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Original Article

Reduced effectiveness of the selective hip dysplasia screening programme over time

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ABSTRACT

INTRODUCTION. Selective ultrasound screening for developmental dysplasia of the hip (DDH) was implemented in the North Denmark Region, Denmark in 2010 but has yet to be evaluated. This study aimed to evaluate selective hip screening in the North Denmark Region from 2010 to 2021, focusing on referral rates, the incidence of DDH and referral causes.

METHODS. We retrospectively identified 3,812 infants (2,091 females) who were referred during a 12-year period for hip ultrasound at Aalborg University Hospital in Denmark. DDH was defined as an ultrasound classification of Graf type IIb or worse. Referral rates and the incidence of DDH and their development over the study period were examined.

RESULTS. During the study period, there was a steadily increasing referral rate, with the annual referral rate doubling from 200 (3.3%) in 2010 to 402 (6.8%) in 2021, whereas the incidence rate increased only minimally from 0.15% to 0.22%. The most common referral causes were breech position (41.4%) and positive clinical exam (27.8%).

CONCLUSIONS. During the study period, the annual referral rate nearly doubled without a corresponding increase in DDH incidence, suggesting that the screening programme has become less efficient over time. This might explain the relatively low incidence of DDH in the North Denmark Region compared with national and international studies.

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TRIAL REGISTRATION. This study was registered in the North Denmark Regional internal Project Register as IDF2022-187.

Developmental dysplasia of the hip (DDH) is a disorder characterised by underdevelopment of the hip joint. It covers a range of abnormalities, from mild deformities of the acetabulum or femur to complete hip dislocation. DDH is the leading congenital musculoskeletal disorder among newborns, with an incidence rate of approximately 1.0% [1]. When DDH is detected early (< 6 months), treatment typically involves using braces, harnesses or casts [2]. However, delayed diagnosis and treatment might cause soft-tissue contractures and bone deformities in the acetabulum or femur that can lead to fixed hip positions, which require more extensive surgical procedures to achieve stabilisation [3]. The long-term effects of these deformities may be reduced hip function and a higher risk of developing premature arthritis, leading to early total hip replacement [3].

Since 2010, the North Denmark Region has conducted targeted ultrasound (US) screening for DDH. All newborns are screened for DDH shortly after birth by a midwife or paediatrician and at a five-week routine check-up by a general practitioner (GP). According to national guidelines, the screening consists of clinical examinations, including the Barlow and Ortolani manoeuvres, the Galeazzi test and a test for limited hip abduction [4]. In addition, all newborns are screened for DDH risk factors. If the clinical screening and/or risk factor screening

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are positive, the newborn is referred to Aalborg University Hospital (AAUH) for a confirmatory hip US, within six weeks (or within two weeks if clinical instability is detected), thus following international protocols for selective DDH screening.

Few studies have examined the performance of the Danish selective US screening programme for DDH, and none have examined its effectiveness over time [2, 5].

The aim of this study was to evaluate selective hip screening in the North Denmark Region from 2010 to 2021, focusing on the development of referral rates for hip US, the causes of referral and the incidence of sonographically verified DDH, defined as a Graf type IIb or above.

METHODS

Study design and setting

This was a retrospective cohort study of infants referred for hip US at AAUH in the North Denmark Region over a 12-year period from 1 January 2010 to 31 December 2021. Reporting follows the STROBE guidelines for observational studies [6].

Participants

The inclusion criteria consisted of infants examined with a hip US at AAUH suspected of DDH.

The patients' hip US examination reports and referral information were identified through a search in the electronic patient record system (EPJ) using the examination code for hip US (UXUG15) and/or diagnosis codes for DDH (DQ658F, DQ656, DR294, DQ652, DQ650, DQ651, DQ658, DQ658D, DQ658E, DM243, DQ659, DQ655). These codes are part of the Danish national health classification system, which is based on the ICD-10 and NOMESCO classification systems for diagnostic and procedure codes.

The exclusion criteria included duplicate referrals, control US examinations and referrals made for examinations unrelated to DDH.

A search of the Statistics Denmark database identified 68,321 live births in the North Denmark Region during the study period.

Variables

Referred infants received a hip US by a radiologist or specifically trained radiographer. The hip US was performed according to the methodology by Graf, with alpha angles measured with the infant in the lateral decubitus examination position [1].

Based on the reported alpha angles, we defined a Graf type for each hip according to Graf's classification [7]. DDH was defined as Graf type IIb or worse.

Data sources

During the study period, the risk factors used as referral criteria for hip US in the North Denmark Region were: breech presentation in utero or during birth, multiple births, family history of DDH, oligohydramnios, syndromes, and congenital deformities, e.g., clubfeet and torticollis. Some infants were referred due to other factors such as asymmetrical skin folds and malformation of the os sacrum. These patients are included in this study despite not fitting the regional referral guidelines and are recorded as “other”.

Referrals based on first-degree relatives with DDH and referrals described as 'familial disposition' were logged as familial disposition. Referrals based on second- or third-degree relatives with DDH were logged as 'other'.

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Finally, referrals based on other factors unrelated to the regional referral guidelines were logged as “other”.

We defined a positive clinical hip examination as any positive clinical finding identified by the primary screener, whether it results from specific tests (Barlow, Ortolani, Galeazzi) or from non-specific findings (hip 'click', limited hip abduction, or other non-specific positive signs).

Data for the study were automatically extracted from the EPJ into an Excel spreadsheet and subsequently manually reviewed and entered into a secure, GDPR-compliant REDCap database hosted at the AAUH.

Statistical methods

Patient demographics, referral rates and US findings underwent descriptive analysis. Continuous variables were presented as means and SD. Normality was confirmed by visual inspection of histograms. Reasons for referral and hip US classifications are presented as counts and percentages. The incidence is measured at the patient level, with the worst-affected hip recorded. Incidence rates of DDH were calculated as the number of patients with at least one Graf type IIb hip/number of infants born. As only a few infants had bilateral DDH (n = 24), this approach had only a minimal impact on the overall incidence and was used to ensure comparability with existing literature [1].

Differences in the proportional distribution of referral causes by year were analysed using chi-square statistics.

Two bar charts were constructed; one depicting the incidence of DDH to the referral rate for hip US from 2010 to 2021, including referrals for infants with multiple referral reasons, and another showing the proportions of each referral reason relative to the total.

A significance level of 5% was applied. All analyses were performed in STATA version 18.0 (StataCorp, College Station, TX, USA).

Trial registration: This study was registered under the internal North Denmark Regional Project Register IDF2022-187.

RESULTS

Participants

During the 12-year study period, 68,321 infants were born in the AAUH catchment area, and 4,778 hip US examinations were performed. Of these examinations, 966 were excluded (duplicate examinations (n = 269), unrelated referrals (n = 73), control hip US (n = 624)), resulting in a total of 3,812 infants undergoing a primary hip US exam (7,624 hips), forming the study population (**Figure 1**). The study population consisted of 55% females, and the mean age at US examination was 46 days (SD: 30 days).

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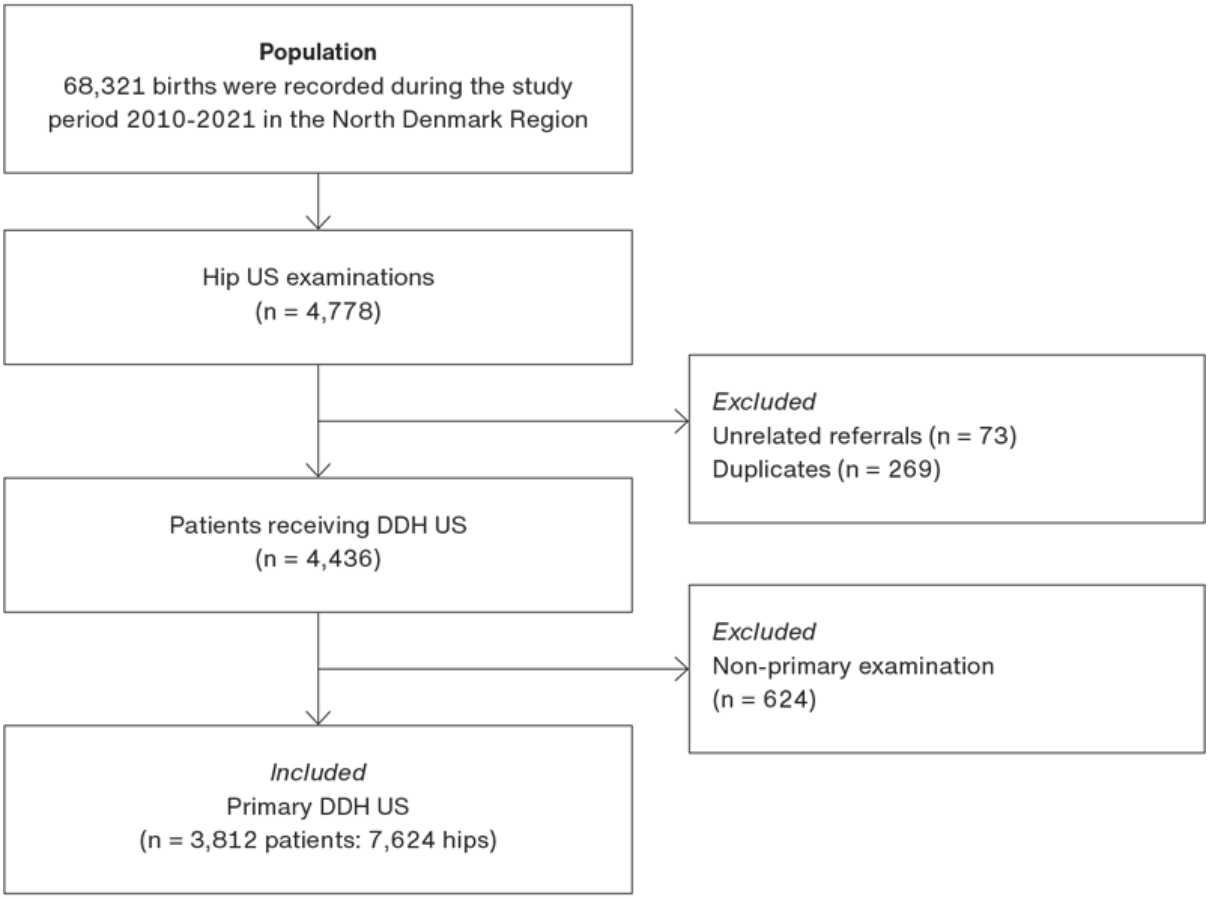
FIGURE 1 Flow chart of the study population inclusion process.



DDH = developmental dysplasia of the hip; US = ultrasound.

Among the 3,812 infants, six had missing referrals. A total of 3,407 infants had a single referral cause, 381 had two referral causes and 18 infants had three referral causes.

Among single referral causes, the most common were breech position (41.5%) and positive clinical exam (27.8%). Among infants with two referral causes, the most common combination was breech position + twin (4.6%). **Table 1** presents the birth rate, referral rate and US hip types, categorised by year.

TABLE 1 Births, referral rates and incidence rates of Graf types and developmental dysplasia of the hip during the study period, 2010-2021

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	Total
Primary hip US examinations, n (%)	200 (5.2)	232 (6.1)	228 (6.0)	235 (6.2)	240 (6.3)	313 (8.2)	341 (8.9)	374 (9.8)	378 (9.9)	429 (11.3)	440 (11.5)	402 (10.5)	3,812 (100)
Births, n	6,032	5,653	5,630	5,221	5,346	5,535	5,837	5,841	5,820	5,817	5,650	5,939	68,321
Referral rate	3.3	4.1	4.0	4.5	4.5	5.7	5.8	6.4	6.5	7.4	7.8	6.8	5.6
Graf hip types, n (%)													
Type I	152 (76.0)	192 (82.8)	188 (82.8)	202 (86.3)	212 (88.3)	291 (93.0)	308 (90.3)	329 (88.0)	338 (89.4)	386 (90.0)	397 (90.4)	380 (94.5)	3,375 (88.6)
Type IIa	39 (19.5)	31 (13.4)	30 (13.2)	26 (11.1)	21 (8.8)	21 (6.7)	29 (8.5)	32 (8.6)	38 (10.1)	34 (7.9)	34 (7.7)	9 (2.2)	344 (9.0)
Type IIb	3 (1.5)	2 (0.9)	1 (0.4)	1 (0.4)	1 (0.4)	0	1 (0.3)	1 (0.3)	0	1 (0.2)	0	0	11 (0.3)
Type IIc	5 (2.5)	5 (2.2)	7 (3.1)	3 (1.3)	4 (1.7)	1 (0.3)	1 (0.3)	8 (2.1)	2 (0.5)	6 (1.4)	1 (0.2)	8 (2.0)	51 (1.3)
Type III	1 (0.5)	2 (0.9)	1 (0.4)	2 (0.9)	2 (0.8)	0	2 (0.6)	4 (1.1)	0	2 (0.5)	7 (1.6)	5 (1.2)	28 (1.3)
DDH, n	9	9	9	6	7	1	4	13	2	9	8	13	90
IR of DDH, %	0.15	0.16	0.16	0.11	0.13	0.02	0.07	0.22	0.03	0.15	0.14	0.22	0.13

DDH = developmental dysplasia of the hip; IR = incidence rate; US = ultrasound.

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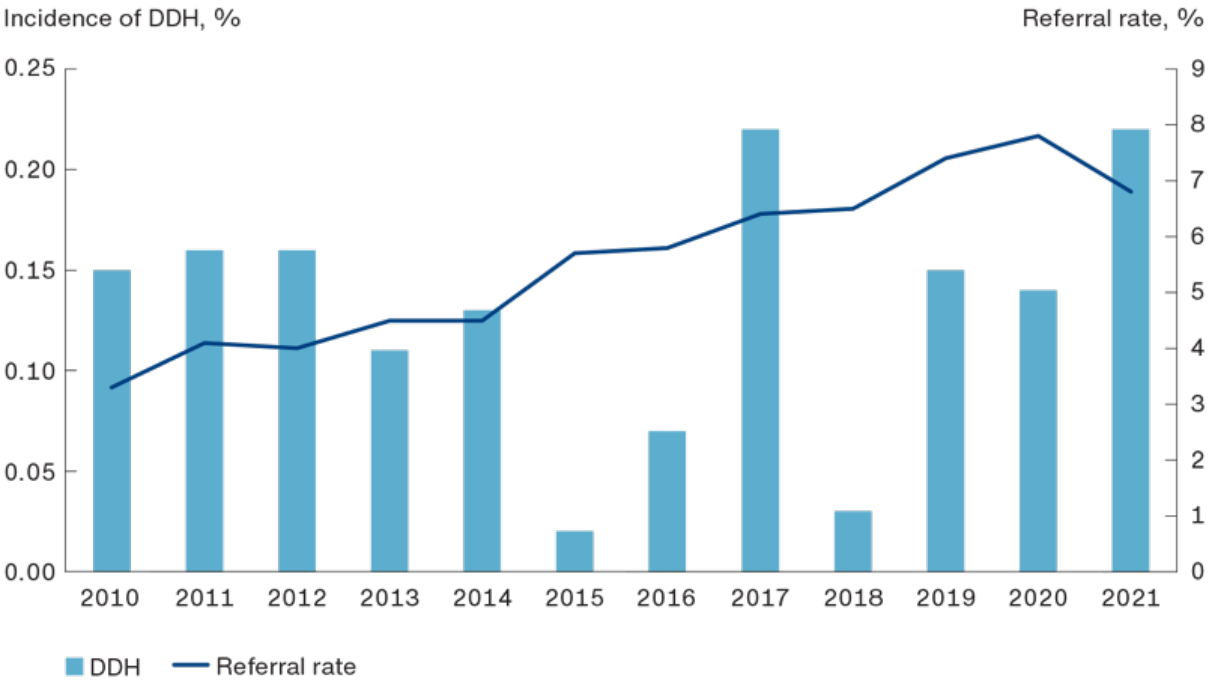
During the study period, the referral rate rose from 3.3% (200 newborns) in 2010 to 6.8% (402 newborns) in 2021. While the incidence of DDH stayed nearly unchanged (0.15-0.22%), the proportion of normal Graf type I hips rose from 76.0% in 2010 to 94.5% in 2021 (**Figure 2**, Table 1).

FIGURE 2 Referral rate and incidence of developmental dysplasia of the hip (DDH) during the study period, 2010-2021. DDH was defined as a Graf type ≥ IIb.



In total, 90 patients were diagnosed with DDH over the 12-year study period, resulting in an overall DDH incidence of 0.13%, ranging from an annual incidence rate of 0.02% in 2015 to 0.22% in 2017. Figure 2 illustrates the DDH referral rate and incidence during the study period.

During the study period, a significant change was observed in all referral causes ($p < 0.001$), except for oligohydramnios and syndromes. The proportion of referrals due to positive clinical examination, breech presentation and family history changed markedly. In 2010, these accounted for 47.5%, 31.7%, and 5.9% of referrals, respectively. By 2021, these figures had shifted to 32.2%, 45.8%, and 16.4% (**Figure 3** and Supplementary table).

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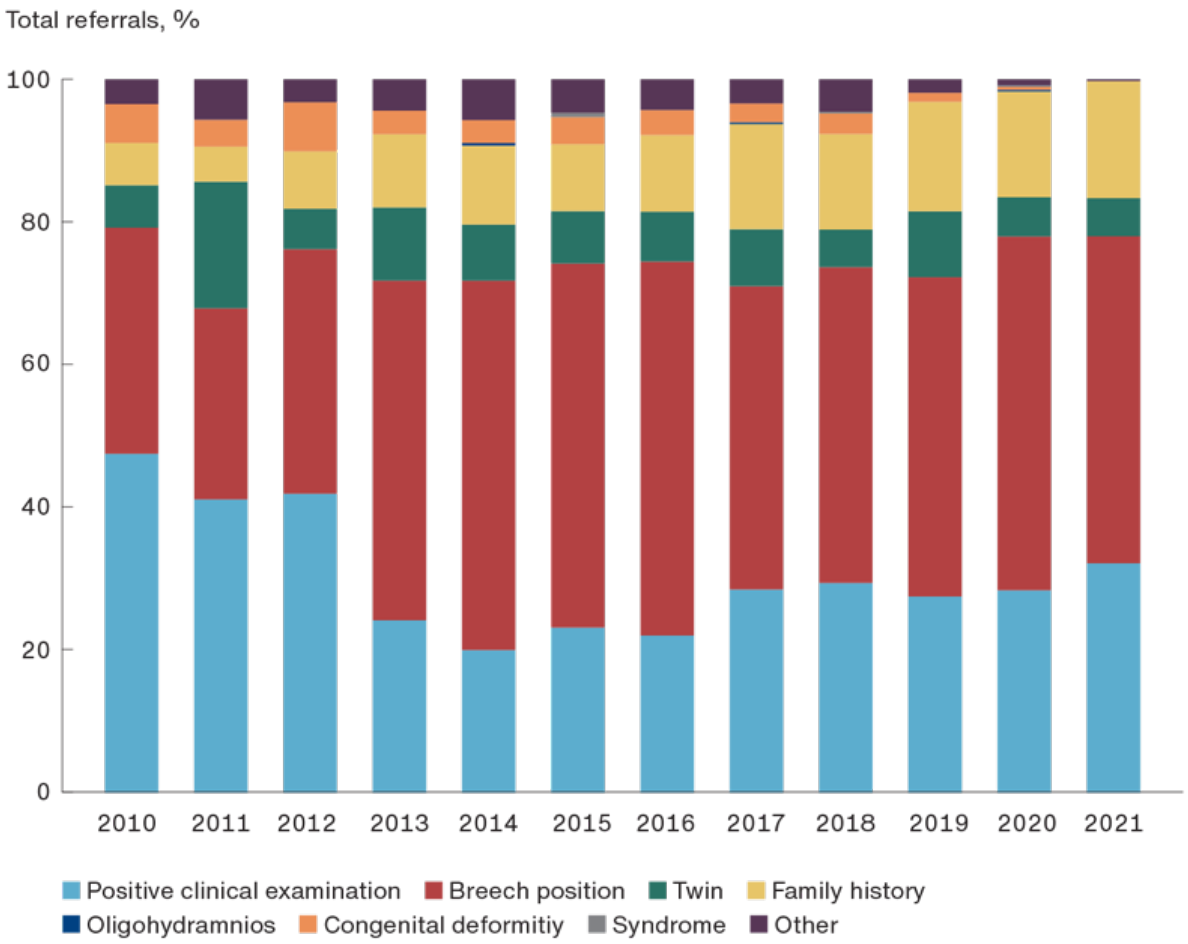
FIGURE 3 Referral causes (also see the Supplementary table).



DISCUSSION

In the 12 years following the implementation of selective US screening for DDH in the North Denmark Region, the DDH incidence rate was 0.13%. However, the annual referral rate nearly doubled during this period without a corresponding increase in incidence, indicating a markedly reduced screening programme efficiency.

Despite having twice the referral rate of other selective screening programmes, the present study found a low incidence rate compared with national and international studies [8].

Other studies of the Danish selective screening programme for DDH reported higher incidence rates of 0.41% and 0.47% [2, 5]. However, these studies defined DDH as a Graf type IIb classification or worse and/or sonographic instability, which may have contributed to a higher incidence rate.

A meta-analysis of 76 studies, including 16.9 million infants, found a mean DDH incidence rate of 2.3% with universal ultrasonographic screening and 0.4% with selective ultrasonographic screening. However, the incidence rates ranged from 0.02% to 12.0%, which may, in part, be attributable to variations in the definition of DDH across the 76 included studies [9].

This highlights the difficulties that arise when comparing DDH incidences and referral rates across different settings and/or definitions of DDH. However, it does seem that DDH screening in the North Denmark Region has

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a lower detection rate than reported in the literature and a higher-than-average referral rate.

Although the number of infants referred for screening increased during the study period, the DDH incidence rate remained nearly unchanged, signifying an increase in unnecessary referrals for hip US.

The declining efficiency of the screening programme may be explained by the inappropriate referral of infants not meeting established risk criteria (e.g. a family history of DDH in second-degree relatives or asymmetrical skin folds), alongside insufficient referral of those who did meet the criteria (e.g. oligohydramnios), suggesting inconsistencies in guideline adherence and or limited knowledge of correct hip examination techniques.

Recognition of region-specific referral criteria for DDH US screening in Denmark is low [10].

In our study, only three infants were referred due to oligohydramnios, even though oligohydramnios complicates 0.5-5.5% of pregnancies [11]. This aligns with the findings of Husum et al., who reported that oligohydramnios is not widely recognised by midwives, GPs and GPs in training as a DDH referral criterion in the Danish selective screening programme for DDH [10].

Furthermore, primary screeners in the Danish DDH screening programme have poor recognition of recommended hip examination techniques [12] and a low positive predictive value for their clinical hip assessments [13].

Combined, the low recognition of risk factors used as referral criteria in DDH screening and a limited understanding of clinical hip screening may explain the relatively high referral rate and low incidence rate of DDH found in this study.

Future studies should evaluate cases of late diagnosed DDH to determine the long-term effectiveness of the screening protocol.

Limitations

In our study, if an infant was in breech position at any point during the pregnancy and a screener referred for that reason, the case was categorised as 'breech position'. However, breech position is a predisposing risk factor only in late pregnancy and during birth [14].

Referrals stating 'familial predisposition' without further specification of which family member had DDH were logged as first-degree family members. This may have led to referrals that included second- or third-degree familial relationships, even though established guidelines state that only first-degree familial predisposition is a valid basis for referral [15].

A total of 183 (4.8%) patients received a primary DDH US without meeting the screening criteria. While the inclusion of these non-risk patients increases the calculated referral rates without increasing incidence rates, we chose to include them to reflect the clinical reality of DDH screening, as non-adherence to screening guidelines may be a driving factor in declining screening efficiency.

CONCLUSIONS

We found an almost doubled annual referral rate without an increase in incidence rate, indicating that the selective screening programme in the North Denmark Region has become less efficient over time.

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