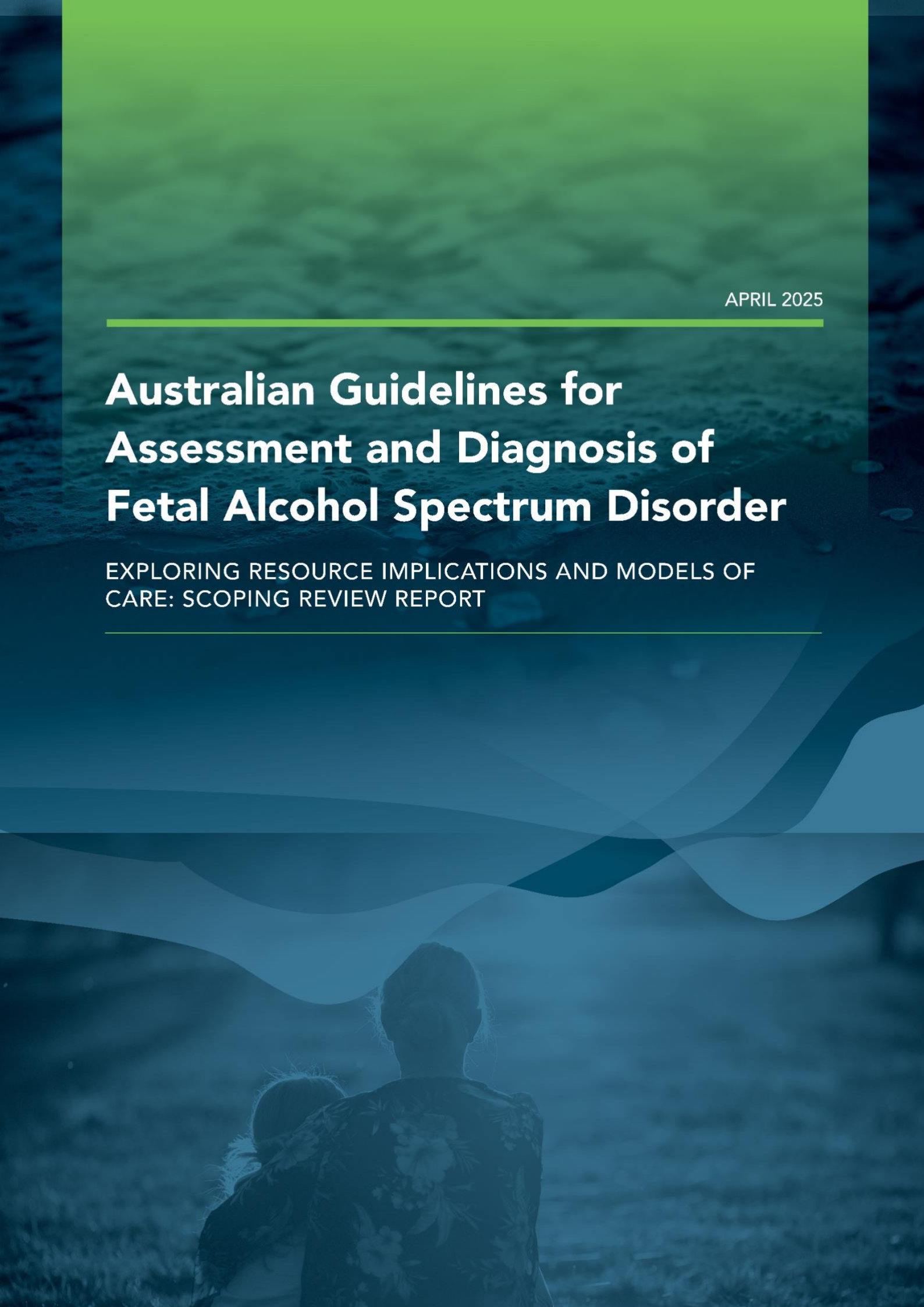
Australian Guidelines for Assessment and Diagnosis of Fetal Alcohol Spectrum Disorder

EXPLORING RESOURCE IMPLICATIONS AND MODELS OF CARE: SCOPING REVIEW REPORT



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Contents

Authors and contributions

Nykola Kent	Wrote the systematic review report with guidance and contributions from other authors, contributed to study selection, data charting and undertook the qualitative synthesis.
Nicole Hayes	Critical review of the protocol and report and contributed to study selection.
Sophia Young	Contributed to study selection and critical review of the report.
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Natasha Reid	Shared responsibility for leading the design, conduct and reporting of the review. Guidance during all stages of the review and contribution to writing of this report. Contributed to study selection and reviewed data charting and synthesis. Oversight and communication with the Project Steering Committee regarding development of the protocol.

Declarations of interest
All authors declare they have no personal, financial, or professional interests that could be interpreted to have influenced conduct or results of this review.

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Summary: Exploring resource implications and models of care for assessment and diagnosis of fetal alcohol spectrum disorder: A scoping review

What is the problem?

Previous reviews have examined various costs associated with fetal alcohol spectrum disorder (FASD), including annual mean costs of care, health burden costs, justice system costs, productivity losses for caregivers, and both the monetary and non-monetary costs of reduced quality of life. However, there no previous reviews focused on understanding the resource implications and models of care for the assessment and diagnostic process.

What is the importance?

This review offers important preliminary insights into the resource implications and models of care involved in assessment and diagnosis of FASD, filling a gap in the existing literature.

What are the key findings?

A total of 11 studies were included in the final qualitative synthesis. The primary patient costs were attributed to the lengthy time required for diagnosis (up to 47 hours). The primary service costs were linked to costs of clinicians and support personnel, as well as the involvement of multi-disciplinary teams in the assessment process. Estimates of the diagnostic costs varied and were limited across studies. Several models of care, mainly from Canadian clinics, were examined. These models aimed to capitalise on available services to improve patient care and reduce service costs. This study provides important preliminary insights into the costs and key service features involved in the diagnostic assessment of FASD. However, the limited number of available studies and variability in data highlights the need for formal costing studies.

1. Background and rationale

Internationally, multiple guidelines to inform assessment and diagnostic practices (e.g., Astley 2013; Bower & Elliott, 2016; Cook et al., 2016; Hoyme et al., 2016). Implementing these guidelines is likely to incur high costs due to the common recommendation that assessments involve health professionals from multiple disciplines (e.g., paediatrician, psychologist, occupational therapist, and speech pathologist). However, to the author's knowledge there have been no reviews to understand the costs of providing assessment and FASD diagnostic services.

Previous studies have estimated the costs of FASD to the health system (Fuchs et al., 2009) and justice system in general (Popova et al., 2015), productivity losses for caregivers (Ericson et al., 2017), and the cost of reduced quality-of-life, measured using both a monetized and nonmonetized quality adjusted life year approach (Miller et al., 2006; Roosen et al., 2008), have been estimated. For example, a systematic review estimated the annual per-person costs of care to be $24,308 USD per adult and $22,810 USD per child based on cost estimates from 32 studies in 4 countries (United States, Canada, New Zealand and Sweden; Greenmyer et al., 2018). However, these data are not specific to the costs required to support assessment and diagnostic procedures.

Access to specific information regarding the resource implications of assessment and diagnosis of FASD guide policy and practice developments and support allocation of public health funding. For example, in 2018, the Australian Commonwealth Government invested $7.2 million AUD to help state governments, service providers, and communities address the prevention, screening and diagnosis of FASD, as well as its support and management, and in tailoring service needs to communities (Australian Government Department of Health and Aged Care, 2018). However, there are no data available to validate the success of this commitment (Panton et al., 2023), the extent to which this funding was helpful, and the true costs of providing the model of care stipulated by the current Australian Guide to Diagnosis (Bower et al., 2017).

2. Objectives

The current scoping review is part of a broader project to review and update the Australian Guide for Diagnosis of FASD (Hayes et al., 2022; Hayes et al., 2023; Hewlett et al., 2023; Reid et al., 2023). In line with the Australian National Health and Medical Research Council standards for clinical practice guidelines (NHMRC, 2011) the evidence review process should include identifying evidence related to cost-effectiveness and resource implications of clinical practice. This scoping review aimed to investigate the available evidence examining the costs and key service features involved in the assessment and diagnostic procedures that can consider FASD as one possible outcome. Additionally, where available results regarding successes and challenges of available models of care were also summarised.

Research question

What are the cost and resource implications to be considered when undertaking assessments that can consider FASD as one possible outcome?

3. Methods

3.1 Protocol and registration

The current scoping review was registered with Open Science Framework (OSF; osf.io/58gnh) and reported according to the Preferred Reporting Items for Systematic Review and Meta-Analyses

extension for Scoping Reviews (PRISMA-ScR; Tricco et al., 2018). A scoping review was selected as the most appropriate synthesis type due to the focus on identifying the available types of evidence, clarifying key concepts and definitions, mapping the key resource implications and models of care reported in these studies, identifying and analysing current knowledge gaps, and reporting the key implications for practice and policy (Munn et al., 2018, Tricco et al., 2016).

3.2 Eligibility criteria

No restrictions were placed on publication year, study location, study designs or languages. Studies were restricted to peer-reviewed publications.

3.3 Information sources

Studies were identified through a systematic search of six electronic databases (PubMed, EMBASE, Web of Science, PsycINFO, Cochrane Library, CINAHL), completed on the 15th of February 2021. Reference lists of included studies were additionally searched to identify any relevant articles. An updated search was completed on 6th of December 2022.

3.4 Search

Search terms were either alcohol-related (e.g., “prenatal alcohol” or “fetal alcohol spectrum disorder” or “fetal alcohol syndrome” etc) or cost/resource-related (e.g., “cost-effectiveness analysis” or “cost-benefit” or “economic impact” etc.). Appendix A details the full search strategy for each electronic database. Initial search and removal of duplicates was undertaken by SY.

3.5 Selection of sources of evidence

Inclusion criteria were peer-reviewed studies focused on the potential costs and/or resources associated with undertaking diagnostic assessments for FASD. Studies focused on the direct costs of assessment and diagnostic service provision, resource considerations in development or delivery of services providing assessment and diagnosis of FASD, and development and/or comparison of different types of models of care/clinical models of service delivery specific to assessment and diagnosis of FASD.

Exclusion criteria were: (1) wrong type of publication (e.g., conference abstracts, books, theses, reviews, commentaries, clinic reports, government reports); (2) wrong topic (e.g., alcohol use generally not during the prenatal period); (3) not related to assessment/diagnosis; and (4) not related to cost of resources associated with assessment/diagnosis. Two independent reviewers screened the titles and abstracts and full texts (NH, SY) in Covidence, and a third reviewer (NR) resolved conflicts.

For this review, costs were defined by two different categories: 1) service costs, including staffing resources, training resources, travel to communities, testing materials, and 2) patient costs, including travel, time off work, and childcare.

3.6 Data charting process

Information pertaining to study author (date), study location (country and context), study design, study aims, the number of clinics surveyed, and key data collected/measures was summarised. Data were charted by three authors (SY, DS and NK).

3.7 Synthesis of results

Content analysis (Cavanagh, 1997, Downe-Wamboldt, 1992) was utilised to synthesise results. Content analysis was selected to support identification of the characteristics of the costs and models of care considered by the included studies (Pollock et al., 2023). In general, content analysis involves coding and grouping available text into similar groups and then counting the number of times each of the groupings occurs (Hsieh and Shannon, 2005). The content analysis process for the current study was both inductive and deductive. The two deductive areas of interest from the research question (i.e., costs and models of care) were categorised based on information contained within each of the included studies.

Information on resource implications related to patient and service costs was summarised under the following subheadings: patient time, professional time required for diagnosis, multidisciplinary teams, estimates of cost of diagnosis, estimates of clinical capacity and how different diagnostic programs accessed funding (Appendix B). Information included in studies that investigated models of care and the associated successes/advantages and losses/challenges of each model were summarised separately to the studies related to costs (Appendix B). Some studies addressed multiple areas of interest (i.e., costs and models of care) and these were included in each area. Following the coding of each interest area (i.e., costs and models of care) frequencies of the studies addressing each type of cost and models of care were calculated. The content analysis was undertaken by one author (NK) and checked by a second author (NR).

4. Results

4.1 Search results

A total of 1,805 records were identified through a search of six databases, and four additional records were identified from reference list searching. Following removal of duplicates and screening of titles and abstracts, 51 articles were eligible for full-text screening. After exclusion of 43 records, and addition of three studies identified in an updated search, 11 studies were included in the final qualitative synthesis (Figure 1).

4.2 Characteristics of sources of evidence

Of the included studies, eight were conducted in Canada, one in Australia and two were international surveys. Six of the studies were based on information collected through questionnaires and surveys of FASD clinics (Clarren & Lutke, 2008; Clarren et al., 2011; Dugas et al., 2022; Peadon et al., 2008; Panton et al., 2022; Reid et al., 2022a), three studies were summaries or critical reviews of specific FASD clinics (McFarlane, 2011; McFarlane & Rajani, 2007; Temple et al., 2015), one was a retrospective chart review of clinical data (Patel et al., 2019) and one study reported a non-experimental cost estimation model (Popova et al., 2013). An overview of the included studies can be found in Appendix B and a visual summary of the included studies, and the sub-topics addressed by the included studies can be found in Figure 2.

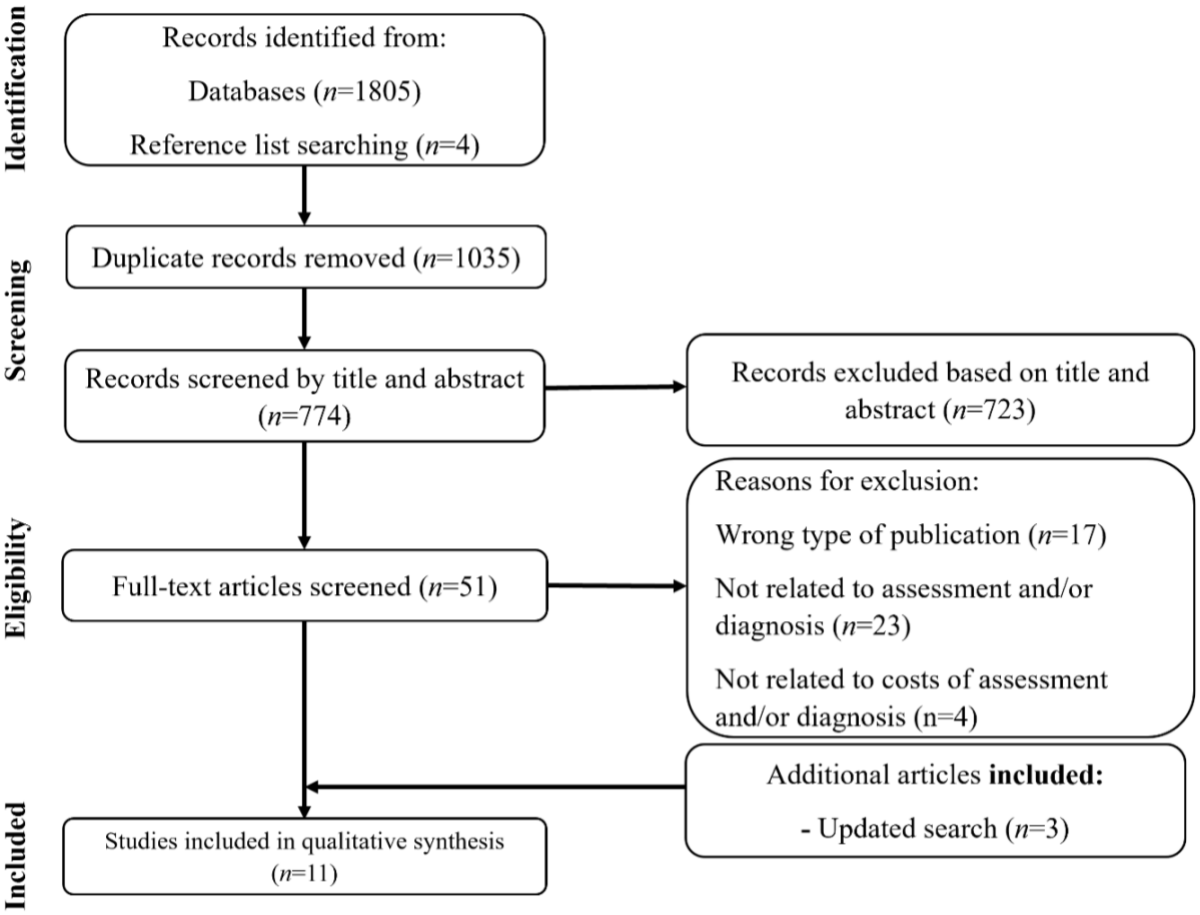

Figure 1. PRISMA flow chart of selection process for included studies.

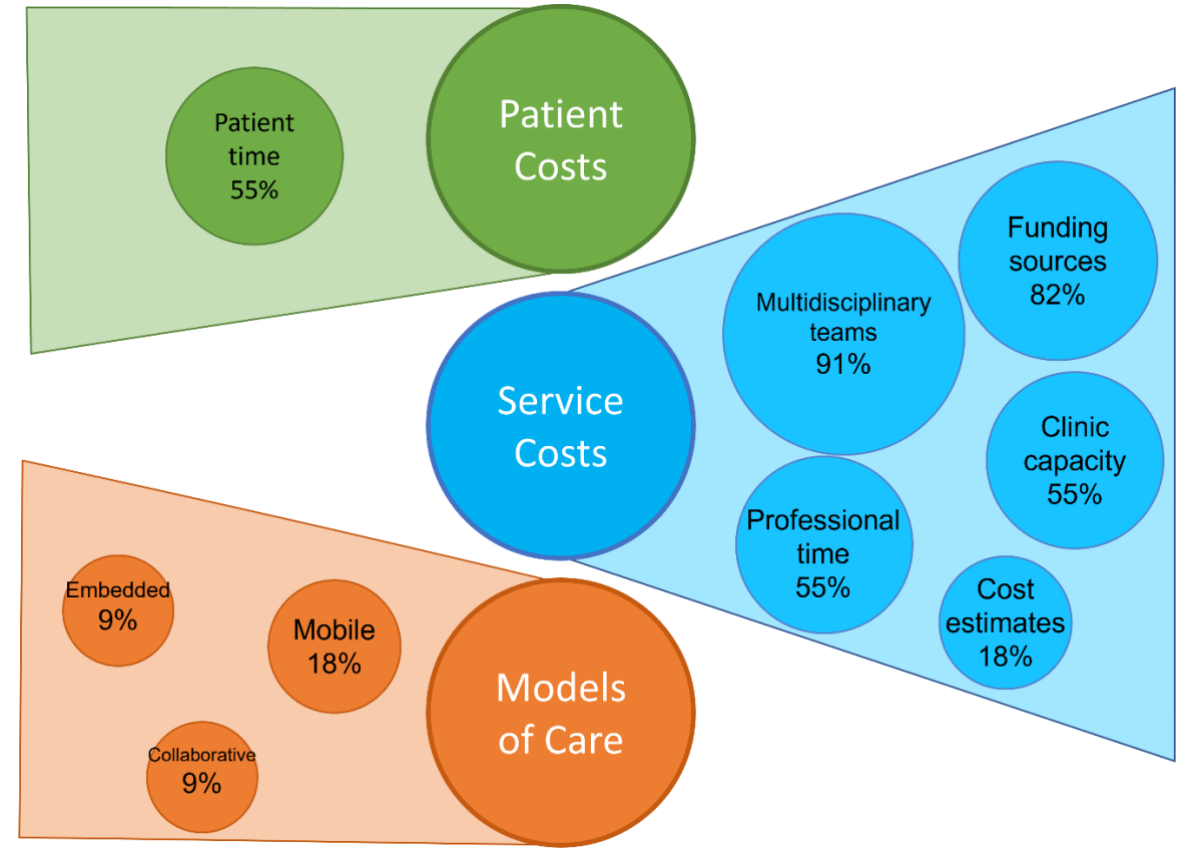

Figure 2. Summary of the patient costs, service costs and models of care addressed by the included studies. The size of the bubble represents the percentage of papers that addressed each sub-topic relative to the total number of papers included in the scoping review

4.3 Synthesis of results

4.3.1 Patient Costs (*n*=6)

Six studies provided estimates of the patient time required for diagnosis, which could be translated to time taken off work leading to loss of income (if employed) and time required for child-care. The estimates of time required by patients for diagnosis ranged between 0.5 hours and 47 hours (Clarren & Lutke, 2008; McFarlane & Rajani, 2007; McFarlane, 2011; Peadon et al., 2008; Popova et al., 2013; Temple et al., 2015). The lowest estimate was provided from an international survey of 34 clinics that focussed specifically on the diagnosis of children, and the highest estimate was provided by a collation of all data available on FASD diagnostic clinics in Canada (Peadon et al., 2008; Popova et al., 2013). A mobile diagnostic team in Canada that provided diagnostic services to rural communities estimated that the diagnostic assessment for a child required four hours, while adult diagnostic assessment required a full day (McFarlane, 2011; McFarlane & Rajani, 2007). Another study estimated that four hours was required for adult diagnosis, if the patient was able to tolerate all of the required tests in one sitting (Temple et al., 2015). Patient wait times were also estimated to range between 3 and >6 months for some clinics (Clarren & Lutke, 2008; McFarlane, 2011; McFarlane & Rajani, 2007).

4.3.2 Service Costs

The key contributors to service costs that were reported in available studies included the professional time required for diagnosis, which was addressed by six studies, and the need for a multidisciplinary team (MDT), which was addressed by ten studies. Some studies also explored how these factors affected clinical capacity, provided estimations of cost, and provided examples of how clinics access funding.

4.3.2.1Professional Time Required for Diagnosis (*n*=6 studies)

Several studies provided estimates of the time required for diagnostic assessments, which while important for estimates of patient time and costs, also translated to professional time and thus, service costs. Estimates of direct patient care and indirect patient care time were provided in six of the included studies (Clarren & Lutke, 2008; McFarlane, 2011; McFarlane & Rajani, 2007; Peadon et al., 2008; Popova et al., 2013; Temple et al., 2015). Typically, studies only provided estimates of the time required for diagnostic assessments, which was stated to range between 0.5 hours and 47 hours, depending on the age of the patient (Clarren & Lutke, 2008; McFarlane, 2011; McFarlane & Rajani, 2007; Peadon et al., 2008; Popova et al., 2013; Temple et al., 2015). Clarren and Lutke (2008) provided estimates of the time commitment of each clinician involved in the diagnostic process, which included a total of 12.5 hours of direct patient care (2.5 hours for Paediatrics, 2.5 hours for Speech and Language Pathology, 1.5 hours for Occupational therapy and 6 hours for Clinical Psychology) and 13.5 hours of indirect patient care (3 hours for Paediatricians, 3 hours for Speech and Language Pathologist, 3 hours for Occupational Therapists, 4.5 hours for Clinical Psychologists). Time for indirect patient care included chart review, team discussion without clients scoring of tests and report preparations, consultations with primary physicians, and referrals, totalling 26 hours of total staff time for diagnosis of one patient (Clarren & Lutke, 2008). It is important to note that this did not include liaison with any external agencies (i.e., schools). Another study conducted in Australia estimated that running an MDT FASD diagnostic clinic required approximately 20 hours of clinical time and 10 hours of administration/coordination time, totalling 30 hours of staff time per assessment (Panton et al., 2022).

4.3.2.2 Multidisciplinary Teams (*n*=10 studies)

The primary cost of FASD diagnostic clinics stemmed from the diverse range of professionals involved in the assessment process. The efficacy and necessity of an MDT in FASD diagnostic clinics and services was

explored in ten studies (Clarren & Lutke, 2008; Clarren et al., 2011; Dugas et al., 2022; McFarlane, 2011; McFarlane & Rajani, 2007; Panton et al., 2022; Peadon et al., 2008; Patel et al., 2019; Popova et al., 2013; Temple et al., 2015). Common MDT members included medical doctors (i.e., family physicians or paediatricians), psychologists (i.e., neuropsychologist or clinical psychologists), speech-language pathologists, occupational therapists, social workers, and team/patient/site coordinators (Clarren & Lutke, 2008; McFarlane 2011, McFarlane & Rajani, 2007; Panton et al., 2022; Temple et al., 2015). Additional team members included psychiatrists, geneticists, nurses, nurse practitioners, education specialists, behaviour therapists, Aboriginal Liaison Officers, addictions counsellors, disability services co-ordinators, legal representatives, post-diagnostic outreach workers, family advocates and general support staff (i.e., secretary/clerical workers; McFarlane, 2011; Popova et al, 2013; Temple et al., 2015). Sourcing individuals within these professions who were appropriately trained was reported as a major challenge for FASD specialist clinics and services across several locations and models of care (Clarren et al., 2011; Dugas et al., 2022; McFarlane, 2011; McFarlane & Rajani, 2007; Panton et al., 2022). Additionally, hiring MDT staff and providing appropriate training and resources represented a major service cost (Clarren et al., 2011; McFarlane & Rajani, 2007; Popova et al., 2013).

Despite the high cost of an MDT, an international study conducted in Canada reported that 33 of the 34 sites surveyed had an MDT, with 32 of 34 reporting they had a minimum of one staff member with specialist FASD training (Peadon et al., 2008). A self-administered survey of FASD diagnostic clinics in Canada reported that all clinics employed a clinician and co-ordinator as per the Canadian Guidelines for FASD diagnosis, and 90% used a "multidisciplinary approach" for FASD diagnosis. However, funding restrictions meant that only 46% of clinics reported access to all the specialists required for the evaluation of neurodevelopment, including psychologists, occupational therapists, and speech pathologists (Clarren et al., 2011). Inadequate funding, or funding by time-limited research or government grants, was a commonly cited problem within FASD diagnostic clinics, which led to a significant burden being placed on clinical and administrative teams and often, less than a full team being available on-site (Clarren et al., 2011; Panton et al., 2022; Peadon et al., 2008). Dugas et al. (2022) provided a breakdown of the proportion of clinics that use an MDT to diagnose their patients. In children <18 months old, 4/7 clinics used an MDT, while 15/25 clinics used an MDT for diagnostic assessment of children between the ages of 18 months and 5 years, and 32/40 clinics used an MDT for diagnostic assessment of children and adolescents between 6-18 years of age.

The need for an MDT was found to be especially challenging for rural communities. A mobile support team required a large MDT consisting of community-based professionals contributed from local agencies as part of their usual care workload who had received appropriate training and were able to dedicate one day per month to the clinic (McFarlane, 2011; McFarlane & Rajani, 2007). These professionals received a fee-for-service payment, which reduced patient costs but contributed to increased service costs (McFarlane, 2011).

The difference in the service needs for the diagnosis of children and adults and the effect that this may have on service costs was briefly explored in two studies (McFarlane & Rajani, 2007; Temple et al., 2015). McFarlane et al. (2007) noted that there are differences in the diagnostic teams and follow-up personnel required for children and adults based on their specific needs contributing to differences in service costs. For example, a teenage client who has experienced involvement with the justice system may have a probation officer invited to be part of their diagnostic team. Furthermore, Temple, Ives and Lindsay (2015) noted that additional staff might be required for adults who are undergoing assessment if they do not have a caregiver.

4.3.2.3 Clinical Capacity (*n*=6 studies)

Clarren and Lutke (2008) provided estimates of the diagnostic capacity of Western and Northern Canada across 15 programs, stating that there were 1,140 patient assessment slots, with only three programs routinely providing assessments to adults. In a follow-up study, Clarren, Lutke and Sherbuck (2011) surveyed 39 clinics in Canada and estimated that 2,200 diagnostic slots were available at the time of publication, with less than 10 clinics stating that they were able to see adults. The authors noted that a 17-fold increase is needed to meet the demand for diagnosis, as they estimated that 37,000 individuals in Canada were living with FASD. A more recent study by Dugas et al. (2022) focussed on clinical capacity for the diagnostic assessment of children and adolescents across 41 clinics in Canada and reported that although 2,537 referrals were received ever year, only 1,797 diagnostic slots were available, equating to a clinical capacity of 71%. Smaller clinics in Canada and Australia were very limited in their clinical capacity, with one diagnostic slot per month provided as one full day was required in these settings for diagnosis, equating to space for the diagnosis of 12 individuals per year (McFarlane, 2011; Panton et al., 2022; Temple et al., 2015).

4.3.2.4 Cost estimations of diagnosis (*n*=2 studies)

Cost estimations were provided in two studies, both conducted in Canada (Clarren & Lutke, 2008; Popova et al., 2013). Clarren et al. (2008) reported the costs of public programs ranged between C$2,500 and C$3,000 per individual, while not-for-profit private program costs ranged between C$2,000 and C$5,000 per individual. Popova et al. (2013) used a non-experimental cost estimation model to determine the cost of FASD diagnoses in Canada, based on the estimates of clinical capacity and professional time provided by Clarren et al. (2011). According to their model, the total cost per person ranged between C$3,110 to C$4,570, including screening and referral, clinic, diagnosis, and general support. Although the other study found that the overall cost of FASD diagnostic services per year was between C$3.6 million and C$7.0 million [37]. It is important to acknowledge that both studies only examined the cost of billable clinical hours and did not account for other service costs. For example, the estimations did not include facility costs (office space, medical records, or other personal costs to run a facility), the cost of instruments (e.g., psychological instruments), which could add an additional cost of C$330 to C$500 per person, consultation with additional professionals, and, as stated previously, did not include the cost of professionals' time in the preparation of assessments and writing of final reports (Popova et al., 2013).

4.3.2.5 Funding for FASD diagnostic clinics (*n*=9 studies)

It was evident from available studies that the professionals required to form the MDT, the time required and access to required resources were key considerations in the diagnosis of FASD. However, access to consistent funding for FASD clinics was reported to be difficult. Seven studies provided specific information on funding for FASD clinics in Canada (Clarren & Lutke, 2008; Clarren et al., 2011; Dugas et al., 2022; McFarlane, 2011; McFarlane & Rajani, 2007; Peadon et al., 2008; Temple et al., 2015), one study reported challenges with funding in Australian clinics (Panton et al., 2022) and another study discussed the importance of funding, political support and recognition of FASD internationally as a potential facilitator for creation and implementation of unified diagnostic guidelines (Reid et al., 2022a). In Canada, the clinics surveyed received provincial and federal funding, funding from non-government organisations, (NGOs) not-for-profit organisations, in-kind donations, research grants or private funding sources, depending on geographic location (Clarren & Lutke, 2008; Dugas et al., 2022; McFarlane, 2011; McFarlane & Rajani, 2007; Peadon et al., 2008; Temple et al., 2015). Patient fees or insurance were stated as the primary source of funding for clinics that did not receive other types of contributions (Peadon et al., 2008). In Australia, the demand for FASD diagnostic clinics was reported to far outweigh the service capacity, with Panton et al. (2022) highlighting the need for funding for efficient clinical models. The authors

described how their funding model incorporated fee-for-service partnerships from private organisations, NGOs, and government funding, and required financial and logistical support to sustainably meet the diagnostic demands. Furthermore, maintaining links to partner agencies including disability services, justice and child protection services was noted to be critical to ensure individuals were able to access well-established government agencies after assessment and that funding was not tied to a single source (Panton et al., 2022). Cost and/or lack of funding was cited as a significant barrier to implementation of a unified diagnostic approach, which may help to improve clinical capacity (Reid et al., 2022a).

4.3.3 Models of Care (*n*=4)

Four studies detailed specific models of care within different FASD diagnostic clinics (McFarlane, 2011; McFarlane & Rajani, 2007; Patel et al., 2019; Temple et al., 2015) and one study evaluated implementation of different models of care across six FASD diagnostic clinic sites in Australia (Panton et al., 2022). To meet the specific needs of rural communities, the efficacy of the same mobile diagnostic and assessment team was assessed in two studies conducted five years apart (McFarlane, 2011; McFarlane & Rajani, 2007). The diagnostic team was comprised of agency service providers from throughout the service area who would come together on pre-determined clinic days once a month to the community in the closest proximity to the client's home. The advantages of a mobile diagnostic team included that: 1) the model required minimal sustainable funds for team coordination, infrastructure, or clinic cost, 2) there were only short wait times for patients (3-4 months), and 3) the mobility of the team reduced perceived barriers to service for patients. In contrast, the challenges associated with the sustainability of a mobile diagnostic assessment team included: 1) limited availability of rural professionals and 2) difficulties accessing sustainable funds.

An embedded model of care was explored in one study conducted in Canada (Temple et al., 2015). The diagnostic team was comprised of professionals who were already employed by the agency with a particular focus on the diagnostic assessment of adults. Some reported strengths of this model of care were that it enabled provision of a more seamless service to patients and costs were reduced as the service was capitalising on pre-existing staff and links to external supports. However, given that the patient demographic were adults, patient cancellations and missed appointments were reported to be common, which was a major challenge for the clinic.

A collaborative care model was explored in a study from Canada, in which the clinic of interest focussed specifically on the diagnosis of youth in care through the Children's Aid Society of Toronto (CAST) services (Patel et al., 2019). This model involved a three-step approach which included: 1) completion of a neurobehavioral screen by child protection workers; 2) paediatric assessment of individuals who screened positive in the neurobehavioral screen; and 3) integration of findings through a full psychiatric assessment. Provision of diagnostic services in an out-of-home care setting supported access to a high-risk population and aimed to reduce costs by utilising pre-existing services offered by CAST. However, youth with more subtle FASD presentations were not identified by the initial screening tool, which may lead to increased costs later in their life as they did not receive appropriate interventions.

Panton et al. (2022) explored the relative challenges and successes of new FASD specialist MDT clinics and their respective models of care as they were being rolled out across six sites in Australia. Some challenges identified across the six sites included high staff turnover, particularly in the roles of co-ordinator, paediatrician and psychologist, lack of appropriate planning for local areas meaning the needs of the region were not met, challenges with maintaining community engagement and differences in the interpretation of the Australian FASD Guide to Diagnosis. Factors noted for sustainable success included strong executive support, co-localisation of members of the MDT (e.g., all based in a single hospital), and

steps taken to facilitate FASD awareness within local networks, such as community reference groups, community engagement sessions and local clinical training. This study highlighted that a nationally consistent diagnostic approach, data collection and referral processes, may not be appropriate for all areas and that it is important to acknowledge the specific needs of the region.

5. Discussion

5.1 Summary of main findings and comparison with previous studies

The current scoping review aimed to explore the available evidence regarding resource considerations and models of care involved in the assessment and FASD diagnostic process. The available literature explored service costs more often than patient costs. The included studies highlighted that generally, service costs were attributed to professional time and staffing of MDTs. Available studies reported that large amounts of professional time were required for diagnosis, including direct and indirect care. However, there were only a small number of studies available that provided specific cost estimations of the services provided. Multiple models care were identified in the literature that primarily aimed to capitalise on the services that were available, which was intended to improve patient care and reduce service costs.

Staffing of MDTs represented one of the primary service costs (Clarren and Lutke, 2008, Clarren et al., 2011, McFarlane, 2011, Popova et al., 2013). An MDT approach has been recommended to most accurately document the full spectrum of outcomes seen in individuals with PAE and was embraced in some form by all models of care in the studies included within this review. However, such approaches were noted to be costly, and MDTs were not always available. There was also great variability in the professional time required for assessment, likely due to differences in the mix of professionals within diagnostic teams, and the community to which the service was being provided. Notably, in some remote communities, a skilled MDT may only be available for the duration of the research funding. Estimates of time required for diagnosis were likely inaccurate as consultation with additional professionals and report preparation time were not included in costs estimates. There was likely significant underestimation in the costs of resources required for diagnoses as facility costs and assessment tool costs were not always included in estimates.

Notably, there was limited number of studies available to review, the majority from one country (i.e., Canada). Although estimates of clinical capacity in Australia were not specifically captured in available research, Panton et al. (2022) stated that the demand for diagnosis outweighed the Australian diagnostic service availability, although demonstrated that with their government funded models of care project, FASD diagnostic activity increased significantly over a two-year period, including an uptake of referrals to the National Disability Insurance Scheme (Panton et al., 2023). Estimates of clinical capacity from Canadian studies similarly indicated that diagnostic demands were not being met and outlined the need for increased clinical capacity or more efficient models clinical care (Clarren and Lutke, 2008, Clarren et al., 2011, Popova et al., 2013). Initial acquisition of funding and availability of professionals were reported to be significant barriers to clinical capacity in an international survey of 55 clinics (Reid et al., 2022a). Given the current state of publicly funded healthcare, there is a risk that the currently recommended comprehensive assessment process in FASD diagnostic guidelines (e.g., Cook et al., 2016; Bower et al., 2017) is too costly for service providers to offer at a capacity that meets the current demand. However, the level of risk is difficult to quantify given the limited research available regarding the economic feasibility of delivering a comprehensive multidisciplinary FASD assessment.

Results from the current review demonstrate the diversity of possible models of care, some of which capitalise on pre-existing systems and professionals. An embedded model of service delivery for FASD that

capitalizes on established services (e.g., general child development services), may reduce costs through use of pre-existing skilled staff and allow patients access to local external supports (e.g., financial/job support, psychosocial support, vocational supports, housing support etc; Temple et al., 2015). This approach is highly relevant to rural communities where implementation of diagnostic assessments within existing clinics and support from community agencies would help to increase clinical capacity within those regions (McFarlane, 2011, McFarlane and Rajani, 2007). For example, a remote region in Queensland, Australia has been trialling the integration of assessment and diagnosis of FASD within existing multi-disciplinary primary care teams (Shanley et al., 2019, Reid et al., 2021). Similar methods have been implemented in other Australian regions where the Victoria Fetal Alcohol Service (VicFAS), a state-wide team that uses a flexible model of care, including telehealth, outreach clinics, school visits and collaboration with community clinicians, have assisted local health services to provide supports to children with FASD and their families (Monash Health, 2022). These types of flexible service delivery approaches may help to better support individuals who live in rural and remote areas as more continuity of care can be provided by health professionals known to the individuals attending for assessment and their families (Reid et al., 2023, Reid et al., 2022b).

Prioritising a more widespread and nuanced understanding of the cohorts of children who may be at increased risk, such as those in out-of-home care or in contact with the justice system and providing additional diagnostic services within these systems of care may support increased clinical capacity. This would reduce costs by training and using available staff to support the diagnostic process (e.g., case managers, clinicians already working in these settings; Department of Health, 2018). For example, Patel et al. (2019) demonstrated the effectiveness of using a collaborative care model that integrated community assessment by a child protection worker who screened youth coming into care using a neurobehavioral screening tool before referring children who screened positive to a paediatrician. Notably, expanding the capacity of child and family health services, education system (e.g., learningwithfasd.org.au), and justice workforces (Heanue et al., 2022; Passmore et al., 2018) to recognise FASD is one of the potential enablers of change identified in the National FASD Strategic Action Plan 2018-2028 (Department of Health, 2018).

As there was only limited evidence available regarding patient costs, with the included studies providing very broad estimates of time the required for diagnostic assessment, it is thus challenging to draw any meaningful conclusions regarding patient costs from the available literature. Investigating ways to streamline the diagnostic process; upskilling available professionals across a wide range of service settings and locations; and supporting collaborative care across settings may reduce wait-times and patient travel, thereby reducing patient costs. Ensuring that highly skilled members of the MDT can spend most of their time on discipline specific clinical tasks while administrative staff prepare the required documents and support report writing is another potential cost-effective measure.

From the available studies it was evident that there were very few services available for adults with FASD, which increases the demand on already limited resources. Of the included studies, only one study was solely focused on adults (Temple et al., 2015), with others highlighting the low clinical capacity for adult diagnosis (Clarren et al., 2011; McFarlane, 2011, McFarlane and Rajani, 2007). Low clinical capacity for providing services to adults may be contributing to the health care costs for adults, which were estimated in a systematic review to be 40% higher than those for children, while loss of productivity associated with FASD was estimated to be 6.3-fold greater in adults compared to children (Greenmyer et al., 2018). Methods to lessen the financial burden on adults undergoing diagnosis could include the provision of childcare services at diagnostic clinics or, if patients struggle to take time off work, using telehealth to reduce travel time and income lost due to time off work. However, little evidence exists on adult patient

costs; therefore, future research should focus specifically on increasing accessibility of adult services and understanding the costs associated with the diagnosis of FASD in adults.

5.2 Limitations and Future Directions

A key limitation of the scoping review was the small number and predominately descriptive nature of the studies that were identified. This made it challenging to group studies based on their content and draw meaningful conclusions from the available literature. Some critical studies may have been missed in the literature searches if cost was a minor feature of the study and thus not mentioned in the title or abstract. Further, the search focused on peer-reviewed journal articles and did not include grey literature, which may have excluded relevant clinical information regarding costs and current of models of care. Additionally, most included studies were conducted in Canada, with only one study identified from Australia. Therefore, the generalisability of the findings to the Australian context is limited and there was no opportunity to be able to compare the potential differential impacts of diagnostic criteria on the specific models of care developed and delivered and the associated service and patient costs. There was also very limited information regarding the specific dollar values of the costs of service delivery (e.g., costs of different practitioners and their time spent per assessment) and no ability to compare different costs across different models of care. Future research is required globally to understand in more detail the specific costs at both the service and patient level, other resource requirements and models of care that can support increased access to assessment and diagnosis of FASD.

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7. Appendices

Appendix A: Search strategy

PubMed	(prenatal alcohol[Title/Abstract] OR prenatal ethanol[Title/Abstract] OR fetal alcohol[Title/Abstract] OR foetal alcohol[Title/Abstract] OR fetal alcohol spectrum disorder[Title/Abstract] OR foetal alcohol spectrum disorder[Title/Abstract] OR fetal alcohol syndrome[Title/Abstract] OR foetal alcohol syndrome[Title/Abstract] OR partial fetal alcohol syndrome[Title/Abstract] OR partial foetal alcohol syndrome[Title/Abstract] OR alcohol-related neurodevelopmental disorder[Title/Abstract] OR alcohol related neurodevelopmental disorder[Title/Abstract] OR alcohol-related birth defects[Title/Abstract] OR alcohol related birth defects[Title/Abstract] OR static encephalopathy[Title/Abstract] OR neurobehavioral disorder alcohol exposed[Title/Abstract] OR neurobehavioral disorder associated with prenatal alcohol exposure[Title/Abstract]) AND (Cost-effectiveness analysis[Title/Abstract] OR cost-benefit[Title/Abstract] OR equity[Title/Abstract] OR utility[Title/Abstract] OR economic impact[Title/Abstract] OR cost of diagnosis[Title/Abstract] OR cost estimate[Title/Abstract] OR clinical capacity[Title/Abstract] OR service delivery implications[Title/Abstract] OR resource implications[Title/Abstract] OR risks[Title/Abstract] OR benefits[Title/Abstract] OR model of care[Title/Abstract] OR feasibility[Title/Abstract])
EMBASE	((((((((((((((('prenatal'/exp OR prenatal) AND ('alcohol'/exp OR alcohol) OR 'prenatal'/exp OR prenatal) AND ('ethanol'/exp OR ethanol) OR fetal) AND ('alcohol'/exp OR alcohol) OR foetal) AND ('alcohol'/exp OR alcohol) OR fetal) AND ('alcohol'/exp OR alcohol) AND ('spectrum'/exp OR spectrum) AND ('disorder'/exp OR disorder) OR foetal) AND ('alcohol'/exp OR alcohol) AND ('spectrum'/exp OR spectrum) AND ('disorder'/exp OR disorder) OR fetal) AND ('alcohol'/exp OR alcohol) AND ('syndrome'/exp OR syndrome) OR foetal) AND ('alcohol'/exp OR alcohol) AND ('syndrome'/exp OR syndrome) OR partial) AND fetal AND ('alcohol'/exp OR alcohol) AND ('syndrome'/exp OR syndrome) OR partial) AND foetal AND ('alcohol'/exp OR alcohol) AND ('syndrome'/exp OR syndrome) OR 'alcohol related') AND neurodevelopmental AND ('disorder'/exp OR disorder) OR 'alcohol'/exp OR alcohol) AND related AND neurodevelopmental AND ('disorder'/exp OR disorder) OR 'alcohol related') AND ('birth'/exp OR birth) AND defects OR 'alcohol'/exp OR alcohol) AND related AND ('birth'/exp OR birth) AND defects OR static) AND ('encephalopathy'/exp OR encephalopathy) OR neurobehavioral) AND ('disorder'/exp OR disorder) AND ('alcohol'/exp OR alcohol) AND exposed OR neurobehavioral) AND ('disorder'/exp OR disorder) AND associated AND with AND ('prenatal'/exp OR prenatal) AND ('alcohol'/exp OR alcohol) AND ('exposure'/exp OR exposure) AND (((((((('cost effectiveness':ab,ti AND analysis:ab,ti OR 'cost benefit':ab,ti OR equity:ab,ti OR utility:ab,ti OR economic:ab,ti) AND impact:ab,ti OR cost:ab,ti) AND of:ab,ti AND diagnosis:ab,ti OR cost:ab,ti) AND estimate:ab,ti OR clinical:ab,ti) AND capacity:ab,ti OR service:ab,ti) AND delivery:ab,ti AND implications:ab,ti

	OR resource:ab,ti) AND implications:ab,ti OR risks:ab,ti OR benefits:ab,ti OR model:ab,ti) AND of:ab,ti AND care:ab,ti OR feasibility:ab,ti)
Web of Science	prenatal alcohol OR prenatal ethanol OR fetal alcohol OR foetal alcohol OR fetal alcohol spectrum disorder OR foetal alcohol spectrum disorder OR fetal alcohol syndrome OR foetal alcohol syndrome OR partial fetal alcohol syndrome OR partial foetal alcohol syndrome OR alcohol-related neurodevelopmental disorder OR alcohol related neurodevelopmental disorder OR alcohol-related birth defects OR alcohol related birth defects OR static encephalopathy OR neurobehavioral disorder alcohol exposed OR neurobehavioral disorder associated with prenatal alcohol exposure (Topic) AND Cost-effectiveness analysis OR cost-benefit OR equity OR utility OR economic impact OR cost of diagnosis OR cost estimate OR clinical capacity OR service delivery implications Or resource implications OR risks OR benefits OR model of care OR feasibility (Topic)
PsycInfo	Abstract: prenatal alcohol *OR* Abstract: prenatal ethanol *OR* Abstract: fetal alcohol *OR* Abstract: foetal alcohol *OR* Abstract: fetal alcohol spectrum disorder *OR* Abstract: foetal alcohol spectrum disorder *OR* Abstract: fetal alcohol syndrome *OR* Abstract: foetal alcohol syndrome *OR* Abstract: partial fetal alcohol syndrome *OR* Abstract: partial foetal alcohol syndrome *OR* Abstract: alcohol-related neurodevelopmental disorder *OR* Abstract: alcohol related neurodevelopmental disorder *OR* Abstract: alcohol-related birth defects *OR* Abstract: alcohol related birth defects *OR* Abstract: static encephalopathy *OR* Abstract: neurobehavioral disorder alcohol exposed *OR* Abstract: neurobehavioral disorder associated with prenatal alcohol exposure *AND* Abstract: Cost-effectiveness analysis *OR* Abstract: cost-benefit *OR* Abstract: equity *OR* Abstract: utility *OR* Abstract: economic impact *OR* Abstract: cost of diagnosis *OR* Abstract: cost estimate *OR* Abstract: clinical capacity *OR* Abstract: service delivery implications *OR* Abstract: resource implications *OR* Abstract: risks *OR* Abstract: benefits *OR* Abstract: model of care *OR* Abstract: feasibility
Cochrane Library	(Abstract: prenatal alcohol OR Abstract: prenatal ethanol OR Abstract: fetal alcohol OR Abstract: foetal alcohol OR Abstract: fetal alcohol spectrum disorder OR Abstract: foetal alcohol spectrum disorder OR Abstract: fetal alcohol syndrome OR Abstract: foetal alcohol syndrome OR Abstract: partial fetal alcohol syndrome OR Abstract: partial foetal alcohol syndrome OR Abstract: alcohol-related neurodevelopmental disorder OR Abstract: alcohol related neurodevelopmental disorder OR Abstract: alcohol-related birth defects OR Abstract: alcohol related birth defects OR Abstract: static encephalopathy OR Abstract: neurobehavioral disorder alcohol exposed OR Abstract: neurobehavioral disorder associated with prenatal alcohol exposure AND Abstract: Cost-effectiveness analysis OR Abstract: cost-benefit OR Abstract: equity OR Abstract: utility OR Abstract: economic impact OR Abstract: cost of diagnosis OR Abstract: cost estimate OR Abstract: clinical capacity OR Abstract: service delivery implications OR Abstract: resource implications OR Abstract: risks

	OR Abstract: benefits OR Abstract: model of care OR Abstract: feasibility):ti,ab,kw AND (Cost-effectiveness analysis OR cost-benefit OR equity OR utility OR economic impact OR cost of diagnosis OR cost estimate OR clinical capacity OR service delivery implications Or resource implications OR risks OR benefits OR model of care OR feasibility):ti,ab,kw (Word variations have been searched) #2 (prenatal alcohol OR prenatal ethanol OR fetal alcohol OR foetal alcohol OR fetal alcohol spectrum disorder OR foetal alcohol spectrum disorder OR fetal alcohol syndrome OR foetal alcohol syndrome OR partial fetal alcohol syndrome OR partial foetal alcohol syndrome OR alcohol-related neurodevelopmental disorder OR alcohol related neurodevelopmental disorder OR alcohol-related birth defects OR alcohol related birth defects OR static encephalopathy OR neurobehavioral disorder alcohol exposed OR neurobehavioral disorder associated with prenatal alcohol exposure):ti,ab,kw AND (Cost-effectiveness analysis OR cost-benefit OR equity OR utility OR economic impact OR cost of diagnosis OR cost estimate OR clinical capacity OR service delivery implications Or resource implications OR risks OR benefits OR model of care OR feasibility):ti,ab,kw (Word variations have been searched)
CINAHL	AB (prenatal alcohol OR prenatal ethanol OR fetal alcohol OR foetal alcohol OR fetal alcohol spectrum disorder OR foetal alcohol spectrum disorder OR fetal alcohol syndrome OR foetal alcohol syndrome OR partial fetal alcohol syndrome OR partial foetal alcohol syndrome OR alcohol-related neurodevelopmental disorder OR alcohol related neurodevelopmental disorder OR alcohol-related birth defects OR alcohol related birth defects OR static encephalopathy OR neurobehavioral disorder alcohol exposed OR neurobehavioral disorder associated with prenatal alcohol exposure) AND AB (Cost-effectiveness analysis OR cost-benefit OR equity OR utility OR economic impact OR cost of diagnosis OR cost estimate OR clinical capacity OR service delivery implications Or resource implications OR risks OR benefits OR model of care OR feasibility)

Appendix B: Summary of data charting

Appendix B Table 1. Summary of studies included in the scoping review

Study	Country	Study Aims	Study Design	Number of clinics	Key Data Collected/Measures
Clarren et al. 2008	Canada	1) Determine FASD clinical activity in Northwest Canada	Cross-sectional survey of clinical programs	15 FASD clinics	Clinical capacity, aggregate diagnostic results, team composition, time of clinical assessment, cost of assessment.
Clarren, Lutke and Sherbuck, 2011	Canada	1) Identify the availability and impact of the Canadian Guidelines in clinics that purport to routinely do FASD diagnosis. 2) Determine the capacity of Canada to perform these necessary medical evaluations.	Cross-sectional survey of clinical programs	39 FASD clinics	Clinical capacity, staffing level, diagnostic processes used, knowledge and implementation of guidelines.
Dugas et al. 2022	Canada	1) Describe diagnostic interdisciplinary team composition for different age groups. 2) Determine the number of clinics that follow the Canadian interdisciplinary diagnostic team guidelines by age group.	Cross-sectional investigation of Canadian FASD clinical capacity	41 FASD clinics	General clinic information (location, sources of funding and services offered), number of referrals, assessments done/year, interdisciplinary team composition by age group, current diagnosis reporting practices, explanatory tools and immediate and post-diagnosis support and counselling.
McFarlane et al. 2007	Canada	1) Describe the history of the Lakeland Centre for FASD relative to development of the model. 2) Describe the diagnostic process used to diagnose children and adults, as well as rural-specific adaptations and challenges.	Summary of clinic history and critical review	1 FASD clinic servicing rural populations	Diagnostic service delivery model, clinic process, model adaptations for rural diagnosis, factors critical to model success, and challenges.
McFarlane et al. 2011	Canada	1) Describe the community-based model developed by the rural Lakeland Centre for FASD 2) Summarise clinical findings, and successes and challenges to date	Summary of clinic history and critical review	1 FASD clinic servicing rural populations	Outcomes not clearly listed but separated into phases of diagnosis (preclinic, clinic days, diagnosis and recommendations, case conference, emotional support, team debriefing,

					outreach support) and clinical observations.
Panton et al. 2022	Australia	1) Develop a nationally consistent diagnostic approach, data collection and referral process to: a. Improve FASD diagnostic capacity. b. Increase FASD awareness, knowledge, and advocacy.	Cross-sectional survey of clinical programs	6 FASD clinics	Demographic information, setting up the project and FASD clinic, community reference groups, models of care, community engagement, sustainability, future goals, and overall successes.
Patel et al. 2019	Canada	1) Determine efficacy of three phase multidisciplinary approach (screening tool by child protection worker, paediatric assessment, psychiatric assessment integration) for diagnosis of FASD for youth in care through Children's Aid services.	Retrospective chart review of clinic data	1 medical clinic	Chart review of participant's clinic files including demographic, comorbidities and treatment recommendation.
Peadon et al. 2008	International	Conduct an international survey to: 1) Describe specialist dedicated clinical service provision for diagnosis and assessment of children with FASD. 2) Establish which countries have specialised services. 3) Describe the models of service used. 4) Compare clinical practice in the services with the published recommendations for assessment of children exposed to alcohol in utero.	Cross-sectional survey of clinical programs	34 FASD clinics	Information relating to clinic population, clinic staff, assessment process, other clinic activity/services provided.
Popova et al. 2013	Canada	1) Estimate the per person cost of FASD diagnosis as well as the annual cost of FASD diagnostic services in Canada.	Non-experimental cost estimation model reliant on reports from	Not applicable	Cost of FASD diagnosis per person (hours required by each specialist multiplied by respective hourly rate),

			respected authorities and survey results of FASD Research Network.		and cost of all cases per year (based on estimates from previous studies).
Reid et al. 2022a	International	1) To collect up to date information regarding guidelines utilised at specialist FASD diagnostic clinics. 2) Collect information from clinicians regarding opinion on feasibility of unified diagnostic approach. 3) Collect information from clinicians as to what they believe to be barriers/facilitators to developing a unified diagnostic approach.	Cross-sectional survey of clinical programs	147 FASD clinics	Clinic's current diagnostic approach, whether they would support a unified method and barriers and facilitators to consistent international FASD diagnostic approach.
Temple et al. 2015	Canada	1) Describe the development and operation of an interdisciplinary FASD diagnostic clinic focussing on adults.	Summary of clinic processes and critical review.	1 FASD clinic	Description of clinic and process, diagnostic outcomes, challenges, advantages, and disadvantages of chosen model.

Appendix B Table 2. Summary of studies that included diagnostic teams, clinical capacity and funding information

Study	Target Population (if stated)	Time for Diagnosis	Diagnostic Team	Cost of Diagnosis	Clinical Capacity	Funding information	Key Considerations
Clarren & Lutke 2008	Children	• Mean time for direct patient care=12.5hrs ○ Paediatrician=2.5hrs ○ Speech and language pathologist=2.5hrs ○ Occupational therapist=1.5hrs ○ Clinical psychologist=6hrs • Mean time for indirect patient care=13.5hrs ○ Paediatrician=3hrs ○ Speech and language pathologists=3hrs ○ Occupational therapist=3hrs ○ Clinical psychologists=4.5hrs	• Average MDT had 4 members (ranged from 2 to 7), including: ○ Physicians ○ Psychologists ○ Speech pathologists ○ Occupational therapists ○ Social workers ○ Patient co-ordinator (likely) ○ Psychiatrists (sometimes) ○ Geneticists (sometimes) ○ Nurses (sometimes) ○ Education specialists (sometimes) ○ Family advocates (sometimes)	• 3 public programs estimated cost of an evaluation at $2500-$3500 (mean and median=$3000) • All 6 NFP private programs had estimated cost of $2000-$5500 (mean and median of $3500)	• 15 programs had total capacity for FASD diagnosis of 1140 patient assessments • Western provinces - total capacity was 816 in 2005, expected to climb to 975 in 2006 • Ratio of FASD diagnoses/full clinic capacity ranging from 48% to 79% • Only 3 programs routinely saw adults • Most patients diagnosed after age of school entry	• 11 programs administered directly through provincial or territorial health systems including university facilities • 4 programs administered through not-for-profit corporations	• Diagnoses are complex and require lengthy assessment in comparison to other developmental concerns. • Need for multidisciplinary team which increases cost. • Further clinical capacity needed, particularly for adults. • Different diagnostic processes may result in different costs; this needs to be explored in future studies.
Clarren, Lutke & Sherbuck 2011	Children and youth (<10 programs specifically for adults)		• Multidisciplinary team • 18 clinics (46%) have full complement of staff professionals (recommended by the		• Of the 39 clinics and estimates from the other five clinics, the total capacity	• Funding restrictions often meant that less than full on-site team	• Guidelines helped clinics acquire funding for staff. • Guidelines improved

			guidelines) to evaluate brain dysfunction ○ 34 had psychologists ○ 25 had occupational therapists ○ 28 had speech pathologists • 90% reported a multidisciplinary approach • 79% reported team development of treatment plan		would be 2,392 in 2010 and 2,288 in 2011 • Estimates suggest that 37,000 diagnostic slots needed/year • Western provinces – increase in capacity to 1,773 in 2010 • <10 programs prepared to see adults		consistency of diagnostic outcomes and increased structured procedures, potentially reducing cost through efficiencies. • Full complement of recommended interdisciplinary professional staff was lacking in majority of clinics: too difficult to source and fund. • Despite cost, need to substantially increase diagnostic capacity, if the goal is to see all those with need.
Dugas et al. 2022	Children and adolescents (<18 months-18 years)		• 7/41 clinics diagnosed in <18months (4 clinics used MDT) • 25/41 clinics diagnosed in 18months-5 years (15 used MDT) • 40/41 clinics diagnosed in 6-18years (32 used MDT)		• 2537 referrals received every year and only 1797 assessments are completed (diagnostic	• 85-95% of regions received provincial funding • In-kind donations and private funding	• Need for National FASD strategy to ensure individuals with FASD and their families have access to

					capacity of 71%) • 17% (7/41) diagnosed infants (<18months) • 60% (25/41) clinics diagnosed preschool children • 98% (40/41) diagnosed in school-aged children	were highest in Central (46%) and Atlantic (57%) Canada • Federal funding highest in Atlantic clinics (43%) • NGO funding highest in Central (15%) and Western/Northern (33%) clinics	services they need • Diagnostic capacity remains important public health issue (similar to findings by Clarren et al, 2011) • Limited diagnostic capacity and lack of resources across Canada highlights critical need for continued FASD support
McFarlane & Rajani 2007	Children and adults	• Child diagnosis ~4 hours + indirect documentation time • Adult diagnosis completed over ~1 day + indirect documentation time	• Children's diagnostic team: ○ Paediatrician ○ Neuropsychologist ○ Speech-language pathologist ○ Public health nurse ○ Aboriginal liaison ○ Mental health therapist ○ Social worker ○ Addictions counsellor ○ Team coordinator ○ Secondary members can be added if needed • Adult diagnostic team: ○ Physician			• Infrastructure funding co-provided by Alberta Ministry of Children Services (demonstration project) and fundraising efforts of the Board of Directors of the Society • For rural delivery: in-kind	• Training was costly, but provision of appropriate training was critical to team functioning, particularly in rural environment. • Training could be administered by established clinics, to assist new clinics developing FASD capacity

			○ Psychologist ○ Mental health therapist ○ Career counsellor ○ Addictions counsellor ○ Aboriginal liaison ○ Disability services coordinator ○ Team coordinator			agreement in place for all team members (except physicians and psychologists, who are paid on fee-for service basis) • Primary funding agencies include: Alberta Children's Services, local school divisions, First Nations communities, Alberta Human Resources and Employment, or Persons with Developmental Disabilities	(e.g., Lakeland Centre provided annual training for practitioners around Canada) • A mobile team model of care was used effectively to reach rural communities.
McFarlane et al. 2011	Adults	• ~1 day for diagnosis	• Diagnostic team comprised of: ○ Team coordinator ○ Physician ○ Neuropsychologist ○ Mental health therapist ○ Psychiatrist ○ Career counsellor ○ Addictions counsellor ○ Cultural liaison		• 1 adult/month (12 adults/year)	• Contributing agencies of local professionals provide them with salary to participate in the team • Fee-for service arrangement for neuropsycholo	• Acquiring interested relevant professionals was difficult due to limited funds available. • In-kind staff support from community agencies was cost-effective

			○ Legal representative ○ Disability services provider ○ Post diagnostic outreach worker			gist and psychiatrist • Honorarium provided to physician • Team coordinator and post diagnostic outreach workers are employed by the centre • In-kind donations from community agencies • Partnerships with all levels of government are cultivated and critical for funding and support • Facility organises fundraisers and attends conferences, liaises with federal, provincial and local gov reps	with positive practical implications for patients. • Community-based clinics using multi-disciplinary approach deemed effective. • Mobile multi-disciplinary teams deemed effective. • Cultural liaison facilitates positive relationship with communities and increases sustainability.
Panton et al. 2022	Children and adolescents		• Multidisciplinary team ○ Paediatrician/physician ○ Psychologist ○ Speech pathologist		• Goulburn Valley Health Service –	• All sites operated on a fee-for-service diagnostic	• Inadequate funding affects diagnostic capacity,

			○ Occupational therapist (where clinically appropriate) ○ Site coordinator (varied clinical background) • PATCHES (Paediatric Child Health & Education Services, Wester Australia) currently use multidisciplinary approach, all new sites were trained on the multi-disciplinary assessment model and		limited to 12 children/year • Demand for diagnosis outweighs the diagnostic service availability in Australia	clinic approach, adapted to fit local needs • Maintaining links with partner agencies (disability services, justice and child protection) helped individuals continue to access support of well-established government agencies post-diagnosis, and also helped ensure that funding was not tied to single source • Combined fee-for service (private or NGO) and government funded (e.g., state child development services) models require financial and	funding needs to be afforded to efficient clinical models (private or blended public-private models) • Co-located multi-disciplinary team is important for success • Should ensure FASD training and education includes ethical, social and political issues that seem to be a barrier toward FASD screening, diagnosis and management • Building local awareness of FASD is important to build local capacity and FASD advocacy • National approach is not appropriate for all areas

						logistical support to sustainably meet the diagnostic demand	
Patel et al. 2020	Children and adolescents in child welfare services		Three step diagnostic process involves: • Neurobehavioral Screening Tool (NST) utilized by child protective workers to screen youth coming into care at CAST • Youth screening positive would then be assessed by paediatrician at CAST to determine if FASD diagnosis possible • Referral for psychiatric assessment by psychiatrist				• Stressed importance of collaborative care model and an interdisciplinary approach to diagnosis. • Integrated community assessment (facilitated by screening from child protection worker) enables more timely and efficient screening process.
Peadon et al. 2008	Children	• Number of visits required for diagnosis ranged from 1-3 (median=1 visit) • 2 clinics reported that time required varied from child to child • Duration of visits was 0.5-6 hours (median=3.25 hours)	• 33/34 clinics staffed by multi-disciplinary team • 33/34 clinics had at least one medical and psychology professional • Clinic that was not run by a multidisciplinary team was staffed by dysmorphologist • 32/33 clinics had at least one member of staff who had undergone speciality training			• Funding from many sources: ○ Charitable and community sources (4 cases) ○ Research grants (7 clinics) • Patient fees – self-pay or insurance and	• Health professionals seek information on FAS, referral services, and a register of health professionals with diagnostic expertise. • Multiple models of care are

			• 22/33 clinics all staff had undergone specialty training			did not receive state, federal, research or charitable contributions (2 clinics)	possible including community-based, as well as urban-specialist with outreach; models need to suit the location and context. • To establish FASD diagnostic services, strategies are needed to ensure funding, and suitably trained professionals.
Popova et al. 2013	All clinics in Canada (model used to estimate costs), age of population not specified	• Total Time=32-47 hours/person o Screening and referral=1-2 hours/person o Intake into diagnostic clinic=2-4 hours/person o Diagnosis=23-33hours/person o General support=6-8 hours/person	• Specialists of the multidisciplinary team involved in each step of the process: o Screening and referral=physician/ paediatrician/family doctor/social worker/probation officer (not limited to these individuals) o Intake=coordinator/social worker o Diagnosis= ▪ Physical/developmental /medical assessment/ examination =physician/paediatrici	• Total cost=$3,110-$4,570/person o Screening and referral=$150-300/person o Intake into diagnostic clinic=$160-$320/person o Diagnosis estimated to cost $2,650-$3,750/person o General support=$150-200/person			• Majority of cost represents need for multidisciplinary team. • Current clinic capacity low and clinics struggle with limited access to needed expertise. • Many costs not accounted for in this study, including indirect practitioner time, cost of testing materials, cost of office space,

			an/ developmental paediatrician/family doctor (specially trained in FASD diagnosis) ▪ Dysmorphology assessment= dysmorphologist/ geneticist ▪ Neurobehavioral assessment – developmental paediatrician, psychologist, speech and language pathologist, occupational therapist and coordinator for case management ▪ General support – secretary/clerical worker	• Total cost of diagnosing FASD in Canada ranges from $3.6-5.2 million up to $5.0-7.3 million/year			study, or for follow-up care. • Although large initial outlay, return on investment and long-term benefit likely high.
Reid et al. 2022a	Not specified					• Funding, political support, recognition of the importance of FASD internationally recognised as potential facilitators for creation and implementation of unified diagnostic guidelines	• Unified FASD diagnostic criteria necessary to standardise global disease management, clinical care and research outcomes • Implementing consistent diagnostic criteria will enable improved

						• Cost/lack of funding was barrier to implementation of a unified diagnostic approach • Some settings do not have resources and specialist access which influences capacity for implementation of universal guidelines	patient outcomes and better collaborative research efforts • Several barriers exist to this process, some of which include cost/lack of funding and lack of ability to standardise due to differences in resources, training and specialist access
Temple, Ives and Lindsay, 2015	Adults	• Intake/assessment day=4 hours (if the individual is able to tolerate it)	• Diagnostic team composed of: o Clinic coordinator o Nurse practitioner o Consulting physician o Clinical psychologist o Speech-language pathologist o Behaviour therapist • Additional supports provided: o Primary medical/nursing care o Psychiatry o Psychology o Speech-language pathology o Social work/counselling o Service coordination o Occupational therapy o Behaviour therapy		• 1 adult/month (12 adults/year)	• Provincially-funded community health agency for individuals with intellectual and developmental disorders of all ages	• Very few evidence-based services available for adults with FASD, increasing demand on limited resources. • Embedded model of service delivery, whereby service provided within developmental/ disabilities sector, provided more seamless service and reduced costs due to

			○ Audiology • Increased need for specialised staff due to absence of carer for adult patients				capitalising on pre-existing staff and links to external supports.

Appendix B Table 3. Summary of studies that evaluated different models of care

Study	Model of Care	Successes/Advantages	Losses/Challenges
McFarlane et al. 2007	Mobile diagnostic team and follow-up support personnel	• Model requires minimal sustainable funds for team coordination, infrastructure or clinic costs • Wait times only 3-4 months	• Patient cancellations/ no-shows are common
McFarlane et al. 2011	Mobile diagnostic team and follow-up support personnel	• Average wait time of 3 months once the initial pre-clinic work is completed • Mobile team provides specialised hospital/services that are often not available in rural communities	• Availability of rural professionals both as primary team members and filling back up positions • Adult patients do not often have stable support network – may not always be able to attend scheduled appointments, so every clinic must have back up patients
Panton et al. 2022	• Multi-disciplinary teams • CDU – created 3 models of care covering women's education (focus on women's health and prevention), ages 0-6, and ages 7-18 • FASD Tasmania – 3 MoCs to cover different age ranges (0-4years – infants, 4-12 years – children, 12+years – young people) • Goulburn Valley Health Service – 4 MoCs (0-5 years – early childhood, 5-16 years – education and 10-16 years – justice) • MDT colocation important for success	• Sustainable sites noted strong executive support ("enthusiasm" and "proactive response", "active" involvement, "good team leadership" and "lobby[ing] hard" for funding" was key for success • Sites that have access to MDT (hospital-based) had greater success with clinic roll-out • Increased local FASD awareness • Improved reporting on national prevalence due to increased reporting of diagnoses • Increased local networks (developed through the CRG, community engagement sessions and local clinical training)	• Not all clinicians available for diagnostic assessments at hospitals (FASD Tasmania) – "geographically challenging for clinicians to attend" • Ensuring that MoC would be clinically useful for the region and target services • Gaining buy-in and traction for the MoC through local stakeholders and clinicians • High staff turnover in key roles (coordinator, paediatrician and psychologist) was barrier to success • Attempting to embed a specialist FASD diagnostic service within a large metropolitan primary health care setting (FASD Tasmania and DDHS) • Low support from clinicians created a barrier to developing an agreed MoC • Lack of appropriate planning for local areas – not able to apply national models to all areas

			• Difficulty maintaining community engagement, esp. with agencies resisting FASD diagnosis • Involving affect regulation and occupational therapy (motor processing, sensory processing and visual-motor integration) in the diagnostic process efficiently was challenging as these domains were not often adequately assessed • Accurately obtaining information about PAE from the biological mother • Inadequate funding • Reluctance from paediatricians to diagnose children (issues around ethical issues, maternal consent and stigma) – important to provide training and education to address these issues
Patel et al. 2020	Collaborative care model	• Overcomes barrier of limited access to specialised diagnostic teams by tying together pre-existing services offered by community children's welfare agency to facilitate the process of FASD diagnosis and treatment • 78% of youth who were suspected received a diagnosis • More timely diagnosis of FASD and identification and treatment recommendations for psychiatric co-morbidity in youth with FASD	• Youth with more subtle presentations were not scored on the screening tool and may have been missed
Temple et al. 2015	Embedded model of care	• Clinicians necessary for team are already part of agency • Reduce infrastructure costs – materials, office space, meeting rooms and administrative supports already exist within host agency • Patient support requirements already offered within developmental sector	• Patient cancellations/ no-shows are common • Some patients may not want to be involved as they do not want to be identified as having an intellectual or developmental disability • Some service providers may see individuals with FASD and intellectual disability as not fitting their mandate and be reluctant to include them in programs, lead to challenges with finding appropriate interventions and follow-up services