

# Respiratory and esophageal morbidity in adults with repaired esophageal atresia

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av  
Vladimir Gatzinsky  
Leg.Läkare

Fakultetsopponent:  
Docent Tomas Wester  
Barnkirurgiska kliniken, Karolinska Universitetssjukhuset, Stockholm

Avhandlingen baseras på följande delarbeten:

- I. **Gatzinsky V, Jönsson L, Johansson C, Göthberg G, Sillén U, Friberg LG**  
Dysphagia in Adults Operated On for Esophageal Atresia-Use of a Symptom Score to Evaluate Correlated Factors.  
*Eur J Pediatr Surg. 2011 Mar;21(2):94-8.*
- II. **Gatzinsky V, Jönsson L, Ekerljung L, Friberg LG, Wennergren G**  
Long-term respiratory symptoms following oesophageal atresia.  
*Acta Paediatr. 2011 Sep;100(9):1222-5.*
- III. **Gatzinsky V, Wennergren G, Jönsson L, Ekerljung L, Houtz B, Redfors S, Sillén U, Gustafsson P**  
Impaired peripheral airway function in adults following repair of esophageal atresia.  
*Accepted for publication, December 2013, J Pediatr Surg*
- IV. **Gatzinsky V, Andersson O, Eriksson A, Jönsson L, Abrahamsson K, Sillén U**  
pH multichannel intraluminal impedance in adults operated for esophageal atresia - what can it tell us?  
*In manuscript*



UNIVERSITY OF GOTHENBURG

# Respiratory and esophageal morbidity in adults with repaired esophageal atresia

Vladimir Gatzinsky

Department of Pediatrics, Institute of Clinical Sciences,  
Sahlgrenska Academy, University of Gothenburg

**Background:** Esophageal atresia (EA) often leads to persistent esophageal and respiratory symptoms, as well as impaired esophageal and lung function in adulthood. The reasons for this, and the connections between symptoms and documented abnormalities, are not fully understood.

**Purpose:** We wanted to investigate a cohort of adults previously operated on for EA in order to describe the prevalence of symptoms and functional abnormalities, as well as to investigate whether, and if so how, they are connected.

**Methods:** Seventy-three of 79 patients operated on for EA in Gothenburg in 1968-1983 agreed to participate in the first part of the study involving questionnaires relating to symptoms from the esophagus and respiratory tract. Twenty-nine patients agreed to undergo further investigations of pulmonary and esophageal function.

**Results:** From the questionnaire studies, we found that, even though the overall quality of life was good, a considerable number of patients had troublesome symptoms. Both esophageal and respiratory symptoms were frequent. Fifty-seven percent experienced swallowing disturbances (dysphagia) which appeared to be associated with regurgitation, which was in turn noted in 40%. Thirty-two percent experienced heartburn. Different respiratory symptoms, such as wheeze and long-standing cough (44% and 30% respectively), were much more common in this cohort than in the general population. Asthma was reported by 30%, even though no predisposing factors were noted. Impaired respiratory function, either obstructive and/or restrictive, was noted in 22/28 (79%). The obstruction was mainly in the peripheral airways, 17/28 (61%) subjects (measured by multiple-breath inert gas washout, MBW), while only six (21%) subjects displayed values indicating central obstruction. Nine patients had restrictive disease. Airway hyper-responsiveness was frequent and associated with atopy and airway inflammation. However, respiratory symptoms or doctor-diagnosed asthma (DDA) did not correlate with any specific lung function test abnormality. There was a high prevalence of gastro-esophageal reflux (GER) measured by pH multichannel intraluminal impedance (pH-MII) involving both pathological reflux episodes with a pH of < 4 and of > 4 (5/15 and 10/15 subjects respectively). Dysphagia correlated to the number of weakly acidic reflux episodes, while esophageal mucosal damage (14/24 subjects with esophagitis, two of whom had Barrett's esophagus) correlated to the reflux index (RI) and the number of episodes of weakly acidic reflux. Lower esophageal sphincter incompetence to any extent was frequent (21/24 subjects) and correlated to the number of acid reflux episodes and RI.

**Conclusion:** A high prevalence of both respiratory and esophageal symptoms remains in adulthood. The impaired pulmonary function appears to be more pronounced than previously described. Even non-acidic reflux episodes appear to contribute to the esophageal morbidity. New investigative modalities such as MBW and pH-MII have helped us in further describing and understanding the late sequelae of EA. Classical asthma appears to be difficult to diagnose in this patient group. Given the high prevalence of both respiratory and esophageal morbidity, further studies and long-term follow-up, including MBW and pH-MII, are warranted.

**Keywords:** esophageal atresia, long-term outcome, pulmonary function, gastro-esophageal reflux, dysphagia