Scoliosis creation by growth modulation of the chest cage in a fetal ovine model

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Introduction

- Thoracic insufficiency syndrome is the inability of the thorax to support normal respiration and lung growth
- Congenital scoliosis and fused ribs can create a 3-D deformity of the chest wall affecting the normal thoracic growth and function

Introduction

- Treatment with expansion thoracoplasty has been recently suggested
 - Campbell et al. JBJS 2004
- Improvements in lung volumes have been reported following expansion thoracoplasty
 - Campbell et al. JBJS 2004
 - Gollogly et al. JPO 2004

Introduction

- Even if lung volume improves, it remains unclear if the actual volume improvements result in greater number of alveoli and if the lung is more functional.
- The augmentation in lung volume could be a distention of the existing lung parenchyma

Rationale

- Animal model of chest wall deformity mimicking the thoracic insufficiency syndrome are rare
- Congenital anomalies represent a disease continuum that starts early in during fetal life and progresses thereafter
- Such an animal model would be useful in evaluating the impact of expansion thoracoplasty.

Methods

- 14 pregnant ewes
- Surgery performed between 65-70 days of gestation (term: 140 days)
- U/S demonstrated 1-2 fetuses.
- Ewes intubated, monitored, surgical prep.
- Surgery performed on 15 lambs.

Methods

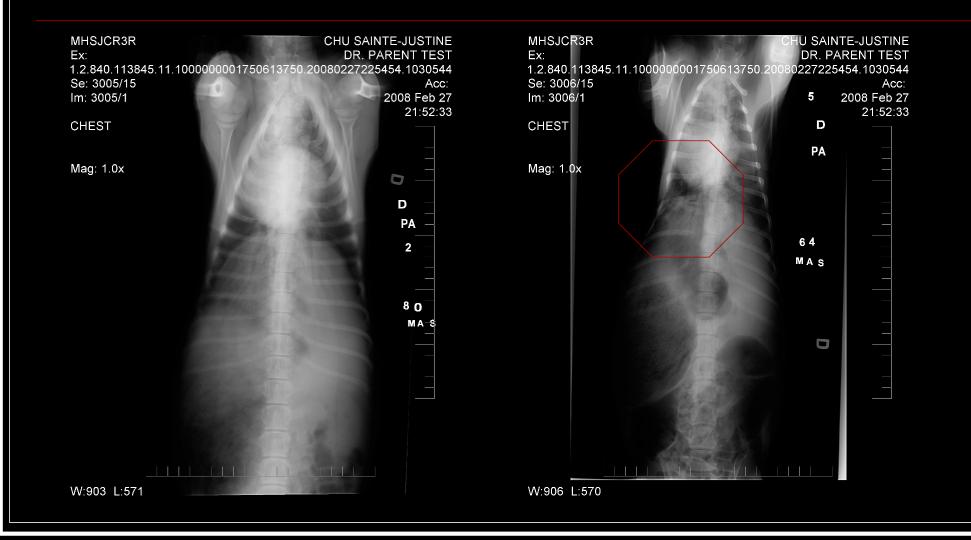
- Thoracic deformity was created by tying 3 ribs together or adding a resection of the 7th rib in addition to partial destruction of the vertebral growth plate (n = 9).
- Resection of 2 ribs n=6
- Pulmonary function tests done on the second group



Results

- All 15 lambs who underwent surgery were born at term through vaginal delivery (one shoulder dystocia) and 6 controls
- Two lambs died in the first hours of life (severe deformity)
- Another lamb died of failure to thrive (severe deformity)
- All other lambs survived to two months

X-Ray: control & mild scoliosis



X-Ray: Moderate & Severe scoliosis



Lamb suffering from most severe defect







Pulmonary function tests

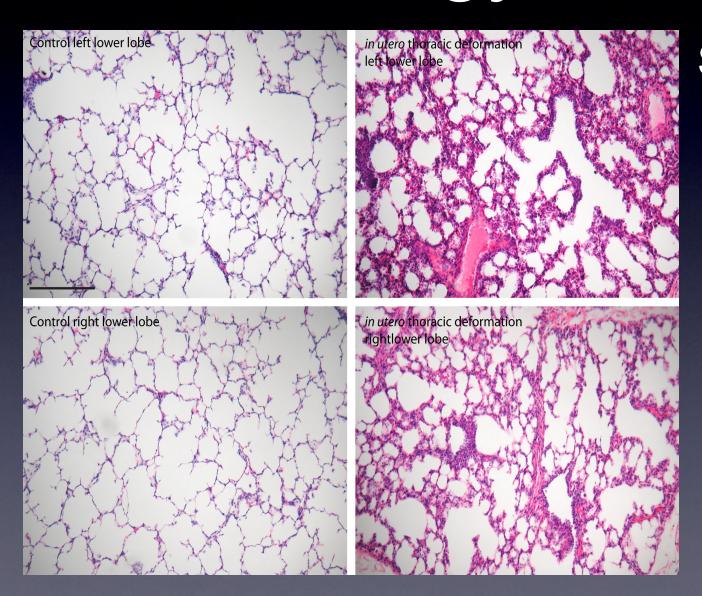
	Sex	Age (Days)	IC (alive)	Respiratory system compliance (alive)	IC (dead) 2 lungs-Left lung- Right lung	Ratio left lung IC/ right lung IC (dead)	Compliance (dead) both lungs-Left lung- Right lung
L227 C	M	3	47	13	62.5-21.5-36	0.6	8.5-2.5-5
L405 C	M	3	48.5	15	64.5-17-38.5	0.4	8.5-2.5-5.5
L406 C	F	3	38	95	44 5-11-24	0.5	6.5-1.5-3
Mean C			43	12.5	57-16.5-33	0.5	8-2-4.5
L235 Def	M	2	35	8.5	•		
L236 Def	F	2	20.5	5.5	-		
Deceased Def	M	1	-	-	10.5-6-8.5	0.7	1.5-0.5-1.5
L376 Def	F	1	12.5	3	12.5-4.5-9	0.5	2-0.5-1.5
L238 Def	F	2	28.5	4.5	26-7.5-16	0.5	2-1.5-1.5
L239 Def	F	2	40	Л	50-14 5-30	0.5	3-1-2
Mean Def			24	4.5	24.5-8-16	0.55	2-1-1.5

Histology

Normal

Left

Left



Scoliotic

Right

Right

Conclusion

- It is possible to create a chest wall deformity in utero in an ovine model with survival of the lamb until birth.
- The thoracic deformity created in utero does not correct spontaneously within the first two months of life.
- Lung development adversely affected by thoracic deformity.
- Pulmonary function tests also adversely affected.

Conclusion

We have created a model of congenital thoracic deformity that can be used to study the impact of the deformity on vital functions and to evaluate treatment strategies.

