Oligohydramnios and Spontaneous Pneumothorax

1. Pneumothorax as a clinical manifestation of autosomal recessive polycystic kidney disease in neonates

Author(s): Skalova S.; Maly J.; Racicka K.; Rejtar P.; Kopriva J.; Stekrova J.; Obeidova L.
Source: Pediatric Nephrology; Sep 2015; vol. 30 (no. 9); p. 1678
Publication Date: Sep 2015
Publication Type(s): Journal: Conference Abstract
Available in full text at Pediatric Nephrology - from Springer Link Journals
Available in full text at Pediatric Nephrology - from ProQuest

Abstract: Introduction: Autosomal recessive polycystic kidney disease (ARPKD) belongs to a group of congenital hepatorenal fibrocystic syndromes. The only gene known to be associated with ARPKD is PKHD1 and its mutations are for ARPKD diagnostic. Clinical manifestation depends on the severity of kidney and liver disease. The most common manifestation of ARPKD is the perinatal form. Cases description: We report 3 neonates with pneumothorax as a clinical manifestation of ARPKD. Patient No.1 was a boy born in term. His antenatal screening tests were negative. He required resuscitation and mechanical ventilation immediately after delivery because of lung hypoplasia and bilateral pneumothorax. Extremely enlarged kidneys were detected on ultrasound and complete anuria was recorded in the clinical course. He expired during the first day of life due to refractory respiratory failure. Patient No. 2 was a girl born in the 37th gestational week. After the birth she had tachydyspnea due to right-sided pneumothorax with improvement of the respiratory symptoms after single puncture. Postnatal ultrasonography revealed enlarged hyperechogenic kidneys. Glomerular filtration rate was normal. She required hypertension treatment and NaCl supplementation since day 3. Patient No. 3 was a boy from the second pregnancy monitored regularly for oligohydramnion since the 30th gestational week. He was born in term. Right-sided pneumothorax was diagnosed shortly after birth with resolution after single puncture. Postnatal ultrasonography revealed enlarged hyperechogenic kidneys. The diagnosis of ARPKD was confirmed clinically and genetically also in his 6-year old brother. Conclusions: Spontaneous pneumothorax as the only respiratory problem was the first manifestation of ARPKD in two of our reported patients. Based on our experience, we recommend postnatal ultrasonographic evaluation of kidneys in all neonates and infants with the history of oligohydramnion or respiratory problems in neonatal period.

Database: EMBASE
2. Oligohydramnios-induced lethal pulmonary hypoplasia secondary to prelabor rupture of membranes

Author(s): Yu M.; Guo C.; Li X.; Lu Z.; Zhuang S.

Source: Journal of the American College of Cardiology; Oct 2016; vol. 68 (no. 16)

Publication Date: Oct 2016

Publication Type(s): Journal: Conference Abstract

Available in full text at Journal of the American College of Cardiology - from ProQuest

Abstract:OBJECTIVES To improve the recognition and management of oligohydramnios-induced lethal pulmonary hypoplasia (LPH) secondary to prelabor rupture of membranes (PROM). METHODS We present a case of oligohydramnios-induced LPH secondary to PROM. Relevant literature was reviewed. RESULTS The male infant was born at 30+3 week's gestation by caesarean. Ultrasonic examination showed the amniotic fluid index (AFI) was 0 cm, the estimated fetal weight (EFW) was 1153 g, and the estimated fetal gestational age (GA) was 28+4 weeks. His mother had amniotic fluid leak for 5 days and twin pregnancy. He developed cyanosis and lethargic after birth. Endotracheal intubation, positive pressure ventilation, chest compression were performed. He didn't show improvement and died at 2 hours after birth. Chest X-rays showed bell-shaped thorax, bilateral pneumothorax and pneumomediastinum. Ultrasonic examination of the other fetal showed the AFI was 63 cm, the EFW was 1443 g, and the estimated fetal GA was 29+6 weeks. The male infant developed cyanosis and lethargic after birth and showed improvement after endotracheal intubation. This case had a history of PROM, anhydramnios and inconsistent development in twins. The clinical and radiologic manifestations were corresponding to LPH diagnosis. According to literature of 144 cases of LPH secondary to midtrimester PROM, GA at PROM, latency period and AFI were predictors of LPH. It was hypothesized that dry lung syndrome might be functional pulmonary hypoplasia. CONCLUSIONS To prevent LPH, PROM management included AFI, fetal biometric indices and pulmonary vessels examination. Neonatal management might include volume-targeted, high-frequency ventilation and nitric oxide.

Database: EMBASE

3. Adverse effects of surfactant therapy in preterm infants with pulmonary hypoplasia due to oligohydramnios

Author(s): Kawase Y.; Hine K.; Saito K.; Mizukaki N.; Hagihara S.; Arai H.; Yoda H.

Source: Journal of Maternal-Fetal and Neonatal Medicine; Jun 2014; vol. 27 ; p. 217

Publication Date: Jun 2014

Publication Type(s): Journal: Conference Abstract

Abstract: Brief Introduction: Oligohydramnios after premature rupture of membranes is known to cause severe respiratory failure in preterm infants. Surfactant is often administered as an early rescue therapy in these severe cases before a definitive diagnosis of respiratory distress syndrome. We reviewed the adverse effects of surfactant therapy in pulmonary hypoplasia due to oligohydramnios. Materials & Methods: A retrospective study of the preterm infants with birth weight<2000g and<34weeks of gestational age who had pulmonary hypoplasia due to oligohydramnios admitted from Jan 1994 to Dec 2013. 34 infants were included. The gestational age, the birth weight, the timing and the duration of rupture of membranes, and the initial treatments and their complications were reviewed in each case. Clinical Cases or Summary Results: The mean gestational age and birth weight were 27.1 +/- 3.1(weeks) and 942 +/- 379(g). 16 of 34 infants were treated with high frequency oscillation. Surfactant was administered in 30 of 34 infants. 22 infants were treated with the inhalation of nitric oxide. 12 infants had pneumothorax and 8 infants had intraventricular hemorrhage and all these cases had received surfactant therapy. 6 infants died within 24 hours after birth and all the cases had received surfactant therapy.
Conclusions: Surfactant therapy was associated with severe complications in preterm infants with pulmonary hypoplasia due to oligohydramnios and may be associated with poor prognosis. In these conditions, it is better to avoid the immediate surfactant therapy after birth.

Database: EMBASE

4. Recurrent severe oligohydramnios and fetal pulmonary hypoplasia associated with ErbB4 mutation.

Author(s): Kamath-Rayne, Beena D; Saal, Howard; Lang, Stephanie; Habli, Mounira

Source: Obstetrics and gynecology; Feb 2013; vol. 121 (no. 2)

Publication Date: Feb 2013

Publication Type(s): Case Reports Journal Article

Available in print at Patricia Bowen Library and Knowledge Service West Middlesex university Hospital - from Obstetrics and Gynecology

Available in full text at Obstetrics and Gynecology - from Ovid

Abstract: BACKGROUND Pulmonary hypoplasia resulting from oligohydramnios or anhydramnios can cause severe respiratory compromise in newborn patients. We report a case of recurrent oligohydramnios in a mother with an ErbB4 mutation and speculate that the effects on the placenta through decreased vascularization contributed to oligohydramnios and subsequent pulmonary hypoplasia in the newborn. CASE The pregnant mother in this case had two subsequent term pregnancies complicated by severe oligohydramnios. Both pregnancies resulted in live born female neonates with pulmonary hypoplasia, pneumothoraces, and pulmonary hypertension. The mother and second newborn, who died, were found to have the ErbB4 mutation. Examination of the placenta with that pregnancy showed decreased vascularity. CONCLUSION ErbB4 may have important effects on placental development and hydramnios that also may affect neonatal pulmonary hypoplasia.

Database: Medline

5. Long-term follow-up of antenatal oligohydramnios of renal origin (ROH)

Author(s): Klaassen I.; Schmidtke S.; Mueller-Wiefel D.E.; Kemper M.J.; Laube G.F.

Source: Pediatric Nephrology; Sep 2010; vol. 25 (no. 9); p. 1974

Publication Date: Sep 2010

Publication Type(s): Journal: Conference Abstract

Available in full text at Pediatric Nephrology - from Springer Link Journals

Available in full text at Pediatric Nephrology - from ProQuest

Abstract: Objectives: Prognosis of ROH has been regarded as unfavorable, although clinical data are scarce. Due to progress in neonatal intensive care and the treatment of infants with chronic kidney disease, the overall prognosis of these children has improved considerably. The aim of this study was the evaluation of complications and long-term follow-up in patients with ROH. Methods: Data on 39 fetuses (26 male, 13 female) with ROH from 1990 to 2010 were evaluated. Results: Primary diseases included urinary tract malformations (n=26), ARPKD and ADPKD (n=9) and other (n=4). Of 9 non-survivors (23%), 6 died within the neonatal period. 27 patients required mechanical ventilation (16 with associated pneumothorax). The surviving 30 children have a current median age of 5.6 (0,116) years. All developed CKD, which could be managed conservatively in 14 patients (median GFR 50.1 (range 19.9-130) ml/min/1.73m2). 16 patients reached ESRD at a median age of 3 months (range 2
days to 8.2 years); two received a preemptive kidney transplantation (KT), and 14 started peritoneal dialysis. 11 of these patients underwent successful KT at a median age of 3.1 (range 1.1-12) years. Two of them had a combined hepatorenal transplantation due to ARPKD. Cognitive and motor development was normal in 25 of 30 patients (83%) and showed a delay in 5 children. Conclusions: In ROH long-term prognosis is encouraging and range from CKD stage 1 to combined hepatorenal transplantation. The rate of perinatal problems and complications is high and may increase, since more severely affected fetuses are actively treated.

Database: EMBASE

6. Lethal cutis laxa with contractural arachnodactyly, overgrowth and soft tissue bleeding due to a novel homozygous fibulin-4 gene mutation.

Author(s): Hoyer, J; Kraus, C; Hammersen, G; Geppert, J-P; Rauch, A
Source: Clinical genetics; Sep 2009; vol. 76 (no. 3); p. 276-281
Publication Date: Sep 2009
Publication Type(s): Case Reports Journal Article

Abstract: Cutis laxa is characterised by redundant, inelastic skin with deep wrinkling and additional variable systemic involvement. Mutations in fibulin-4 (EFEMP2) and fibulin-5 (FBLN5) were described to be causative for autosomal recessive cutis laxa type 1 in a few families each. The female patient was born to healthy consanguineous parents. Pregnancy was remarkable for fetal overgrowth and oligohydramnios. The newborn girl showed extreme bradycardia and died perinatally. Apart from overgrowth, cutis laxa, arachnodactyly of hands and feet with contractures of the third to fifth finger, medial rotation of feet, spina bifida of the os sacrum, microcephaly and facial dysmorphism were noted. Autopsy showed collapsed lungs with hypoplastic diaphragm and signs of cervical soft tissue bleedings due to fragility of vessels. Histologic examination showed fragmentation of elastic fibres with formation of cystic cavities in the medial layer of the aorta and central lung vessels. Sequencing of the elastin, fibulin-4 and fibulin-5 genes revealed a homozygous missense mutation (p.Cys267Tyr) in the fibulin-4 gene in the patient. Our observation increases the number of cases with fibulin-4 mutations to three and extends the phenotypic spectrum of fibulin-4 mutations by microcephaly, overgrowth and arachnodactyly.

Database: Medline

**Author(s):** Klaassen, Ilka; Neuhaus, Thomas J; Mueller-Wiefel, Dirk E; Kemper, Markus J

**Source:** Nephrology, dialysis, transplantation : official publication of the European Dialysis and Transplant Association - European Renal Association; Feb 2007; vol. 22 (no. 2); p. 432-439

**Publication Date:** Feb 2007

**Publication Type(s):** Journal Article

Available in full text at Nephrology Dialysis Transplantation - from Oxford University Press;
Collection notes: To access please select Login with Athens and search and select NHS England as your institution before entering your NHS OpenAthens account details.

Available in full text at Nephrology Dialysis Transplantation - from Highwire Press

**Abstract:** BACKGROUND Prognosis of fetuses with renal oligohydramnios (ROH) is often still regarded as poor. Neonatal complications and the long-term follow-up of fetuses with ROH in two pediatric centres are described. Method. 23 fetuses (16 males, 7 females) were included as patients. Primary diseases included congenital anomalies of the kidney and urinary tract (n = 16), autosomal recessive polycystic kidney disease (n = 4) and renal tubular dysgenesis (n = 3). The analysis includes retrospective chart review. RESULTS Seven children died (30%), the majority (n = 4, 17%) within the neonatal period due to pulmonary hypoplasia and renal insufficiency. Fourteen patients (61%) required postnatal mechanical ventilation for a median of 4 (range 1-60) days; 11 infants had an associated pneumothorax. All 16 surviving children have chronic kidney disease (CKD) at a current median age of 5.7 years (range 0.5-14.5), managed conservatively in eight patients [median glomerular filtration rate 51 (range 20-78) ml/min/1.73 m(2)]. Eight patients reached end-stage renal disease at a median age of 0.3 years (range 2 days to 8.3 years), including one patient with preemptive kidney transplantation. Five of the patients requiring dialysis underwent successful renal transplantation at a median age of 3.5 years (range 2.5-4). Growth was impaired in seven children requiring growth hormone treatment. Cognitive and motor development was normal in 12 (75%) of the 16 patients and showed a delay in four children, including two with associated syndromal features. CONCLUSION ROH is not always associated with a poor prognosis and long-term outcome in survivors is encouraging. The high incidence of neonatal complications and long-term morbidity due to CKD requires a multidisciplinary management of these children.

**Database:** Medline


**Author(s):** Herman, T E; Siegel, M J

**Source:** Journal of perinatology : official journal of the California Perinatal Association; Sep 2000; vol. 20 (no. 6); p. 397-398

**Publication Date:** Sep 2000

**Publication Type(s):** Case Reports Journal Article

Available in full text at Journal of Perinatology - from Nature Publishing Group

Available in full text at Journal of Perinatology - from ProQuest

**Database:** Medline
9. Nonfatal symptomatic spontaneous pneumothorax in neonates: Association with white ethnicity and lack of association with major urinary tract malformations

Author(s): Brenner J.S.; Karlowicz M.G.
Source: Clinical Pediatrics; Apr 1997; vol. 36 (no. 4); p. 241-243
Publication Date: Apr 1997
Publication Type(s): Journal: Article
Database: EMBASE

10. Fetal renal maldevelopment with oligohydramnios following maternal use of piroxicam.

Author(s): Voyer, L E; Drut, R; Méndez, J H
Source: Pediatric nephrology (Berlin, Germany); Oct 1994; vol. 8 (no. 5); p. 592-594
Publication Date: Oct 1994
Publication Type(s): Case Reports Journal Article
Available in full text at Pediatric Nephrology - from Springer Link Journals
Abstract: A female neonate, born by cesarean section at 37 weeks of gestation, presented with respiratory distress syndrome, right pneumothorax and anuria. A sonogram showed increased echogenicity, with neither hydronephrosis nor macroscopic cysts. Peritoneal dialysis was started on the 14th day because of renal insufficiency, but the newborn died on the 33rd day. Family history was unremarkable, except that the mother received piroxicam at about the 26th week of gestation. A sonogram at the 28th week showed oligohydramnios. Histopathological study of the kidneys revealed crowded glomeruli and only few differentiated proximal convoluted tubules in the inner cortex, abnormally differentiated microcystic tubules and microcystic glomeruli in the outer cortex. Periodic acid-Schiff staining showed only traces of brush border in the dilated tubules of the outer cortex. Immunoperoxidase staining for epithelial membrane antigen was positive in the luminal border of all tubules. Electron microscopy confirmed the presence of brush border remnants and other proximal tubular characteristics in some segments. The renal abnormality bears some similarities to that found in familiar renal tubular dysgenesis, but it fits better with those described after maternal use of angiotensin converting enzyme inhibitors and nonsteroidal anti-inflammatory drugs. The lesion in this case appears to have resulted from fetal exposure to piroxicam. Recently, a second pregnancy ended in a completely normal female newborn.
Database: Medline
11. Pulmonary hypoplasia associated with oligohydramnios: report of five cases.

**Author(s):** Hsu, W C; Hung, H Y; Huang, F Y; Kao, H A; Lee, H C; Shih, C C; Chen, P F

**Source:** Zhonghua Minguo xiao er ke yi xue hui za zhi [Journal]. Zhonghua Minguo xiao er ke yi xue hui; 1990; vol. 31 (no. 6); p. 388-395

**Publication Date:** 1990

**Publication Type(s):** Case Reports Journal Article

**Abstract:** From January 1986 to December 1988, 5 cases of oligohydramnios with pulmonary hypoplasia were experienced. Clinically, all of the newborn infants had unique faces and evidence of respiratory distress with tachypnea, cyanosis, intercostal retractions, nasal flaring and grunting respiration presented within minutes after birth. Usually, they had no response to respiratory therapy. Most of them showed small lung volume by chest roentgenogram and were prone to develop pneumothorax. All 5 cases did not survive more than a few hours. Autopsy findings disclosed pulmonary hypoplasia. Four of them had urinary tract malformation, and the other one had the history of prolonged leakage of amniotic fluid.

**Database:** Medline


**Author(s):** Rupprecht, V E; Kabus, M; Schwarze, R

**Source:** Kinderarztliche Praxis; Nov 1989; vol. 57 (no. 11); p. 545-551

**Publication Date:** Nov 1989

**Publication Type(s):** Case Reports Journal Article

**Abstract:** Within a period of 3 years 4 mature male neonates were observed with an unusual complex of symptoms: subvesical obstruction (valve) with spontaneous pneumothorax/pneumomediastinum. All infants showed immediately postnatal respiratory insufficiency. There are pathogenetical relations between renal and urinary tract malformations and pulmonary changes, which are recognized in Potter's sequence with pulmonary hypoplasia. The authors postulate a deranged dynamic of pulmonary fluid by oligohydramnios, causing increased vulnerable lungs also in such cases without typical x-ray signs of pulmonary hypoplasia. A spontaneous pneumothorax/pneumomediastinum in newborn infants should be the sign to perform nephro-urologic (sonographic) examination.

**Database:** Medline
13. Pulmonary hypoplasia in a regional perinatal unit.

**Author(s):** Knox, W F; Barson, A J

**Source:** Early human development; Jul 1986; vol. 14 (no. 1); p. 33-42

**Publication Date:** Jul 1986

**Publication Type(s):** Research Support, Non-u.s. Gov't Journal Article

**Abstract:** 83 cases of pulmonary hypoplasia were found in 709 perinatal necropsies in St. Mary's Hospital between 1976 and 1983. 49 of these infants were inborn, representing an incidence of 1.4 per 1000 births. Diaphragmatic hernia was present in 43% and was the commonest reason for referral, usually to the neonatal surgical unit, from an outside hospital. Renal malformation, often with oligohydramnios, was seen in 25%, and 11% had no other major disease. The respiratory symptoms associated with pulmonary hypoplasia in the absence of diaphragmatic hernia seem to have encouraged the referral of these infants to the neonatal medical unit. However, more than half the liveborn infants with pulmonary hypoplasia died during the first postnatal day. In comparison with first week deaths from other causes, short survival was a particular feature of pulmonary hypoplasia in infants weighing less than 2.5 kg and pneumothorax and pulmonary haemorrhage were commoner in term infants with hypoplastic lungs.

**Database:** Medline

14. Radiographic chest contour and pulmonary air leaks in oligohydramnios-related pulmonary hypoplasia (Potter's syndrome).

**Author(s):** Leonidas, J C; Bhan, I; Beatty, E C

**Source:** Investigative radiology; 1982; vol. 17 (no. 1); p. 6-10

**Publication Date:** 1982

**Publication Type(s):** Journal Article

**Abstract:** Review of the clinical records, chest roentgenograms, and autopsy material of 17 infants dying from oligohydramnios related pulmonary hypoplasia confirmed that two roentgen signs appeared with high frequency. These were pneumomediastinum/pneumothorax (82%) and a bell-shaped chest contour (59%). The bell-shaped chest is valuable in predicting the most severe degrees of lung growth impairment in these infants, and was present in all cases in which oligohydramnios was caused by bilateral renal agenesis. Either pneumomediastinum/pneumothorax or the bell-shaped chest configuration, when present alone, are of low diagnostic value as they are relatively nonspecific signs; their combination, however, has predictive value for Potter's syndrome and may be important in evaluation infants with less typical clinical manifestations of Potter's syndrome.

**Database:** Medline
15. Radiographic chest contour and pulmonary air leaks in oligohydramnios-related hypoplasia (Potter’s syndrome)

**Author(s):** Leonidas J.C.; Bhan I.; Beatty E.C.

**Source:** Investigative Radiology; 1982; vol. 17 (no. 1); p. 6-10

**Publication Date:** 1982

**Publication Type(s):** Journal: Article

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**Database:** EMBASE

16. Value of chest radiography in the diagnosis of Potter’s syndrome at birth

**Author(s):** Leonidas J.C.; Fellows R.A.; Hali R.T.

**Publication Date:** 1975

**Publication Type(s):** Journal: Article

**Abstract:** The authors describe the appearance of the chest roentgenogram in cases of pulmonary hypoplasia associated with Potter’s syndrome or maternal oligohydramnios. Their experience is based on 7 newborn infants with ‘Potter’s syndrome’. The ‘air block syndrome’ was present in all 6 infants with renal nonfunction. The presented series of patients with pulmonary hypoplasia is small, since the disease is relatively uncommon. However, there is enough evidence from this patient material, as well as from observations of published roentgenograms, to warrant the conclusion that pneumomediastinum and/or pneumothorax in the newborn, associated with decreased thoracic volume and poorly expanded and ‘structureless’ lungs, should be viewed with suspicion of renal nonfunction and/or maternal oligohydramnios.

**Database:** EMBASE


**Author(s):** Mooney, J K; Berdon, W E; Lattimer, J K

**Source:** The Journal of urology; Feb 1975; vol. 113 (no. 2); p. 272-278

**Publication Date:** Feb 1975

**Publication Type(s):** Journal Article

**Abstract:** Infants with posterior urethral valves may seem to have diverse and unrelated symptoms when, in fact, the clinical findings are all related to the primary effect of the valves during various stages of early growth. In some, prenatal urinary obstruction leads to such severe oligohydramnios that the fetus is stillborn. Others, somewhat less affected, are born alive but have severe respiratory distress from hypoplastic (stiff) lungs and die of respiratory problems. Still others can be associated with massive ascites and urinomas, and be stillborn or die soon after birth. In less severe cases the neonates may have unexplained respiratory distress with pneumomediastinum or pneumothorax as the only indication of obstructive urologic disease with deficient urinary output. Urinomas or ascites
may later develop beyond the neonatal period as the post-natal obstructive effects of the valves accumulate and the urinary system ruptures and decompresses itself. Finally when the urinary system does not decompress itself, the back pressure can lead to rapid and progressive renal damage until the kidneys can no longer concentrate urine and lose water. The infant becomes dry, acidotic and paradoxically at this stage, puts out large quantities of dilute urine. Physicians caring for infants should be highly suspicious of posterior urethral valves in any male infant with unexplained respiratory distress or metabolic derangements, abdominal distension or flank masses.

Database: Medline

18. Pneumothorax and renal disease in a newborn

Author(s): Wilkinson R.H.; Wheeler D.B.
Source: Annales de Radiologie; 1973; vol. 16 (no. 3); p. 235-238
Publication Date: 1973
Publication Type(s): Journal
Abstract: There appears to be a spectrum of renal anomalies associated with a variable degree of pulmonary hypoplasia which should be suspected in the newborn infant with pneumothorax. This is particularly true of those infants showing the facies described by Potter. In addition, those infants with pneumothorax and with an obstetrical history of oligohydramnios and who have enlarged kidneys or a diminished urine output should have a urinary tract investigation. This should be performed not only from a prognostic point of view, but occasionally an infant may have a correctible urinary tract abnormality.

Database: EMBASE


Author(s): Renert, W A; Berdon, W E; Baker, D H; Rose, J S
Source: Radiology; Oct 1972; vol. 105 (no. 1); p. 97-105
Publication Date: Oct 1972
Publication Type(s): Journal Article
Abstract: Nine cases of newborns with pneumothorax and/or pneumomediastinum are presented. In 7 this was associated with Severe fetal urologic malformations; oligohydramnios was sometimes noted and pulmonary hypoplasia Was severe. In 2 patients there was a lag of weeks to months between the neonatal respiratory distress and the detection of urologic problems. This may reflect mild pulmonary hypoplasia with survival. The possible association of oligohydramnios and urologic abnormalities of the fetus to neonatal respiratory distress is discussed. Early urologic evaluation is recommended when “unexplained” respiratory problems are noted at birth.

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Database: Medline
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