

Radiographic changes in ribs following clipping of patent ductus arteriosus in preterm infants

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Abstract

Objectives: There are no published reports on the rib abnormalities on the plain chest radiograph in preterm infants following surgical clipping of isolated patent ductus arteriosus. The purpose of this study was to describe changes in the ribs on the plain chest radiograph following surgical clipping of patent ductus arteriosus (surgery groups) in preterm infants compared to non-surgical closure of patent ductus arteriosus (control group).

Methods: Data from preterm infants with patent ductus arteriosus clipping (surgery) via a left postero-lateral thoracotomy were compared with infants in whom the patent ductus arteriosus closed: spontaneously, with medications or use of an occlusive device (controls). Serial pre- and post-closure plain chest radiographs were randomly reviewed by a reader blinded to the route of closure and up to 1 year following the patent ductus arteriosus closure.

Results: Of the total of 196 cases included in the study: 45 of the patent ductus arteriosus closed following treatment with medications, 8 cases closed with an occlusion device, 38 were closed surgically, and in 105 cases, the patent ductus arteriosus closed spontaneously. Compared to the pre-operative period, 36/38 (95%) infants in the surgery group had one or more of the following rib abnormalities: ipsilateral fourth and fifth rib fusion, narrowing of the ipsilateral fifth intercostal space, thinning of the ipsilateral fourth or fifth rib, or a combination of the above on the chest radiograph compared to 0% in the control group ($p < 0.001$).

Conclusion: Radiographic rib abnormalities are common and appear in infancy following surgical clipping of patent ductus arteriosus in preterm infants. Further studies are needed to clarify the natural history of these abnormalities on thoracic cage and cardiopulmonary functions.

Keywords

Child, heart, patent ductus arteriosus, surgery, radiography

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Introduction

Abnormalities of ribs identified on a plain chest radiography in infants can indicate a systemic or metabolic disease or trauma, including non-accidental trauma.^{1,2}

Patent ductus arteriosus (PDA) is the persistence of the fetal ductus arteriosus between the aorta and pulmonary artery beyond the immediate postnatal period.³ The pathophysiological effects of isolated PDA is dependent on the gestational age of the patient and the size of the PDA.⁴ A hemodynamically significant PDA leads to excess pulmonary blood flow with volume overload on the left side of the heart with resultant heart failure and pulmonary edema. These PDAs require interventions for closure.^{3,4}

Pharmacological therapy using non-steroidal anti-inflammatory drugs (NSAIDs) or acetaminophen may facilitate closure of PDA.⁵ Percutaneous occlusion devices are being

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increasingly utilized in patients who fail to respond to medical therapy.^{6,7} Other patients undergo surgical closure of PDA via a left postero-lateral thoracotomy or thoracoscopy using either ligation of the PDA or more commonly application of a titanium clip (clipping) over the PDA in order to close it.⁴ Reported complications of surgical closure of PDA include vagus nerve and recurrent laryngeal nerve palsies and transient worsening of the respiratory status.^{4,5}

Abnormalities of the ribs on the plain chest radiograph in preterm infants may appear similar to congenital or acquired medical conditions as well as trauma including non-accidental trauma. There are no published reports on the chest wall bony abnormalities in preterm infants, *during infancy*, following PDA clipping.

The purpose of this study was to evaluate rib abnormalities on chest radiography during infancy in two cohorts of preterm infants: one who have undergone PDA clipping via postero-lateral thoracotomy and the second in which the PDA closed non-surgically. We hypothesized that PDA clipping is more likely to be associated with rib abnormalities post-operatively during infancy compared to PDA closure non-surgically.

Methods

This was a retrospective chart review study. Preterm infants with a diagnosis PDA confirmed by echocardiography between 1 January 2012 and 31 December 2018 were included. Infants who had other cardiac defects or had undergone other thoracotomies were excluded. Institutional Review Board approval was obtained prior to data collection. All surgeries were performed via a left postero-lateral thoracotomy with appropriate dissections to identify and separate the PDA prior to application of a titanium clip.

The following data were collected: age, sex, gestational age and weight at the time of diagnosis, manner of closure, age at the time of closure of PDA, findings on plain chest radiograph, the need for invasive mechanical ventilation at the time of closure of PDA, and mortality. Mortality directly related to surgery was defined as death within 7 days following surgery.

Serial chest radiographs obtained for daily management of the patient by the treating physician were reviewed prior to closure and up to 1 year following closure of PDA (surgical or otherwise) by one of the authors (R.A.H.) who was blinded to the method of closure by applying a radiopaque material over the PDA clip that would otherwise be visible on the chest radiograph. The reader (R.A.H.) was trained by a renowned pediatric radiologist on interpreting plain chest radiographs in infants and has been interpreting plain chest radiographs in infants for daily clinical practice for 31 years.

Statistical analysis

Infants were classified into two groups: surgery group (surgery) and those who did not undergo postero-lateral thoracotomy

(controls). *t*-test was used to compare continuous variables; Wilcoxon's rank-sum and chi-square analysis was used for categorical variables. Relative risk with 95% confidence interval (CI) was calculated between both arms of the cohort. Variables with a *p*-value < 0.05 were identified as statistically significant independent risk factors associated with the development of rib abnormalities on the chest radiograph. Regression analyses were implemented to assess if the observed outcome was predicted by the treatment or control group. All analyses were performed using SPSS software program (IBM SPSS®, IL, USA).

Results

A total of 196 preterm infants with PDA with a mean gestational age of 27.5 ± 2 weeks were included in the study. Of these, 38 infants (38/196, 19.3%) underwent surgical clipping of the PDA (surgery group), while 158 (158/196, 80.7%) are considered controls (Table 1). Of the 158 infants in the control, 45 closed with use of medications, 8 cases were closed with an occlusion device, and in 105 infants, the PDA is closed spontaneously.

The surgery group had a lower birth gestational age (25.7 ± 1 vs 29.3 ± 1 weeks, $p < 0.05$) and a lower age at the time of the diagnosis of PDA (26 ± 0.5 vs 29 ± 0.8 weeks, $p < 0.05$) compared to controls (Table 1). There was no statistically significant difference between the two groups with regard to radiographic bony abnormalities prior to surgery (Table 1).

The vast majority of 36/38 (95%) patients in the surgical group had radiographic abnormalities detectable (at a mean postnatal age of 10 ± 2 weeks) compared to 0% in the control group ($p < 0.001$) at the same postnatal age (Table 1). The radiographic abnormalities on the ipsilateral side included (1) narrowing of the fifth intercostal space (57%); (2) thinning/narrowing of the fourth and fifth ribs (23%); (3) fusion (Figure 1) of the fourth and fifth ribs (43%); and (4) a combination of all of the above (95%). 38/38 (100%) of the patients in the surgery group were receiving invasive mechanical ventilation on the day of surgery compared to 77% (121/158) in the control group at the time of confirmation of the closure of PDA by echocardiography ($p < 0.05$). The mortality rate was higher (but not directly related to surgery) in the surgery group compared to controls (2/38 (5%) vs 1/158 (2.1%), $p < 0.05$).

Furthermore, since patients included in the surgery group were younger, regression analysis for gestational age was conducted which showed that the mortality rate was no longer predicted by surgery or control group (odds ratio=1.02; 95% CI=0.76–1.24; $p=0.86$). However, rib abnormalities were still strongly predicted by surgery (odds ratio=146; 95% CI=0.06–146; $p=0.000$). There were no cases of phrenic nerve or recurrent laryngeal nerve injury in any of the patients.

Table 1. Comparison of characteristics of patients with patent ductus arteriosus (PDA) who underwent surgery (surgery group) and matched control (control group) patients.

Variable	Surgical group (N=38)	Control group (N=158)	p
Gestational age at birth (weeks)	25.7 ± 1 weeks	29.3 ± 1 weeks	<0.05
Age at PDA diagnosis (weeks)	26 ± 0.5 weeks	29 ± 0.8 weeks	<0.05
Weight at diagnosis of PDA (kg)	0.870 ± 0.160 kg	1.17 ± 0.20 kg	<0.05
Rib abnormalities on chest radiograph prior to closure of PDA	0/38 (0%)	0/158 (0%)	NS
Rib abnormalities following PDA closure, n (%)	36/38 (95%)	0/158 (0%)	<0.05
Mechanical ventilation on the day of closure	38/38 (100%)	121/158 (77%)	<0.05
Earliest time of post-closure radiographic evaluation, weeks	10 ± 2 weeks	10 ± 2 weeks	NS

NS: not significant.

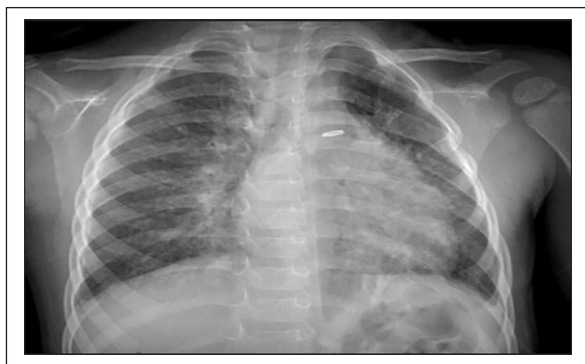


Figure 1. A chest radiograph demonstrating fusion of the fourth and fifth rib with narrowing of the fourth and fifth intercostal spaces on the left side. A patent ductus arteriosus clip is seen as a radiopaque material.

Discussion

This study demonstrates that ipsilateral rib abnormalities on the plain chest radiograph are commonly observed following surgical clipping of PDA in preterm infants. These abnormalities are detected during infancy and persist throughout infancy.

There are no data in preterm infants on the changes in the ribs following PDA closure via postero-lateral thoracotomy during infancy.⁴ The need for insertion of a surgical retractor between the delicate ribs of the preterm infant in order to provide adequate visualization of the ductus arteriosus is likely to lead to some degree of injury to the adjacent soft ribs of the preterm neonate.⁸ Subsequent healing may lead to various abnormalities that we have described in this report, particularly fusion of the ribs.

Jaureguizar et al.⁸ reported on the musculoskeletal sequelae of thoracotomy for tracheoesophageal fistula repair in infants over a period of 16 years. In this study, follow-up chest radiographs, performed at various ages following surgery, demonstrated rib abnormalities in only one-third of the patients. The authors in the study of Jaureguizar et al. also reported fusion of the ribs on the ipsilateral side similar to the current.

Seghaye et al.⁹ reported on thoracic sequelae following surgical closure of PDA in infancy (predominantly with ligation). The median chronological age was 6 years and 1 month at the time of follow-up imaging with plain chest radiographs. There were no radiographic images in infancy, and therefore, it is not clear if these radiographic abnormalities were present during infancy and what was the nature of rib deformities. The current data shed light onto the musculoskeletal complications of surgical clipping of PDA during infancy when rib abnormalities can be confused with other medical conditions including non-accidental trauma.⁸

Rib fractures are rare with accidental injuries in infants and are unlikely to occur during chest compression for cardiopulmonary resuscitation.¹⁰ In the setting of non-accidental trauma, rib fractures are more common at the posterior arc of the rib closer the vertebral bodies.^{1,2} This site is more prone to fracture because the levering force during the physical abuse tends to be directed more posteriorly and the area at which the rib angulates forward receives the brunt of the force leading to fractures in this area.^{1,2} Rib fractures in association with non-accidental trauma tend to involve multiple ribs from both sides, whereas the changes in the ribs following surgical ligation of the PDA tends to involve only the ipsilateral fourth and fifth ribs as demonstrated in this study.¹

Limitations of the study

This study has a number of limitations. First, it is based on retrospective data from a single institution with its own clinical settings; we did not perform a power analysis for calculation of sample size; therefore, the results of this study may not be applicable to other clinical settings.

Conclusion

Rib abnormalities following PDA clipping are common and are detectable on the plain chest radiograph in infancy. Percutaneous occlusion of PDA is likely to replace surgical closure. Therefore, rib abnormalities following PDA closure should decline in the future. However, there are a substantial number of children who have undergone surgical repair, and

these patients need to be recognized and monitored in order to elucidate the natural history of these rib abnormalities and the potential for development of more serious complications such as scoliosis and shoulder height discrepancies.

Authors' note

All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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Author contributions

R.A.H. created the concept, planned the study, revised data collection and the initial draft of the manuscript, and does not have financial conflicts or otherwise. F.K. planned for data acquisition and assisted with data analysis and revision of the manuscript, he has no conflicts to report, financials or otherwise. J.Z.H. wrote the first draft, helped with data collection, and revision of manuscript. He has no conflicts to report. K.H. wrote the first draft, helped with data collection and revision of manuscript.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Ethical approval for this study was obtained from the Institutional Review Board of Hurley Medical Center. IRB approval number is 1332540-2.

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Informed consent

Since this is a retrospective observational study, exemption for obtaining informed consent was granted by the Institutional Review Board of Hurley Medical Center.

Trial registration

This randomized clinical trial was not registered because the study was retrospective, observational, single-centered, therefore, it cannot be registered as randomized trial.

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