


Systematic Review

Giant Sacrococcygeal Teratoma in Infant: Systematic Review

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Abstract**Introduction**

Sacrococcygeal teratoma (SCT) is a rare embryonal tumor that occurs in the sacrococcygeal region, with an incidence of about 1 in 35,000 to 40,000 live births. This study presents a systematic review of giant SCT greater than 10 cm.

Methods

A systematic review of published studies regarding giant SCT in infants was conducted. The studies included met the following criteria: 1) the teratoma was situated in the sacrococcygeal region; 2) all case reports involved infants with a teratoma larger than 10 cm, and 3) the size of the teratoma was verified to exceed 10 cm through diagnostic methods.

Results

The current study included 31 studies that met the inclusion criteria. The studies included patients aged 6.1 to 9.3 months, with a median age of 7.6 months, predominantly female (72.7%). Diagnoses were primarily made in the first and second trimesters (39%) or after birth (33.3%), with cesarean delivery being the most common method (66.7%). Tumors weighed between 1.5 and 5 kg, with an average diameter of 15.6 cm. Surgical resection was performed in 93.9% of cases. The most common complication was respiratory failure (30.3%), and histopathology revealed that 39.4% of tumors were immature teratomas, while 33.3% were mature teratomas. The overall survival rate was 66.7%, with 18.2% of survivors experiencing tumor recurrence. Most complications occurred in the second trimester; however, no significant associations were found concerning the timing of diagnosis. Additionally, tumor size did not significantly impact outcomes.

Conclusion

Routine ultrasound and MRI are essential for the antenatal diagnosis of SCT. Due to the high risk of morbidity with larger tumors, cesarean delivery is advised for tumors over 10 cm. Coccygectomy is the most effective approach to prevent recurrence, highlighting the importance of timely surgical intervention and ongoing follow-up.

1. Introduction

Sacrococcygeal teratoma (SCT) is an uncommon embryonal tumor form in the sacrococcygeal region. It affects around one

among every 35,000 to 40,000 live births [1]. The condition is significantly more common in females, with a female-to-male

ratio of 3:1 to 4:1. Teratomas consist of tissues originating from all three germ layers: ectoderm, endoderm, and mesoderm [2, 3]. Although their exact embryonic origin remains uncertain, SCTs are believed to arise early in gestation from totipotent cells in Hensen's node, a remnant of the primitive streak in the coccygeal region [2]. SCTs can vary significantly in size, with some growing large enough to cause noticeable anatomical changes, such as the anterior displacement of the anus and resulting in clinical symptoms like anal displacement, tightening of the anal canal, and constipation, often due to tumor compression of the bladder or rectum [1, 3]. Obstetric ultrasound during the second trimester is a crucial tool for making antenatal diagnoses of tumors, helping to prevent perinatal and neonatal complications such as fetal hydrops, tumor rupture, placentomegaly, and high-output cardiac failure, all of which are linked to a higher risk of mortality [4, 5]. In 1973, according to the American Academy of Pediatrics Surgical Section (AAPSS), Altman classified SCTs into four types based on the tumor's intrapelvic and intra-abdominal extension and external components. SCTs observed at birth are typically classified as Altman Type I or II, with Type III being rare, while Type IV is usually identified later in life [2]. The management of SCT involves surgical excision, and the likelihood of recurrence after complete removal is minimal [5], leading to a favorable prognosis. However, long-term follow-up is essential to monitor for recurrence [3].

While there have been reports of large SCTs, to the best of our knowledge, based on an extensive literature review and previous case reports, no systematic reviews specifically address SCTs greater than 10 cm in infants. All included references were confirmed for eligibility [6].

2. Methods

2.1. Study design

The present systematic review adhered to the preferred reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.

2.2. Data sources and search strategy

A comprehensive review of all published studies on SCT was conducted by searching databases such as Google Scholar, PubMed/MEDLINE, Cochrane Library, ScienceDirect, CINAHL, Web of Science, and EMBASE. The search used the following keywords: (sacroccygeal) AND (teratoma OR tumor) in combination with (antenatal OR neonate OR infancy OR infant OR child OR children OR newborn OR pediatric).

2.3. Eligibility criteria

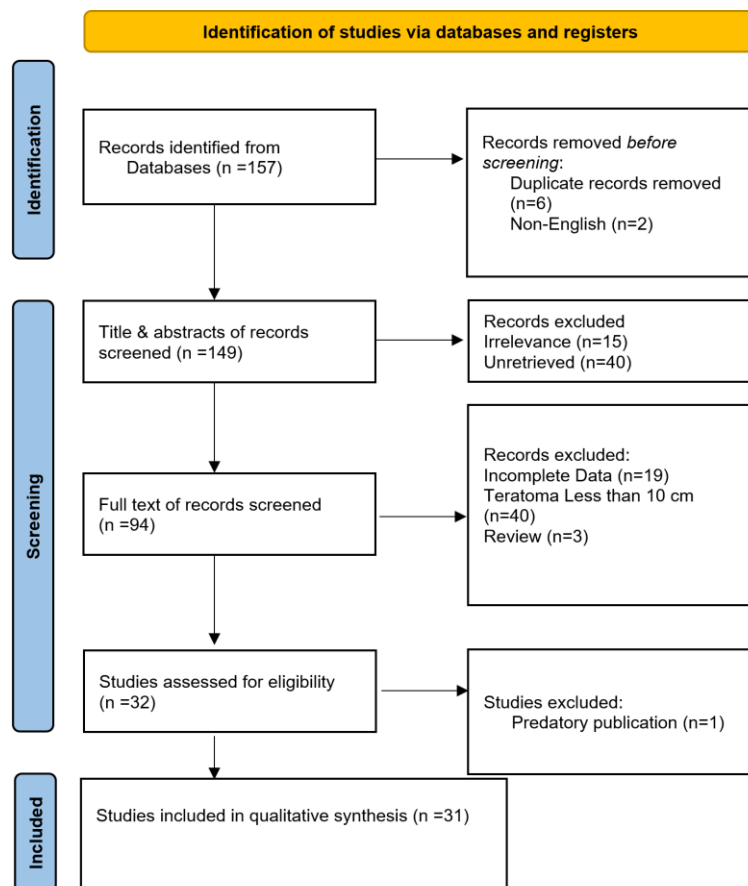


Figure 1. Study selection PRISMA flow chart.

Table 1. Raw data of each included study.

Author [reference]	Year	Country	Gender	Age of delivery (days)	Diagnosis time	Mode of delivery	Weight (kg)	Size of tumor (cm)	Altman classification	Complication	Histopathology	Survival	Recurrence
Chamberlin et al. ^[7]	N/A	USA	N/A	224	1 st trimester	CS	3.7	13.9	N/A	Near cardiac arrest & hypoxia	N/A	Yes	N/A
Sabir et al. ^[11]	2023	Iraq	F	266	2 nd trimester	CS	5.0	17.5	1	None	Immature teratoma	Yes	No
Dey et al. ^[8]	2023	USA	F	206	Prenatal (Unknown trimester)	CS	2.4	11.6	3	Poor respiratory effort	Immature teratoma	Yes	No
Abou-Bekr et al. ^[4]	2022	Algeria	F	N/A	Postnatal	CS	N/A	10	1	None	Mature teratoma	Yes	N/A
Koc et al. ^[9]	2022	Turkey	F	209	N/A	N/A	2.2	10	1	None	Teratoma (unknown type)	Yes	N/A
Meshram et al. ^[2]	2021	India	M	259	2 nd trimester	CS	2.2	24.5	2	Polyhydramnios	Mature teratoma	No	N/A
Zlatan et al. ^[10]	2021	Bosnia and Herzegovina	M	238	2 nd trimester	CS	2.3	15.5	2	Cardiac arrest	Immature teratoma	Yes	N/A
Guitart et al. ^[11]	2020	Spain	N/A	245	2 nd trimester	CS	4.0	18	1	Premature rupture of the membranes & abnormal cardiotocography	Immature teratoma	Yes	N/A
Savitri et al. ^[12]	2019	Indonesia	F	266	2 nd trimester	CS	4.2	11.2	1	Resuscitation needed	Teratoma (unknown type)	Yes	No
Singhal et al. ^[13]	2018	India	F	196	Postnatal	NVD	3.6	16	N/A	Infection	Mature teratoma	N/A	N/A
Konopliitskyi et al. ^[4]	2018	Ukraine	F	280	3 rd trimester	CS	4.3	14.9	1	N/A	Immature teratoma	Yes	Yes
Sop Lee et al. ^[15]	2017	Korea	M	199	2 nd trimester	CS	2.9	12.3	3	Poor respiratory distress	Immature teratoma	Yes	No
Bechtel et al. ^[16]	2014	USA	N/A	226	Prenatal (Unknown trimester)	CS	3.7	13.9	2	Lower limb flaccid paralysis, scoliosis	Immature teratoma	Yes	Yes
Mondal et al. ^[17]	2014	India	F	245	Postnatal	NVD	3.5	16.7	1	Bleeding	N/A	No	N/A
Mbumba et al. ^[18]	2010	France	F	266	1 st trimester	CS	N/A	16	N/A	None	Mature teratoma	N/A	N/A
Roka et al. ^[3]	2010	Nepal	F	N/A	Postnatal	N/A	N/A	22.4	N/A	None	Mature and yolk sac components (Schiller-Duval bodies)	Yes	N/A
Lahdes-Vasama et al. ^[19]	2010	Finland	F	210	2 nd trimester	CS	3.4	16.9	2	Bleeding	Immature teratoma with a malignant component in a small area	Yes	No
Abraham et al. ^[20]	2010	USA	F	238	2 nd trimester	CS	N/A	18.9	3	Premature rupture of membranes & poor respiratory effort	N/A	Yes	N/A

Table 1. Continued...

Den Otter et al. ^[21]	2007	Netherlands	F	192	2 nd trimester	CS	2.2	11	2	Respiratory failure	Mixed teratoma	Yes	Yes
Howman-Giles et al. ^[22]	2007	Australia	F	N/A	Postnatal	N/A	N/A	16	N/A	Left iliac lymph node enlargement, colostomy needed	Immature teratoma	No	Yes
Hosono et al. ^[23]	2004	Japan	F	252	3 rd trimester	CS	4.0	13.1	1	Poor respiratory efforts	Immature teratoma	Yes	No
Ribeiro et al. ^[24]	1999	France	F	266	Postnatal	CS	2.7	11.4	1	None	Mature teratoma	Yes	No
Jona et al. ^[25]	1999	USA	F	189	Prenatal (Unknown trimester)	CS	1.7	26	N/A	N/A	Teratoma (unknown type)	No	N/A
Johnston ^[26]	1998	USA	N/A	245	N/A	N/A	3.9	12.2	N/A	N/A	Mature elements, except for immature neuroectodermal tissue.	Yes	N/A
Robertson et al. ^[27]	1995	USA	F	185	2 nd trimester	CS	1.8	N/A	2	Severe respiratory distress syndrome	Mature teratoma	Yes	No
Lnoue et al. ^[28]	1994	Japan	M	210	2 nd trimester	CS	3.8	10	N/A	Bleeding and circulatory failure	Immature teratoma	Yes	N/A
Nakayama et al. ^[29]	1991	USA	F	231	3 rd trimester	CS	4.3	13.9	2	Respiratory insufficiency & renal failure	Immature teratoma	N/A	N/A
	1991	USA	F	210	Postnatal	CS	3.0	15	3	Respiratory distress	Immature teratoma	N/A	N/A
Worsham et al. ^[30]	1975	USA	F	189	Postnatal	NVD	1.9	13.1	N/A	No respiratory efforts	N/A	No	N/A
Williams et al. ^[31]	1970	Nigeria	F	266	Postnatal	N/A	N/A	30	N/A	Metastasis	Teratoma (unknown type)	Yes	Yes
Schiffer et al. ^[32]	1956	USA	F	196	3 rd trimester	NVD	1.5	20	N/A	Difficult birth	N/A	N/A	N/A
	1956	USA	N/A	N/A	Postnatal	NVD	N/A	15	N/A	Difficult birth	N/A	N/A	N/A
Walker et al. ^[33]	1950	USA	F	224	Postnatal	NVD	2.7	12.5	N/A	N/A	Teratoma (unknown type)	Yes	N/A

N/A: non-available, F: female, M: male, CS: cesarean section, NVD: normal vaginal delivery

The non-English studies and those unrelated to humans were excluded before or during the initial screening. Studies on SCT were included if they met the following criteria: 1) the teratoma was located in the sacrococcygeal region; 2) all case reports involved infants with a teratoma larger than 10 cm, and 3) the teratoma size was confirmed to be over 10 cm through diagnostic methods. Studies published in predatory journals (inappropriately peer-reviewed) and those not meeting the inclusion criteria were excluded.

2.4. Study selection and data extraction

The titles and abstracts of the identified studies were initially screened. This was followed by a comprehensive review of the full text to assess eligibility. Data points collected from the included studies included the year and country of the study, patient age and gender, age at delivery, trimester of detection, postnatal diagnosis, mode of delivery, tumor size and weight, imaging technique, complications, and recurrence rates.

2.5. Statistical analysis

The data were first utilized in a qualitative synthesis, followed by quantitative re-analysis using the Chi-square test and Fisher's exact test for categorical variables and the independent sample t-test for numerical data, conducted with SPSS software version 25.0. A significance level of 0.05 was not established.

3. Results

3.1. Study selection and characteristics

A systematic search initially identified 157 articles. Before screening, eight were excluded due to duplication and non-English language. During the initial title and abstract screening, 55 studies were excluded as they did not meet the inclusion criteria. Subsequently, 94 studies underwent full-text screening, and 32 were further assessed for eligibility. Ultimately, 31 studies [1- 4, 7- 33] were compatible with the inclusion criteria and included in the final analysis (Figure 1). All the studies included were case reports (Table 1).

3.2. Patient characteristics, diagnosis, management and outcome

The included studies characterized patients with delivery ages ranging from 6.1 to 9.3 months, with a median age of 7.6 months. Most patients were female (72.7%), while 12.1% were male, and in 15.2% of cases, gender was unspecified. Diagnoses were predominantly made during the first and second trimesters (39%) or after birth (33.3%), with cesarean section as the most common delivery mode (66.7%). Tumor weights ranged from 1.5 to 5 kg, with an average diameter of 15.6 cm. Surgical resection was the primary treatment, performed in 93.9% of cases, although two patients died before surgery could be undertaken. Complications were frequent, with respiratory failure as the most common (30.3%). Histopathological analysis indicated that 39.4% of tumors were immature teratomas and 33.3% were mature teratomas. The overall survival rate was 66.7%, and 18.2% of surviving patients experienced tumor recurrence (Table 2).

Table 2. The baseline characteristics of the study.

Variables	Frequency / Percentage
Patient demographics	
Age at delivery time, range (median, mean \pm SD), month	6.1 – 9.3 (7.6, 7.6 \pm 0.97)
Gender	
Male	4 (12.1%)
Female	24 (72.7%)
N/A	5 (15.2%)
Diagnosis time	
In the first trimester	2 (6.1%)
In the second trimester	11 (33.3%)
In the third trimester	4 (12.1%)
Diagnosed prenatally with unknown trimester	3 (9.1%)
Postnatal	11 (33.3%)
Unknown	2 (6.1%)
Mode of delivery	
Cesarean section	22 (66.7%)
Normal vaginal delivery	6 (18.2%)
Unknown	5 (15.1%)
Weight of the tumor (kg), range (mean \pm SD)	1.5 – 5 (3.1 \pm 0.96)
Tumor size (cm), range (mean \pm SD)	10 – 30 (15.6 \pm 4.77)
Group of tumors (Altman classification)	
Group 1	9 (27.3%)
Group 2	7 (21.2%)
Group 3	4 (12.1%)
Unknown	13 (39.4%)
Management	
Surgical resection	31 (93.9%)
Patient died before surgery	2 (6.1%)
Complications	
Yes	21 (63.6%)
No	10 (30.3%)
Unknown	2 (6.1%)
Common perioperative complications*	
Respiratory failure	10 (30.3%)
Bleeding	3 (9.1%)
Cardiac failure/problem	3 (9.1%)
Infection	2 (6.1%)
Histopathology	
Immature teratoma	13 (39.4%)
Mature teratoma	11 (33.3%)
Mixed teratoma	3 (9.1%)
Unknown	6 (18.2%)
Survival	
Yes	22 (66.7%)
No	5 (15.1%)
Unknown	6 (18.2%)
Recurrence among survived cases	
Yes	4 (18.2%)
No	8 (36.4%)
Unknown	10 (45.4%)

*Other complications may have been reported

N/A: non-available, SD: standard deviation

3.3. Complications and recurrence

The comparative analysis of complications assessed the trimester of diagnosis, timing of diagnosis, and recurrence rates. Most complications occurred in the second trimester (10 cases),

Table 3. Comparison of trimester at diagnosis, diagnosis time, and recurrence with complication.

Variables	Complication		Total	P-value
	Yes	No		
Trimester at diagnosis	First	1	1	0.34
	Second	10	1	
	Third	3	1	
Diagnosis time	Prenatal	16	4	0.10*
	Postnatal	5	6	
Recurrence	Yes	2	2	0.55*
	No	6	2	

CS; cesarean section, NVD; normal vaginal delivery

*Fisher's exact test

with fewer cases reported in the first and third trimesters; however, the p-value of 0.34 indicates no significant difference across trimesters. Diagnostic timing was categorized into prenatal and postnatal periods. Complications were more common in the 20 cases diagnosed prenatally (16 cases) than in postnatal diagnoses, but Fisher's exact test yielded a p-value of 0.10, suggesting no statistically significant association. For recurrence, complication rates were similar in both recurrence and non-recurrence groups, with a p-value of 0.55, supporting the lack of a substantial correlation. Overall findings reveal no strong statistical relationships between complications and diagnostic timing, trimester of diagnosis, or recurrence (Table 3). The influence of tumor size and weight on complications, recurrence, and survival is demonstrated. The mean tumor size was slightly larger in cases with complications (15.02 ± 3.49 cm) compared to those without complications (14.29 ± 3.87 cm), but the difference was not statistically significant ($p = 0.6$). Recurrence was associated with larger tumors (17.16 ± 7.41 cm) relative to non-recurrent cases (13.42 ± 2.66 cm), although this difference was not significant ($p = 0.14$). Similarly, non-survivors exhibited a higher mean tumor size (19.25 ± 5.65 cm) compared to survivors (14.6 ± 4.84 cm), but the association was not statistically meaningful ($p = 0.41$). For tumor weight, no significant variations were found across outcomes. The mean tumor weight in cases with complications was 3.03 ± 0.88 kg versus 3.47 ± 1.09 kg in those without ($p = 0.39$). Likewise, tumor weight did not show significant differences for recurrence ($p = 0.92$) or survival ($p = 0.35$). The findings indicate that neither tumor size nor weight significantly impacted complications, recurrence, or survival (Table 4).

4. Discussion

SCTs are rare embryonal tumors that develop in the sacrococcygeal region of newborns, the most common site for

germ cell tumors. While SCTs occur more frequently in females—with 72.7% of cases in this systematic review involving female patients, they tend to show a higher malignancy rate in males [16]. Most of these tumors are histologically benign and can be classified into three main types: mature teratomas, which consist of fully differentiated tissues such as bone, teeth, and hair; immature teratomas, which contain embryonal elements or partially differentiated structures that pose a significant risk of malignancy; and malignant teratomas, which include one or more malignant germ cell tumors such as yolk sac tumors, choriocarcinomas, and embryonal carcinomas [5, 21].

SCTs are classified into four types based on the location of internal and external tumors, as defined by the AAPSS. Type I involves an externally visible mass; Type II is characterized by an external mass with a significant intrapelvic component; Type III includes both external and pelvic masses; and Type IV is entirely internal [17]. Prenatal ultrasound (U/S) can effectively identify Types I and II, while Type IV poses a higher risk of malignancy due to its internal location. Routine U/S is typically performed during the second trimester for antenatal diagnosis, though detection is possible as early as the first trimester. In accordance with this data, more than 60 percent of cases in this systematic review are diagnosed prenatally. However, many pregnant women skip first-trimester U/S screenings, making early detection of masses more challenging. In some cases, the mass is large enough to be detected earlier [1].

Magnetic resonance (MR) imaging complements ultrasound by providing a more detailed characterization of the mass's tissue components [16]. It is particularly useful for evaluating the full extent of large lesions. MR imaging can accurately determine the tumor's reach and the pressure it exerts on surrounding organs. It also aids in distinguishing this condition from common differential diagnoses, such as distal neural tube defects, including myelocystocele or myelomeningocele [5].

Table 4. The effects of tumor size and weight on complication, recurrence, and survival.

Tumor characteristics	Complication		P-value	Recurrence		P-value	Survival		P-value
	Yes	No		Yes	No		Yes	No	
Tumor size (mean \pm SD)	15.02 ± 3.49	14.29 ± 3.87	0.6	17.16 ± 7.41	13.42 ± 2.66	0.14	14.6 ± 4.84	19.25 ± 5.65	0.41
Tumor weight (mean \pm SD)	3.03 ± 0.88	3.47 ± 1.09	0.39	3.40 ± 1.08	3.31 ± 1.04	0.92	3.25 ± 0.91	2.34 ± 0.80	0.35

SD; standard deviation

Additionally, elevated levels of tumor markers, such as alpha-fetoprotein (AFP), are used to assess the likelihood of malignancy. High AFP levels may indicate a malignant tumor; however, in newborns, AFP levels are naturally elevated and typically return to normal by around nine months of age, leading to potential misdiagnosis of the condition as malignant [1].

Perinatal morbidity and mortality rates are notably elevated in fetuses with SCT, primarily due to the high frequency of preterm birth. The most severe perinatal complications include premature labor, malignant tumor infiltration, hemorrhage or rupture of the tumor, amniotic fluid obstruction, and heart failure [5]. Factors that increase the risk of complications include rapid tumor growth exceeding 150 cm³ per week, tumor size greater than 10 cm, highly vascularized solid tumors, polyhydramnios, and cardiac decompensation [4, 21].

Cesarean delivery is recommended for mothers carrying fetuses with SCTs larger than 10 cm in diameter, especially when the tumors are highly vascularized or exceed 5 cm. This approach minimizes the risk of tumor rupture and hemorrhage [2]. This systematic review indicates that cesarean section is performed in more than two-thirds of such cases. The incision is typically made in the lower uterine segment, as the uterus often becomes significantly enlarged due to the tumor's size [21].

Timely surgical intervention for SCTs is crucial, particularly when the tumor exceeds 10 cm, as delays can increase the risk of malignancy and recurrence. Surgery is the primary treatment, with over 93% of cases requiring surgical management. Current expert recommendations indicate that coccygectomy is the most effective approach for preventing the recurrence of benign teratomas, with minimal risk to the tumor cyst wall [2,4]. Ideally, this procedure should be performed within one week and not delayed beyond that period. During surgery, careful fetal monitoring is essential to prevent operative mortality, often caused by severe hemorrhage or cardiac arrest, frequently linked to hyperkalemia [4,10]. Additionally, ensuring the availability of cross-matched blood in the operating room is critical [4].

When the coccyx is not removed, the likelihood of cancer recurrence can be as high as 37%. Even when patients have the entire coccyx removed, there is still a chance of the cancer returning, ranging from 11% to 22% [2]. This aligns with the 18% recurrence rate mentioned in this review. Although recurrence is uncommon, several factors could contribute to it, such as incomplete removal of the tumor, failure to completely remove the coccyx along with the tumor, tumor spillage or rupture, and undetected malignant components within the tumor [12].

Although survival outcomes for SCTs are generally positive, the mortality rate for tumors larger than 10 cm is reported to be around 18%, which is nearly similar to the 15% rate mentioned in this review. SCTs can potentially recur years after treatment, highlighting the importance of continuous monitoring into adulthood. Regular follow-up appointments are recommended every 3 to 6 months, including physical examinations, such as rectal exams, diagnostic imaging, and alpha-fetoprotein (AFP) testing, for at least three years [1,16].

5. Conclusion

Routine ultrasound and magnetic resonance imaging play critical roles in the antenatal diagnosis and characterization of SCTs. Given the elevated perinatal morbidity and mortality associated with SCTs, especially due to preterm birth, cesarean delivery is advised for tumors larger than 10 cm to reduce risks of rupture and bleeding. Furthermore, coccygectomy is highlighted as the most effective strategy for preventing the recurrence of benign teratomas, underscoring the importance of timely and thorough intervention in managing these cases.

Declarations

Conflicts of interest: The author(s) have no conflicts of interest to disclose.

Ethical approval: Not applicable, as systematic reviews do not require ethical approval.

Patient consent (participation and publication): Not applicable.

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Use of AI: AI was not used in the drafting of the manuscript, the production of graphical elements, or the collection and analysis of data.

Data availability statement: Note applicable.

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