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ORIGINAL ARTICLE

Radiation burden in patients with esophageal atresia: a systematic review

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Abstract
Esophageal atresia (EA) is the most common congenital esophageal disorder. Radiological imaging facilitates diagnosis, surgical interventions, and follow-up. Despite this, standardized monitoring guidelines are lacking. We aimed to: (1) review the literature regarding radiation burden in children with EA; (2) establish the presence of guidelines for diagnosis and follow-up in children with EA. The systematic review was performed according to PRISMA protocol. Two investigators conducted independent searches (PubMed, Ovid, Cochrane Review) and data extraction. Analysis focused on pre- and post-operative imaging type and frequency to determine the radiation burden. Seven studies met the inclusion criteria (337 patients). All authors agreed upon the need to minimize radiation burden, recommending symptoms-guided management, use of dosimeters, and non-radiating imaging. One study identified a median 130-fold increase in cumulative lifetime cancer risk in children with EA compared with other babies in the special care unit. The most common investigations were X-ray and CT (pre-operatively), and X-ray and contrast swallow (post-operatively). Standardized guidelines focused upon the frequency and type of radiological imaging for children with EA are lacking. Children with EA are subjected to more radiation exposure than the general population. Implementation of non-radiating imaging (ultrasonography, manometry) is recommended.

Keywords Esophageal atresia · Radiation · Exposure · Outcomes · Systematic review

Introduction

Esophageal atresia (EA) is a congenital disorder that affects 1 in 2600 live births [1]. The majority of children with EA will have an associated tracheo-esophageal fistula (TEF). The gross classification of EA sub-types is based upon the esophageal anatomy, as well the relationship between the esophagus and TEF, if present [2]. The length between the two esophageal pouches determines the classification as either short- or long-gap EA. Whilst operative repair with primary anastomosis typically occurs in the first days of life for patients with short-gap EA, patients with long-gap EA typically undergo delayed repair [3]. This aspect of care for patients with long-gap EA leads to increased numbers of investigations when compared with patients with short-gap EA. Patients with EA may also have associated conditions, including the VACTERL association (vertebral, anal, cardiac, tracheo-esophageal, renal and/or limb malformations) [4].

The imaging modalities utilized in EA include chest X-ray (CXR), upper gastrointestinal contrast swallow,

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intra-operative fluoroscopy, and, rarely, computed tomography (CT) imaging [5]. The long-term effects observed in EA patients of undergoing repeated radiological tests have not yet been established. Whilst imaging studies are important, and often critical, in the management of these patients, there are concerns regarding unnecessary exposure to radiation. Yousef et al. [5] highlighted the need for a more thorough understanding of the effects of radiation in pediatric patients. They identified higher radiation-associated risks in the pediatric population, compared with adults, potentially due to a child's increased rate of growth and, hence, cellular divisions. The principle of as low as reasonably achievable (ALARA) radiation has become the standard of care in medical imaging. As the radiation-induced risk varies according to the patient's age and gender, as well as the modality used and the organs being imaged, the relevance to EA patients is yet unknown [5, 6].

This systematic review has focused upon the effects of radiological imaging in children born with EA, as well as the standardized imaging regimens currently in the literature. We aimed to review the literature regarding the radiation burden for children born with EA, and to determine the presence of current imaging guidelines aimed at the diagnosis and follow-up of children born with EA.

Methods

Protocol and registration

The systematic review was registered with PROSPERO (Reference Number: CRD42018100485). The review was conducted according to the PRISMA protocol [7].

Eligibility criteria

Inclusion criteria included:

(1) Any publications analyzing the relationship between radiation exposure and outcomes in children born with EA.
(2) Any publications discussing the effect of any radiological imaging modalities on health outcomes in children born with EA.
(3) EA patients less than 18 years of age.
(4) Publications between 1998 and 2020.

Exclusion criteria included:

(1) Publications in a language other than English.

Information sources

Databases that were searched included Ovid, PubMed and Cochrane Review. None of the publication's authors were contacted.

Search

The searches, using the following terms, were independently performed by two authors (AC, STT) with American and English spelling: "esophageal atresia/oesophageal atresia" and "radiation". MeSH terms included were: "esophageal atresia/oesophageal atresia", "absorption, radiation", "dose-response relationship, radiation", "neoplasms, radiation-induced", "radiation dosage", "radiation dosimeters", "radiation, ionizing/ionising", "radiation protection", "abnormalities, radiation-induced", "radiation effects", and, "radiation, non-ionizing/non-ionising".

Study selection

Data extraction was independently performed by two authors (AC, STT) for the following: type of study, number of patients, type of studied radiological imaging, overall assessment of utility or harm of the studied radiological imaging, patient demographic and quantitative data regarding type and frequency of imaging, lifetime cancer risk, and, complication development in children born with EA. If there was discrepancy in the data extraction, joint discussion was undertaken, with assessment by a third reviewer (SKK) if necessary, to achieve concordance.

Data collection process

Discrepancies in the selection of included articles were discussed and agreed upon. Similar outcomes were compared amongst the publications to draw conclusions, such as type of imaging used pre- and post-operatively, cancer life-risk, and complication rates. Both favorable and unfavorable data regarding the use of radiation imaging for children with EA were recorded. Overall assessment of utility or harm of the studied radiological imaging was reported, as well as patient demographic data.

Data items

Authors reviewed the modality and frequency of radiological tests performed pre- and post-operatively, discrepancies in EA-gap estimate according to investigation type, lifetime cancer risk, and rates of complications. Lifetime cancer risk and development of complications were chosen as outcomes of interest in terms of quantitative long-term measurement, to be able to discuss benefit versus risk of diagnostic and follow-up imaging.

Synthesis of results

The primary aim of the systematic review was to analyze the radiation burden in children with EA. The secondary aim was to determine the presence of current imaging guidelines used to diagnose and follow-up children born with EA. Similar outcome values in each selected paper were compared. For each selected paper, the authors compared the types and frequency of performed imaging, as well as its mentioned radiation burden, to identify which investigations were most frequently used. We focused upon which guidelines and reasoning were used to determine the imaging modality requested and the radiation burden that each of those investigations would carry.

Risk of bias in individual studies and across studies

The Cochrane risk-of-bias tool was used to assess the presence of bias in included publication, as well as in this systematic review [8]. The ROBINS-I tool was not used to assess bias in this systematic review, as no non-randomized control trials were included in this review [9].

Levels of evidence

The Oxford Centre for Evidence-Based Medicine 2011 Levels of Evidence was used to assess all the included studies [10].

Results

Study selection

A total of 36 articles (PubMed—23, Ovid—4, Cochrane—1, reference lists—8) were initially identified (Fig. 1). Seven articles satisfied the inclusion criteria for the quantitative analysis, with a total of 337 patients [5, 11–16] (Table 1). Of the included articles, three were cross-sectional studies, three were reviews, and one was a case report. Two articles were prospective studies, whilst the remaining five articles reported retrospective data. Articles that did not provide data regarding quantity of radiation burden were excluded.

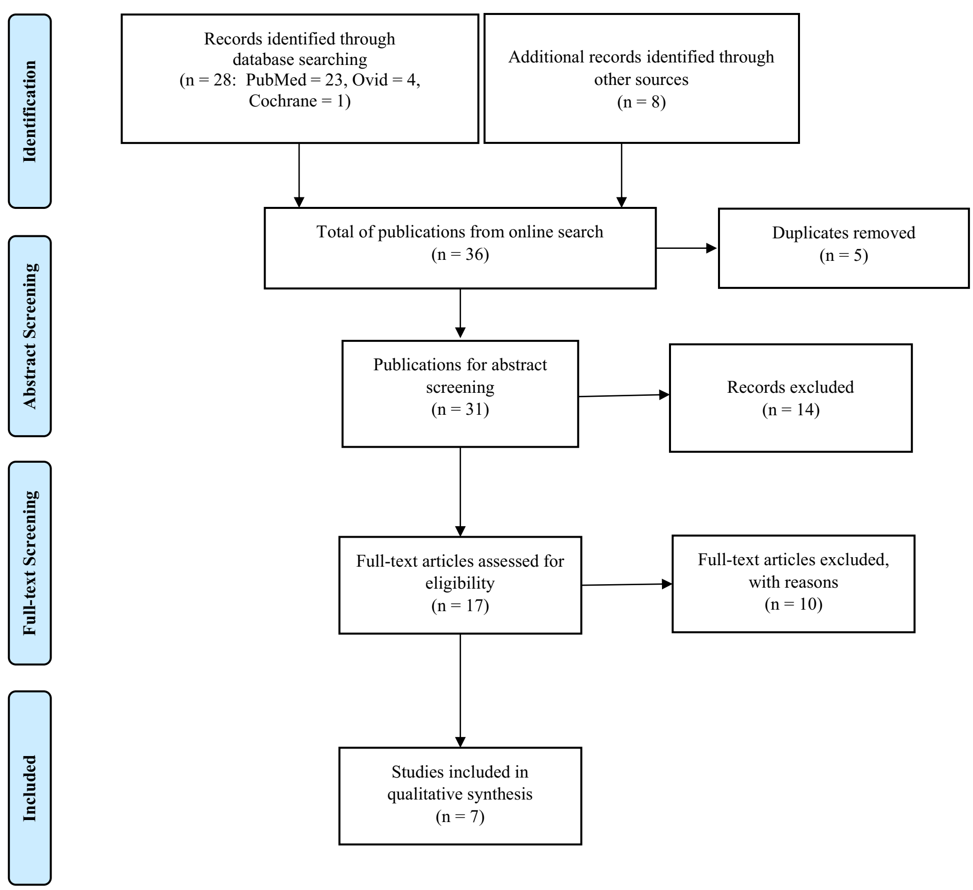

Fig. 1 PRISMA flow-diagram of systematic review methodology

Table 1 Characteristics of selected publications

Author	Year	*N*	Imaging modalities	Radiation exposure–outcome relationship
Roberts et al. [11]	2019	49	X-rays, fluoroscopy, esophagram, CT, nuclear medicine	(1) Fluoroscopy and esophagrams contribute the most to radiation exposure level
				(2) Routine post-operative esophagrams prior to starting feeds are unnecessary if there is no clinical indication for investigations
				(3) Esophagrams findings in asymptomatic patients do not alter management
Yousef et al. [5]	2018	53	All X-rays	(1) Need for minimization of radiation exposure
				(2) The use of dosimetry could help towards this goal
Higano et al. [12]	2017	3	MRI versus X-ray and CT	(1) Unacceptable high exposure to radiation with X-ray use
				(2) MRI considered superior, providing more accurate and descriptive pre-operative information to plan the procedure and to counsel parents appropriately
				(3) MRI considered to be an excellent pre-operative and post-operative tool to monitor EA children for complications
Zamiara et al. [13]	2015	69	All X-rays	(1) Importance to track patients' radiation exposure and hospital visits in order to optimize patients' care
				(2) Need for use of updated and advanced imaging machines
Garge et al. [14]	2013	96	Pre-operative CT	Controversial: safety concerns outweigh its efficacy
Mahalik et al. [15]	2012	30	Pre-operative 3D CT scan	CT radiation increases the lifetime cancer risk in EA children when added to the natural background risk. Its pre-operative use risks outweigh its benefits
Nambirajan et al. [16]	1998	37	Post-operative esophagram	(1) Esophagrams are not required routinely post-operatively
				(2) Management for asymptomatic patients was not altered by esophagram imaging findings

Study characteristics

The demographic data for the patients studied in the seven selected publications were: (Table 2).

(1) EA types: type A—10, type B—6, type C—261, type D—8, type E—13, unknown—39
(2) Gender: males—83, females—68, unknown—186
(3) Gestational age at birth: term—159, preterm—45, unknown—133

Results of individual studies

Roberts et al. [11] analyzed radiation exposure data for a cohort of 49 children with EA over their first year of life. They found that every child had undergone a median of 19 ionizing radiation imaging studies (median radiation exposure of 4.7 mSv), with the majority being X-rays (median of 14 per child, with median radiation exposure of 0.6 mSv), followed by fluoroscopy (median of four per child, with median radiation exposure of 3.3 mSv), and esophagrams (median of two per child, with median radiation exposure of 2.1 mSv). Yousef et al. [5] studied 52 patients, with a focus upon the use of X-rays. They recommended minimizing the use of imaging, and advanced the idea of using dosimeters. Similarly, Zamiara et al. studied 69 patients and highlighted the importance of tracking patient's hospital visits and X-rays performed, in order to minimize the burden [13]. They found a 3-year mean ionizing radiation exposure of 17.4 mSv (median 14.8, range 3.0–59.9 mSv), from a mean of 40 (median 24, range 5–165) radiological studies [13]. Higano et al. [12] studied three patients and evaluated the use of MRI, X-ray and CT. They found MRI to be an excellent pre- and post-operative tool, limiting the radiation burden. Garge et al. [14] studied the use of pre-operative CT in 96 patients. They determined that the burden of radiation outweighed the benefits. Similar results were reported by Mahalik et al., who studied the use of pre-operative 3D CT scanning in 30 patients [15]. They reported a mean exposure of 1.79 mSv in children with EA who underwent pre-operative 3D CT, doubling when radiation levels were combined with natural background radiation [15]. Lastly, Nambirajan et al. looked at the use of post-operative esophagram in 37 patients, and recommended against this investigation as a routine follow-up study [16].

Pre-operative investigations

Garge et al. [14] demonstrated that a pre-operative CT was not required for pre-operative planning, deeming it

Table 2 Demographics of included patients

Author	Esophageal atresia type	Gender	Gestational age	Birth weight (mean ± SD)
Roberts et al. [11]	A—5/49 B—3/49 C—39/49 D—1/49 E—1/49	Male—27/49 Female—22/49	Term—35/49 Preterm (< 37/40)—14/49 Mean—37 ± 3Weeks	2.572 kg (± 0.680 kg)
Yousef et al. [5]	A—2/53 B—1/53 C—41/53 D—5/53 E—2/53 Unknown—2/53	Unknown	Term—42/53 Preterm (< 37/40)—11/53	2.878 kg (± 0.629 kg)
Higano et al. [12]	C—3/3	Male—2/3 Female—1/3	Term—3/3	2.743 kg
Zamiara et al. [13]	A—3/69 B—2/69 C—55/69 D—2/69 E—7/69	Male—32/69 Female—37/69	Term—49/69 Preterm—20/69	Unknown
Garge et al. [14]	C—93/96 E—3/96	Unknown	Unknown	Unknown
Mahalik et al. [15]	C—30/30	Male—22/30 Female—8/30	Term—30/30	2.41 kg (± 0.35 kg)
Nambiraja et al. [16]	EA with TEF—34/37 Pure OA—3/37	Unknown	Unknown	Unknown
Total	A—10/337 B—6/337 C—261/337 D—8/337 E—13/337 Unknown—39/337	Male—83/151 Female—68/151	Term—159/204 Preterm—45/204	2.651 kg

more of a long-term safety concern rather than an effective short-term tool. Though CT may provide useful information regarding lung and heart anatomy, anomalies may be diagnosed with less deleterious means, including echocardiogram and CXR. Berrocal et al. [17] supported this view, though acknowledging that the level of anatomical detail and accuracy using CT was far superior to CXR. Mahalik et al. [15] studied type C EA children and reported a gap length difference of 0.43 ± 1.18 cm between CT and intra-operative findings, whilst Garge et al. found that the chosen surgical approach was unchanged by CT findings [14]. Berrocal et al. [17] deemed that CT and MRI were not useful diagnostic modalities for EA, despite providing greater anatomical detail, and described an increased use of ultrasound as a diagnostic tool for EA. Higano et al. [12] deemed pre-operative 3D MRI to provide a great amount of detail prior to the repair, which was superior to the detail provided by CXR [12]. They also suggested that the level of detail that MRI gave could not only be useful to plan the surgical procedure and evaluate post-operative complications, but also enable superior pre-operative counseling for the families [12].

An ongoing issue in the management of EA patients is the accurate estimation of esophageal gap length. Mahalik et al. [15] explored the different imaging modalities available and found that CXR was unable to provide adequate anatomical detail. Contrast studies, endoscopy and bronchoscopy were mainly useful to provide information regarding the TEF. Whilst CT provided detail regarding the TEF, the esophageal gap, and potential associated mediastinal anomalies, there were concerns regarding the potential long-term radiation risks. Mahalik et al. [15] foresaw that, with the advent of MRI and the improvement in quality and availability, there would be a shift towards this imaging technique in children with EA/TEF.

Post-operative investigations

Nambirajan et al. [16] reported that post-operative anastomotic narrowing on CXR is not related to symptomatic stricture formation. The association between anastomotic leak and development of a symptomatic stricture is controversial. They concluded that CXR and esophagrams were actually not required and predisposed children to complications later in life. All the anastomotic leaks that were identified radiologically did not manifest clinically. No relation was found between the anastomotic narrowing and the development of symptomatic strictures. These findings were supported by Zamiara et al. and Roberts et al. who stated that post-repair esophagram to commence oral feeds, followed by another esophagram at one year of age, was unnecessary [11, 13]. They advocated that significant anastomotic leaks present clinically, with sufficient indications to alter the management accordingly, and leaks found only on radiological imaging were minor and tended to respond to conservative treatment.

Burden of radiation exposure

Yousef et al. [5] estimated lifetime radiation-induced cancer risk in children with EA, basing their calculations on normative prenatal exposure data from the International Commission on Radiological Protection 2007 recommendations (1:200,000). They estimated a median radiation dose exposure of 5.5214 mSv/patient, with an estimated median cumulative lifetime cancer mortality risk of 1:1530 [5]. Of note, they reported an estimated maximum radiation dose exposure of 6.66386 mSv/patient, which carried an estimated median cumulative lifetime cancer mortality risk of 1:1575 and a maximum risk of 1:130. Mahalik et al. [15] demonstrated a doubling of radiation exposure levels in children with EA, compared with the general population, when natural predisposition exposure level was combined with the radiological imaging one. Similarly, Zamiara et al. identified a radiation exposure for this cohort of patients, which was equivalent to 226 flights from Buenos Aires to Sydney [13]. Radiation risk was higher in younger patients, with a fourfold increase in children compared with adults, and higher in females [18]. Berrocal et al. [17] demonstrated a three–fourfold increase in lifetime risk with exposure in the first 10 years of life, compared with radiation exposure at 30–40 years of age. Long-term harm risks were higher with exposure during childhood, compared with adulthood.

Limiting radiation exposure

In the first three years of life, Zamiara et al. identified a radiation exposure level comparable to 6–7 years of background radiation [13]. The authors identified a wide range of cumulative exposure, due to the different fluoroscopy machines used [13]. The more advanced the machines, the lower the radiation exposure, and therefore, the lower the cumulative exposure. They demonstrated a reduction in radiation exposure of a factor of 3 with the use of more advanced technology.

Synthesis of results

Chest X-ray, CT, and rarely MRI were described as pre-operative investigation modalities [5, 12]. All were used to estimate the EA-gap length, with CT and MRI providing greater anatomical detail when compared with X-ray. However, discrepancies were still noted during surgery. Post-operatively, upper gastrointestinal contrast swallow and CXR were identified as the predominant follow-up studies, even though no specific guidelines regarding timelines were provided. The majority of post-operative complications were anastomotic leakage and stricture formation, with median rates estimated to be as high as 19% and 29%, respectively, in some studies, warranting close clinical monitoring and at times consideration for a lower threshold for investigations [19]. Both of these complications led to increased inpatient admission length of stay and subsequent radiation exposure [5].

Levels of evidence

The included studies were assessed to be Level 3 evidence according to The Oxford Centre for Evidence-Based Medicine 2011 Levels of Evidence [10].

Discussion

Esophageal atresia is the most common congenital malformation of the esophagus. Modern management is focused upon ensuring a quality of life (QoL) as normal as possible for surviving patients, with minimization of morbidity and mortality. It is known that children with EA have a higher radiation burden than the general population, with reported higher median radiation dosages [11, 15]. This review found that there is a paucity of guidelines regarding appropriate investigations for patients with EA. In addition, there is a paucity of evidence regarding long-term risks of radiation exposure. Chest X-rays and CT scans were the investigations of choice pre-operatively, whilst upper gastrointestinal contrast swallows and CXR were used post-operatively, as per clinician discretion. A risk–benefit analysis is crucial at the time of every imaging, considering the patient's short- and long-term best level of care, as well as financial costs. The greater the number of ordered tests, the higher the costs, with CT and MRI scans being more expensive than CXR [14]. It has been suggested to increase the use of non-radiating imaging modalities pre-operatively, and to follow-up children post-operatively based on symptomatology [16, 17]. Post-operative routine investigation of asymptomatic patients with radiation imaging is discouraged [11].

Yousef et al.'s [5] findings, in the cohort of children born with EA, correlated with a median of 3 years and a maximum of 42 years of neonatal background radiation exposure. However, it has to be acknowledged that this study based its risk of radiation-induced cancer calculation on prenatal normative data; hence, this becomes a limitation when conclusions are drawn for post-natal radiation exposure. Mahalik et al. and Zamiara et al.'s results also demonstrated a higher burden of radiation in children with EA [13, 15]. Exposure to radiation during medical imaging carries the potential for harm in the long term. Balonov et al. [18] described the different factors that affected the distribution of radiation dose: type of machine used, exposed area, technique used, patient's stature, and radiographer's expertise [5]. Overall, despite a higher exposure radiation dose having been identified, there are no published guidelines to minimize imaging in children with EA.

In order to limit radiation dose, Yakoumakis et al. emphasized the importance of experienced personnel performing the test, exposing the minimum portion of the body to radiation [20]. The use of modern equipment is also pivotal [5, 13, 17]. Zamiara et al. [13] found that the increased radiation exposure in the first few years of life for children with EA was mainly caused by routine investigations, rather than clinically determined investigations, with some patients undergoing post-operative esophagrams prior to commencing oral feeds, with a follow-up study at one year of age. Diagnostic fluoroscopy was identified as one of the main causes of elevated radiation exposure [11]. Cumulative radiation exposure becomes, therefore, relevant, hence the importance of using updated radiological equipment to minimize radiation-induced long-term harm [13].

Similarly, Berrocal et al. discussed long-term complications and the relevance of the appropriate size of the imaged body part [17]. They highlighted the importance of proper radiological technique and equipment, yielding higher-quality images and reducing unnecessary peripheral exposure. Justification and optimization of the imaging procedure are crucial principles that the International Commission on Radiological Protection has recommended [21]. Clinical relevance of the test has to be explicit and crucial for the management of patients. Additionally, the information should not be able to be obtained in any other "less potentially harmful" way [17].

Pre-operative CT has been deemed not necessary for operative planning, and alternative imaging modalities (echocardiograms, CXR) should be used. By doing so, some level of anatomical detail that CT provides is lost, but the radiation exposure level is greatly lowered [14, 15, 17]. The use of pre-operative MRI was also investigated: this provides high levels of anatomical detail, as well as reductions in radiation exposure, but this imaging modality has accessibility limitations [12].

Yousef et al. [5] have advocated for early post-operative surveillance with imaging modalities that do not involve radiation, including USS. They described their use of dosimeters for each patient, to quantitatively monitor radiation exposure. According to current clinical practice, a post-operative esophagram is commonly performed in the first week following EA repair, regardless of the findings on CXR. It has been proposed that post-repair esophagram should be discouraged, as it does not provide any clinically significant detail for the patient's management, in particular for asymptomatic patients [11, 16]. Isolated radiological findings of anastomotic leaks which do not correlate with clinical symptoms are often managed conservatively, hence the recommendation to not expose children with EA to regular investigations when there is no clinical indication that warrants them [11]. Considering the higher aspiration risk that children with EA have and the current clinical practice, Nambirajan et al. and Zamiara et al.'s suggestion of a change in practice highlights the need for further research in this area. Studies making an evidence-based risk–benefit comparison of undergoing esophagrams or not, accounting for short- and long-term consequences, are needed [13, 16].

Considering the increasing evidence of minimal short-term benefits in using imaging to investigate asymptomatic children with EA, paired with minimal or no change in management for this cohort of patients, it may be beneficial to use the patients' clinical presentation to guide decision-making. Symptomatic patients should be followed up with investigations, performing a short- and long-term risk–benefit assessment of every procedure. The use of non-radiating imaging modalities, such as esophageal manometry, USS, and potentially MRI, should be considered as alternative investigation modalities, aiming to limit the level of radiation exposure.

A limitation of the current systematic review is that it identified only seven relevant articles, demonstrating the paucity of relevant data. Quantitative analysis was, therefore, not possible. No selection, performance, detection, nor attrition bias was found in any of the selected publications, according to the Cochrane risk-of-bias tool. All studies reported limitations that authors experienced during their data collection and analysis. The main difficulty was the accurate calculation of radiation exposure values, as several confounders were present. Some of these included radiation burden due to comorbidities or prenatal radiation exposure when interpreting radiation levels for post-natal radiation levels only. The publications accounted for this in their statistical analysis or, if this was not possible, they mentioned it as part of their limitations. The authors of this systematic review acknowledged these limitations mentioned in the selected studies and reported them in their analysis and conclusions. The types of studies included in the systematic review were three cross-sectional studies, three reviews, and one was a case report. Randomization and blinding were, therefore, not possible in any of these studies. Two articles were prospective studies, whilst the remaining five articles reported retrospective data. No selective reporting or attrition bias was found in any of the papers.

Conclusions

In conclusion, there are no current guidelines that clearly state the appropriate investigations for children born with esophageal atresia, nor are there any guidelines for a recommended maximum limit of radiation imaging. The use of radiation dosimeters could be an important adjuvant tool to aid clinicians when considering indications and need for further investigations. This systematic review found that pre-operative chest X-ray, with the use of non-radiation imaging modalities (e.g., ultrasound scans), seem to be as safe and useful as pre-operative computed tomography scans, whilst significantly reducing radiation exposure. Therefore, we discourage the use of pre-operative computed tomography scans.

This systematic review also identified a lack of evidence-based knowledge regarding the indications for esophagrams prior to commencing oral feeds. Only three publications reported evidence against the use of these investigations, as most asymptomatic strictures and leaks do not require any interventions. Manometry could be a useful tool that could be utilized to assess oesophageal motility and function post-operatively, minimizing the radiation burden. Utilizing up-to-date imaging machines with paediatric radiation doses are also crucial.

Acknowledging that survival rates have largely increased for children with esophageal atresia, further research should be aimed at direct, prospective radiation exposure measurement. Studies could involve prospective long-term follow-up studies to quantitatively measure the radiation doses from birth relating to esophageal atresia-related investigations, and reporting rates of any radiation-induced complications in the subsequent years. Other studies could prospectively investigate fluoroscopy studies prior to commencing oral feeds to determine its clinical relevance, as the current literature only reports controversial and a small amount of data on the current topic.

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Declarations

Conflict of interest The authors have no relevant financial or non-financial interests to disclose.

Ethical approval This study was performed in line with the principles of the Declaration of Helsinki.

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