

# Risk of cholesteatoma among non-syndromic orofacial cleft patients and their siblings

A study based on four national registers

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# Background

## Orofacial clefts

- Cleft lip
- Cleft lip and palate
- Cleft palate

1-2 per 1000 live births in DK

## Cholesteatoma

Surgically treated

5-10 per 100.000 person years in DK

on a national basis,

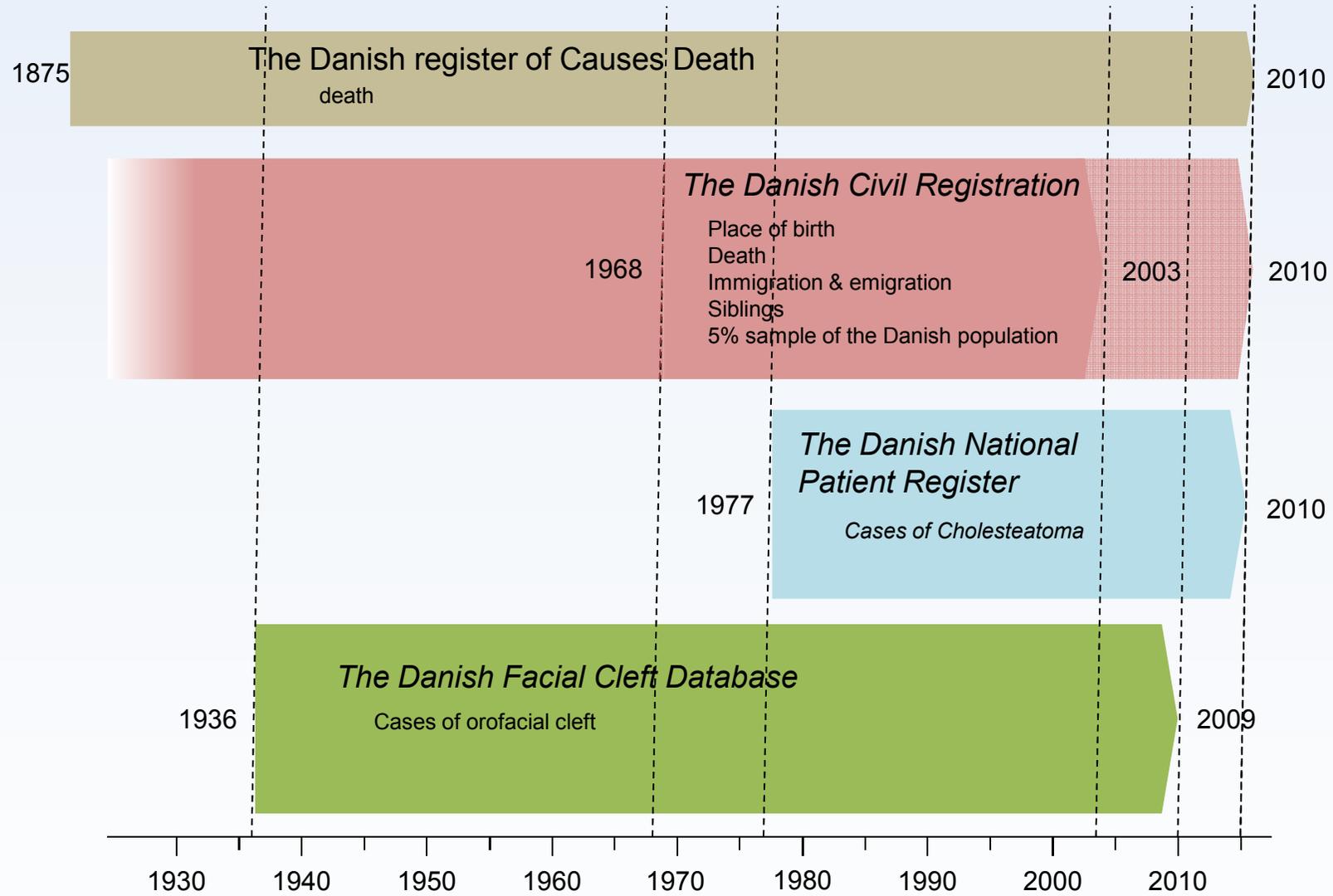
to estimate the risk of cholesteatoma surgery among

- 1) Orofacial cleft patients
- 2) Siblings to orofacial cleft patients

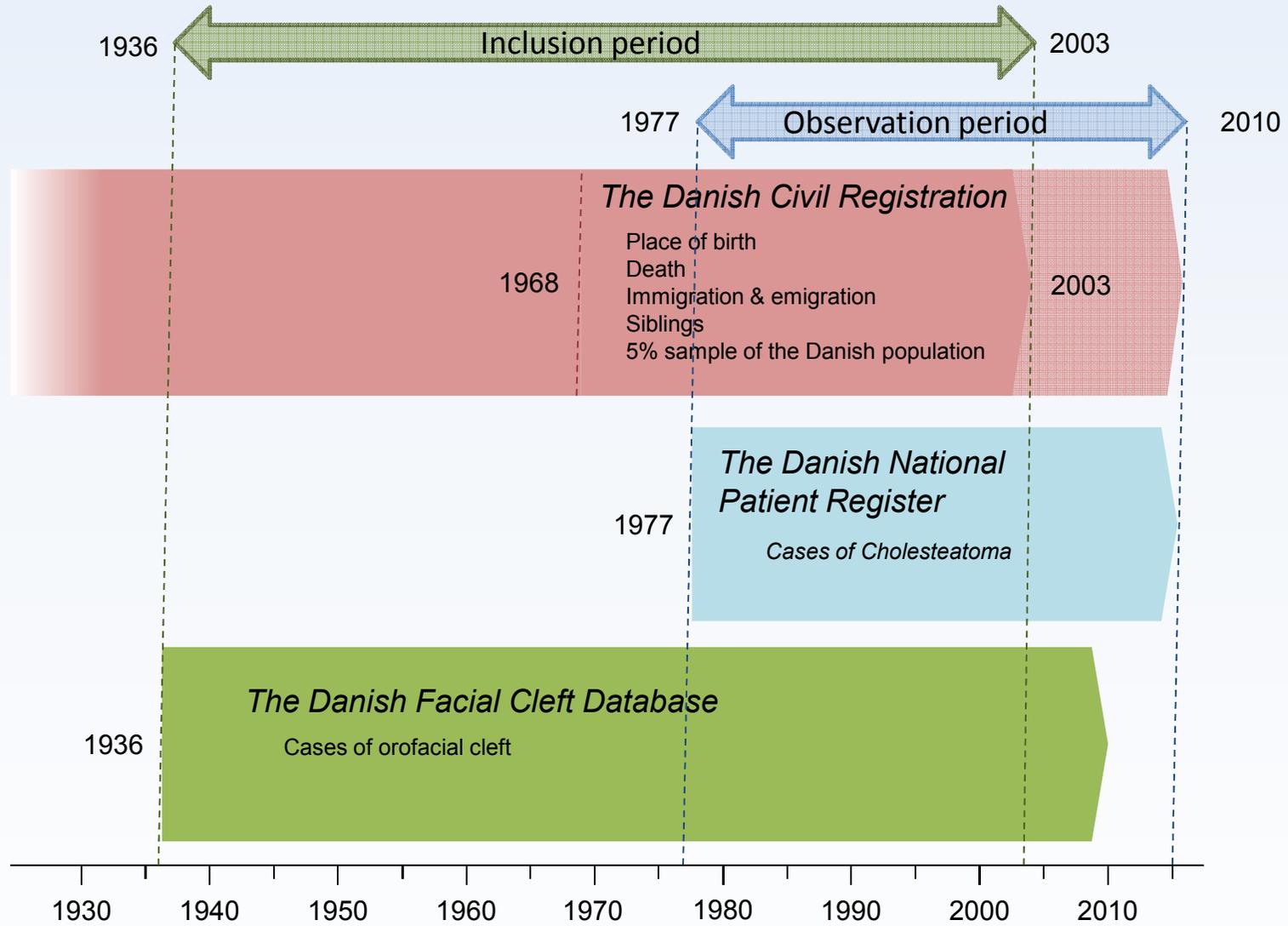
using survival analysis and

a comparison group of a random 5% sample of the Danish population

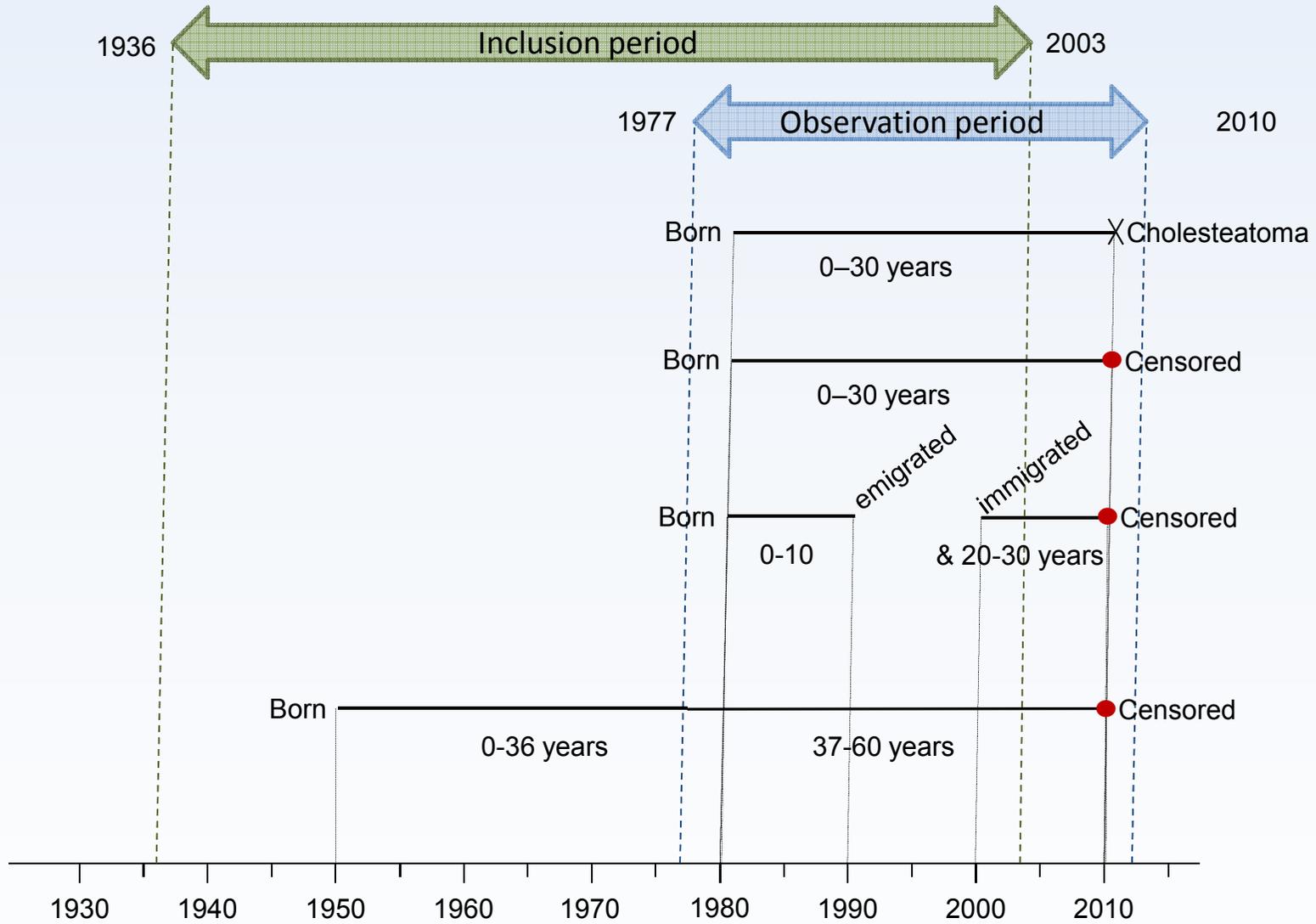
# Material and Methods



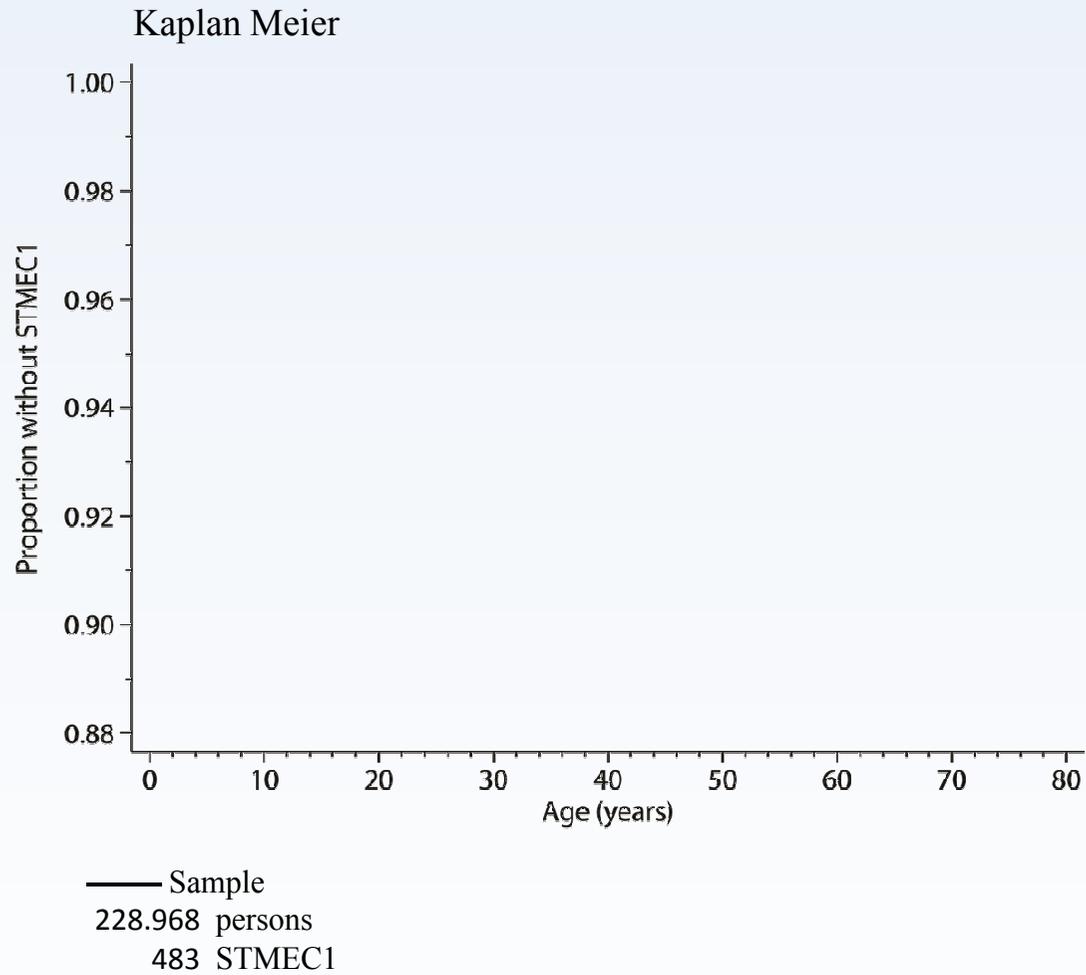
# Material and Methods



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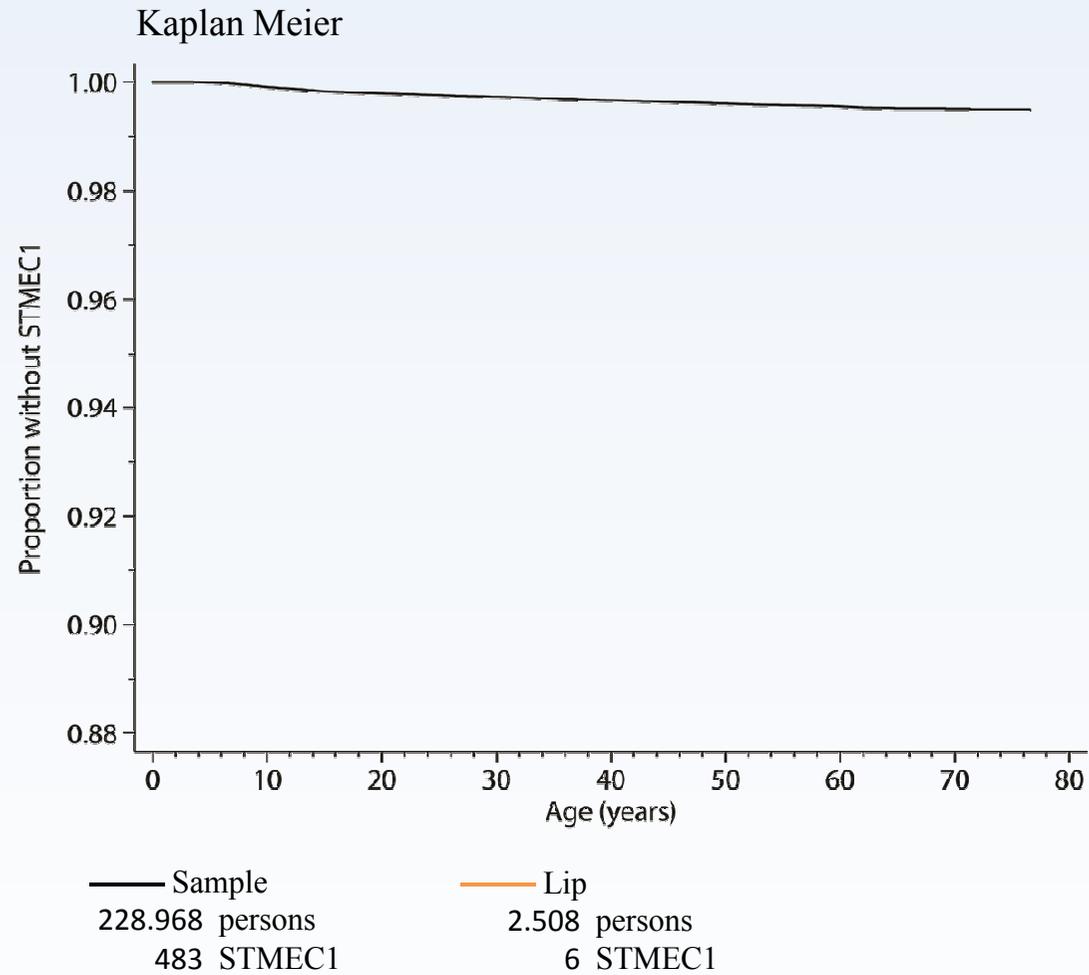


# Results



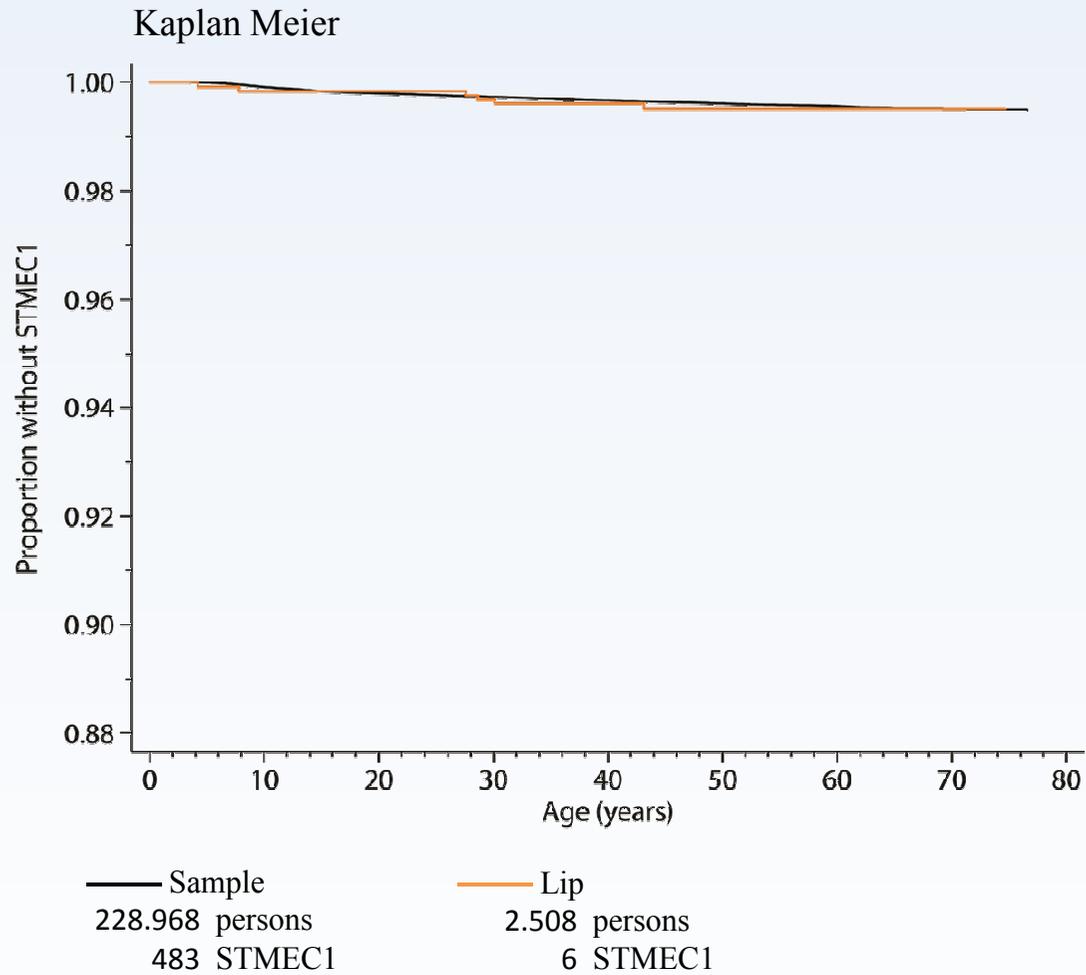
STMEC1: First-time surgically treated middle ear cholesteatoma

# Results



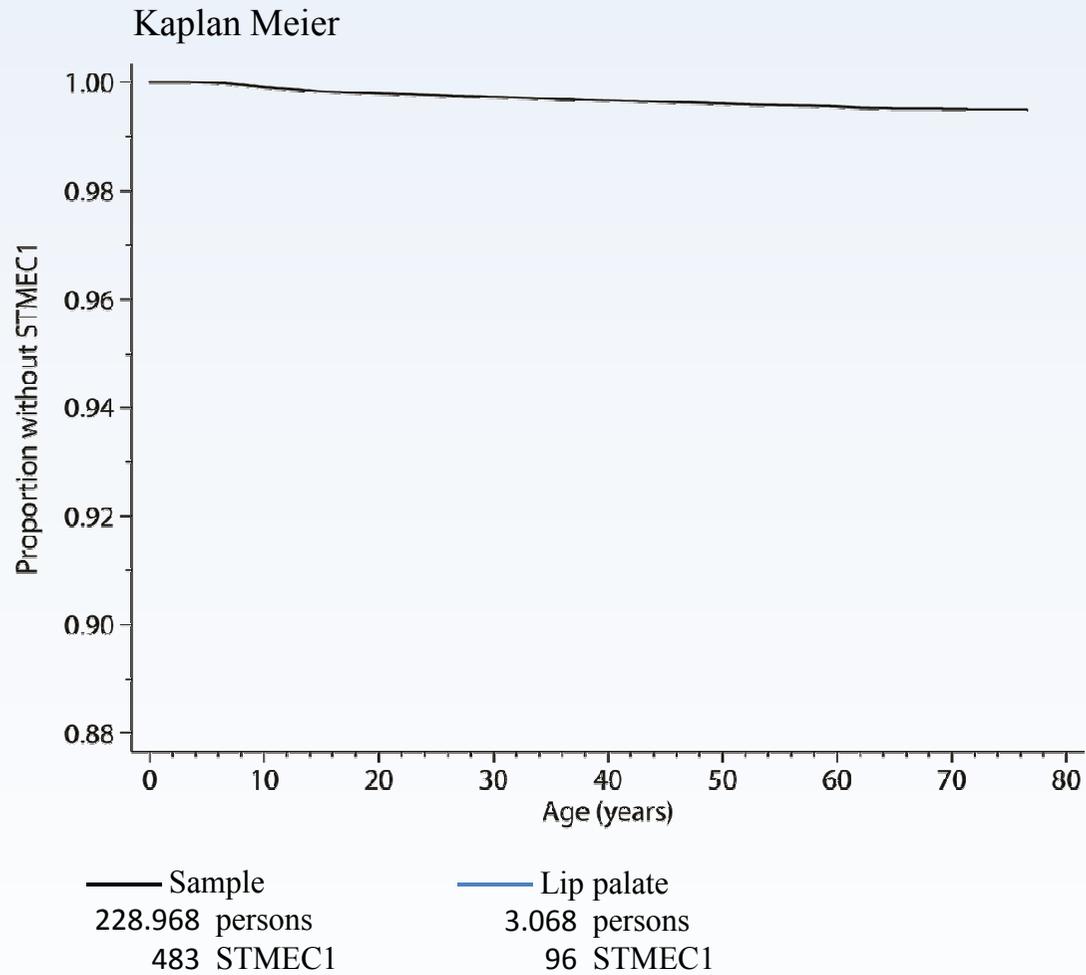
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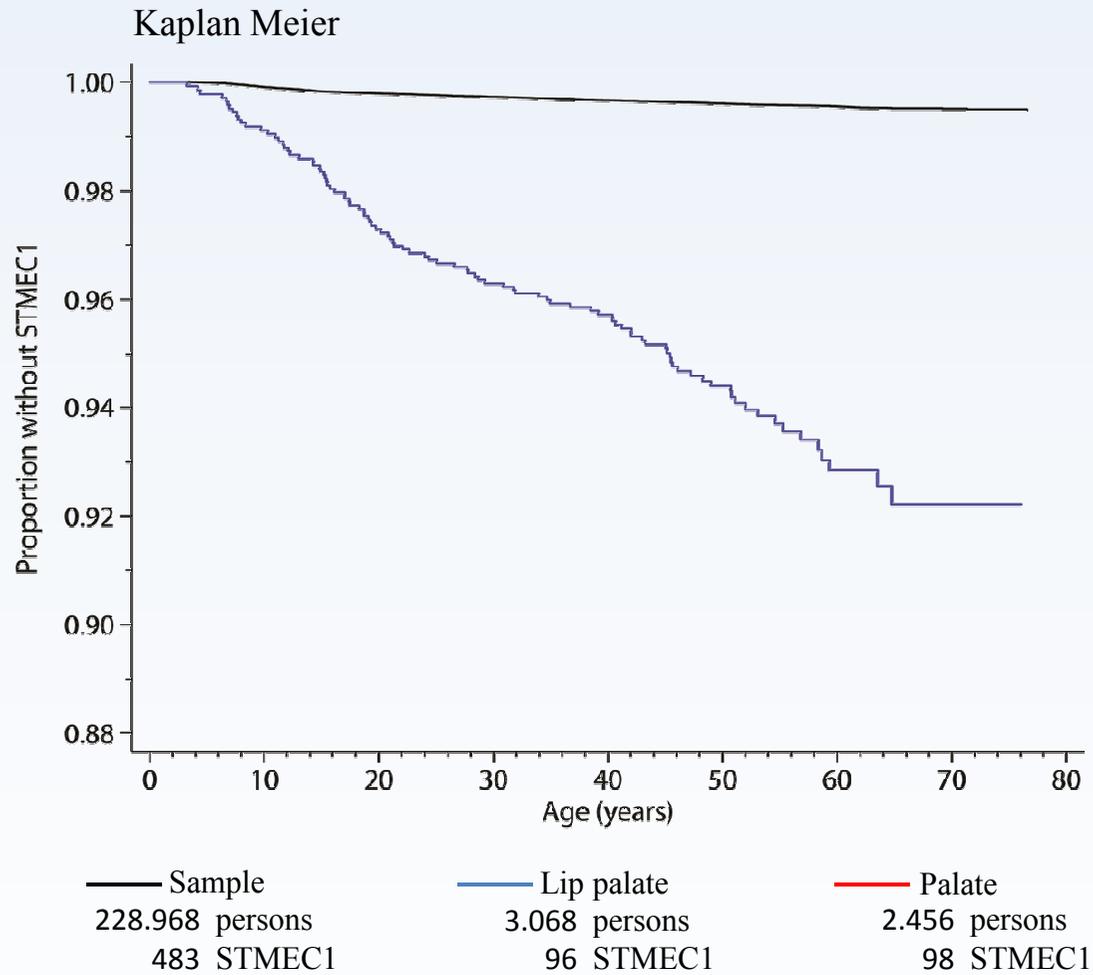
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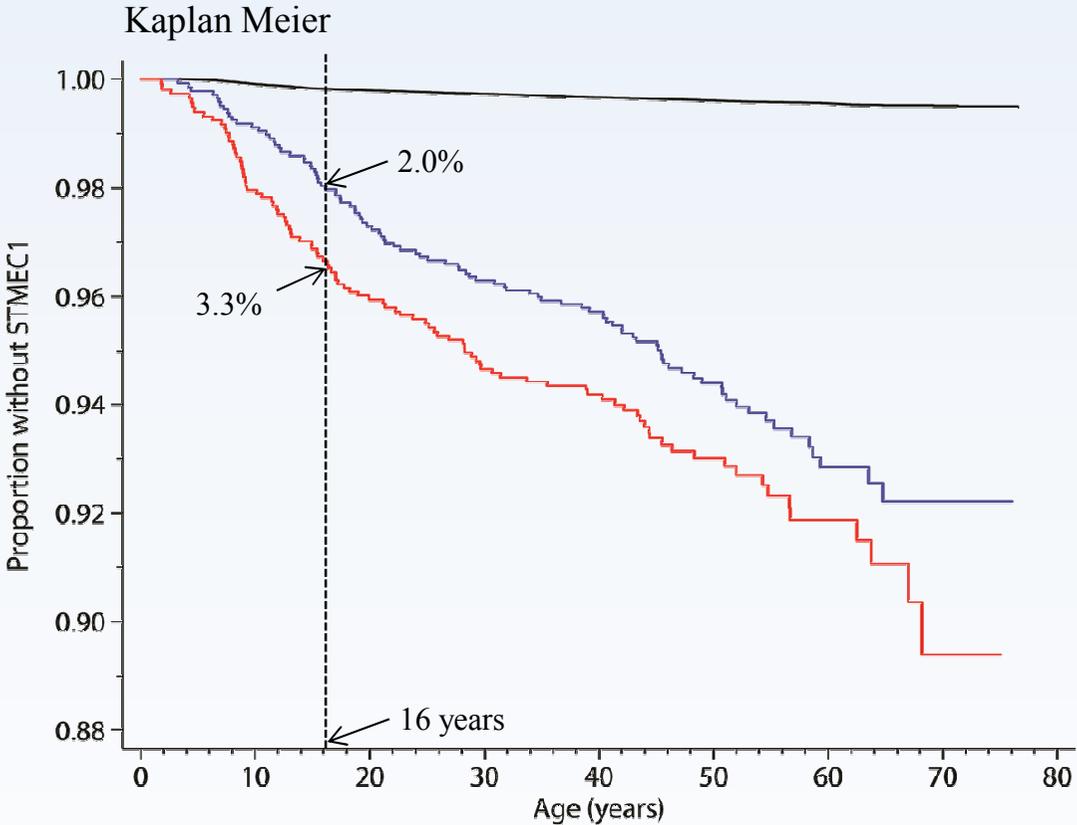
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STMEC1: First-time surgically treated middle ear cholesteatoma

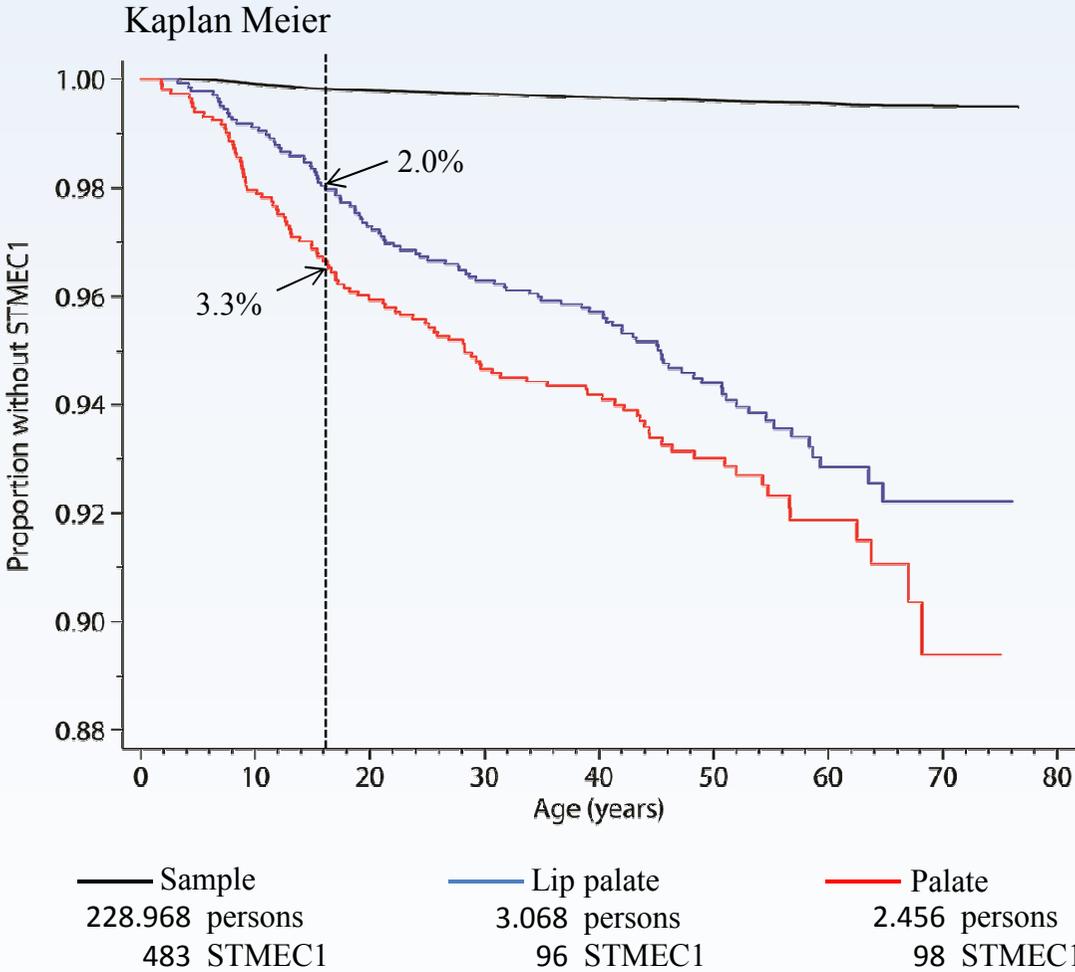
# Results



— Sample	— Lip palate	— Palate
228.968 persons	3.068 persons	2.456 persons
483 STMEC1	96 STMEC1	98 STMEC1

STMEC1: First-time surgically treated middle ear cholesteatoma

# Results



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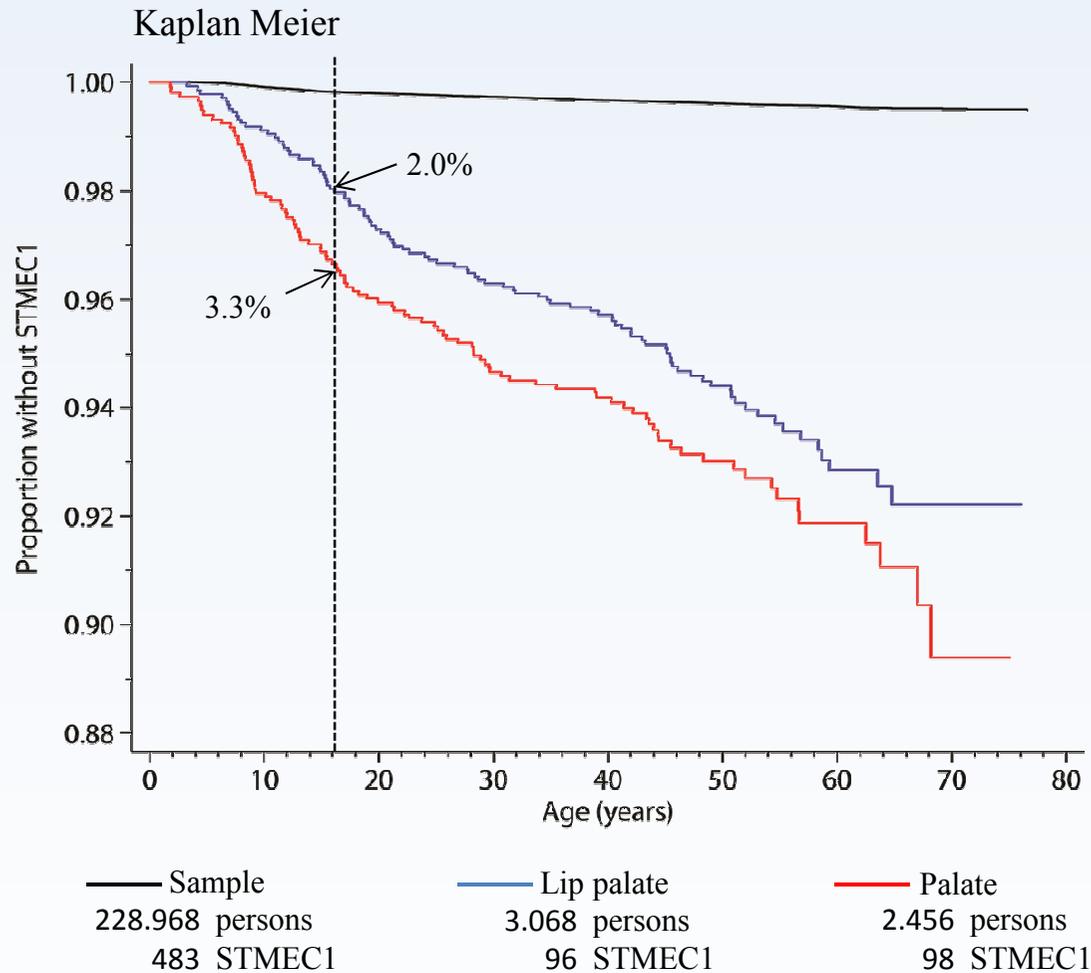
HR for STMEC1 by cleft-type using Cox regression analysis

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STMEC1: First-time surgically treated middle ear cholesteatoma

HR: Hazard rate

# Results




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HR for STMEC1 by cleft-type using Cox regression analysis

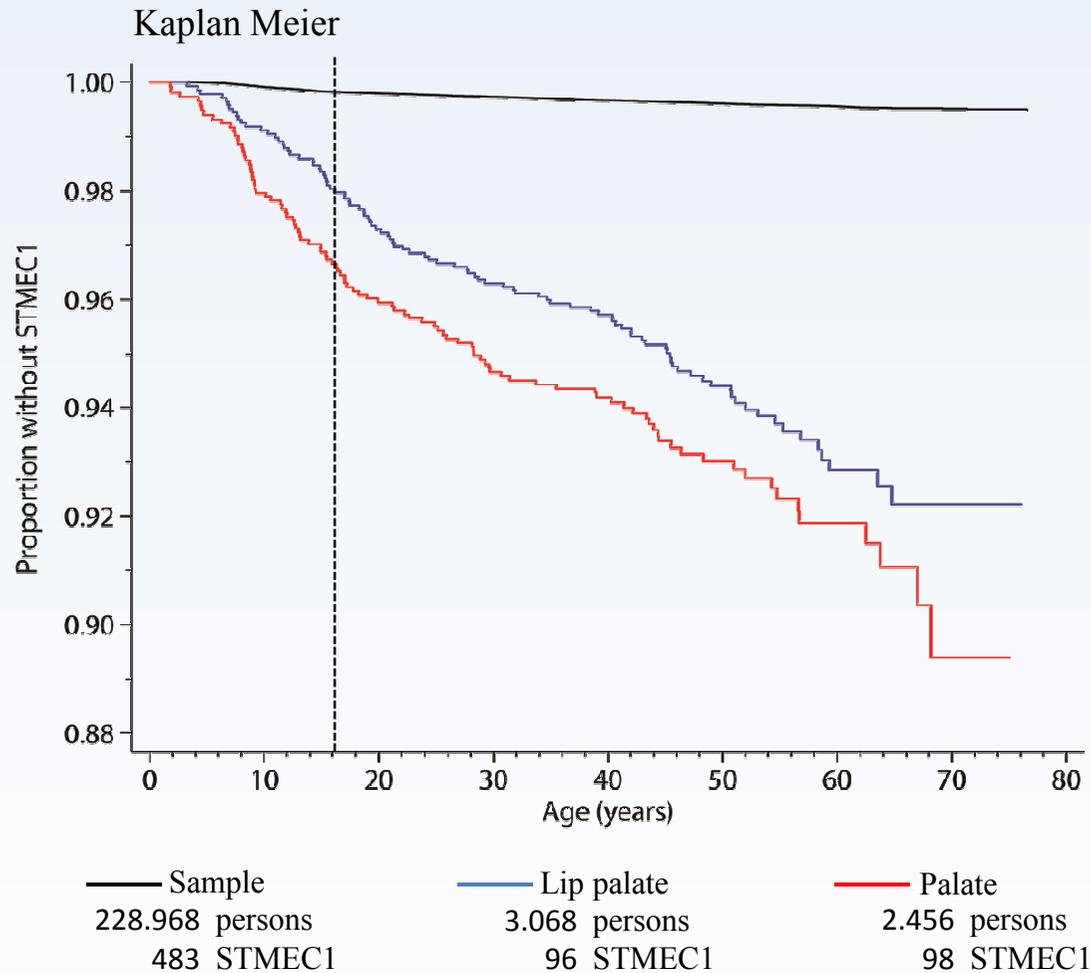
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	HR	95% CI
<b>All</b>		
Cleft lip and palate	14	12-18
Cleft palate	19	16-24

STMEC1: First-time surgically treated middle ear cholesteatoma

HR: Hazard rate

# Results



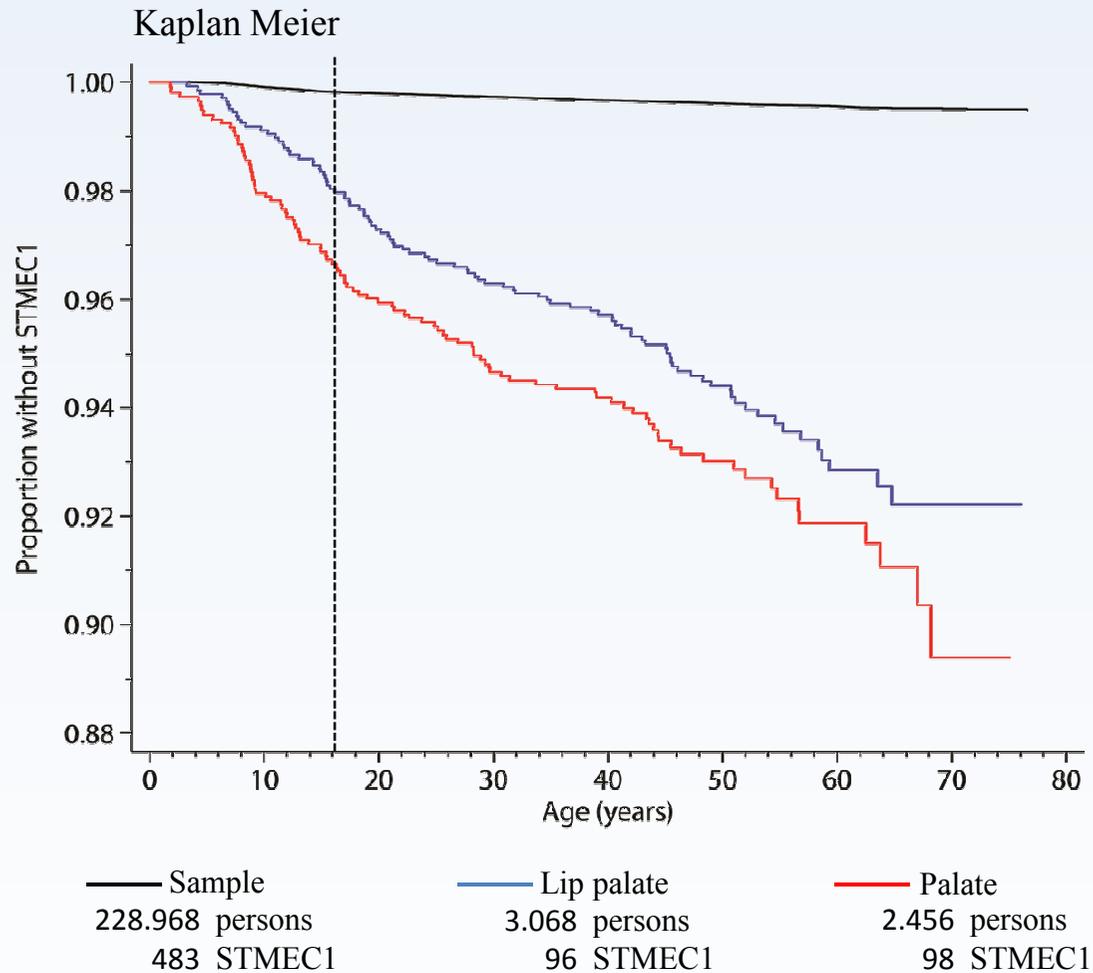
HR for STMEC1 by cleft-type using Cox regression analysis

	HR	95% CI
<b>All</b>		
Cleft lip and palate	14	12-18
Cleft palate	19	16-24
<b>Children</b>		
Cleft lip and palate	11	7-16
Cleft palate	19	14-27

STMEC1: First-time surgically treated middle ear cholesteatoma

HR: Hazard rate

# Results




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HR for STMEC1 by cleft-type using Cox regression analysis

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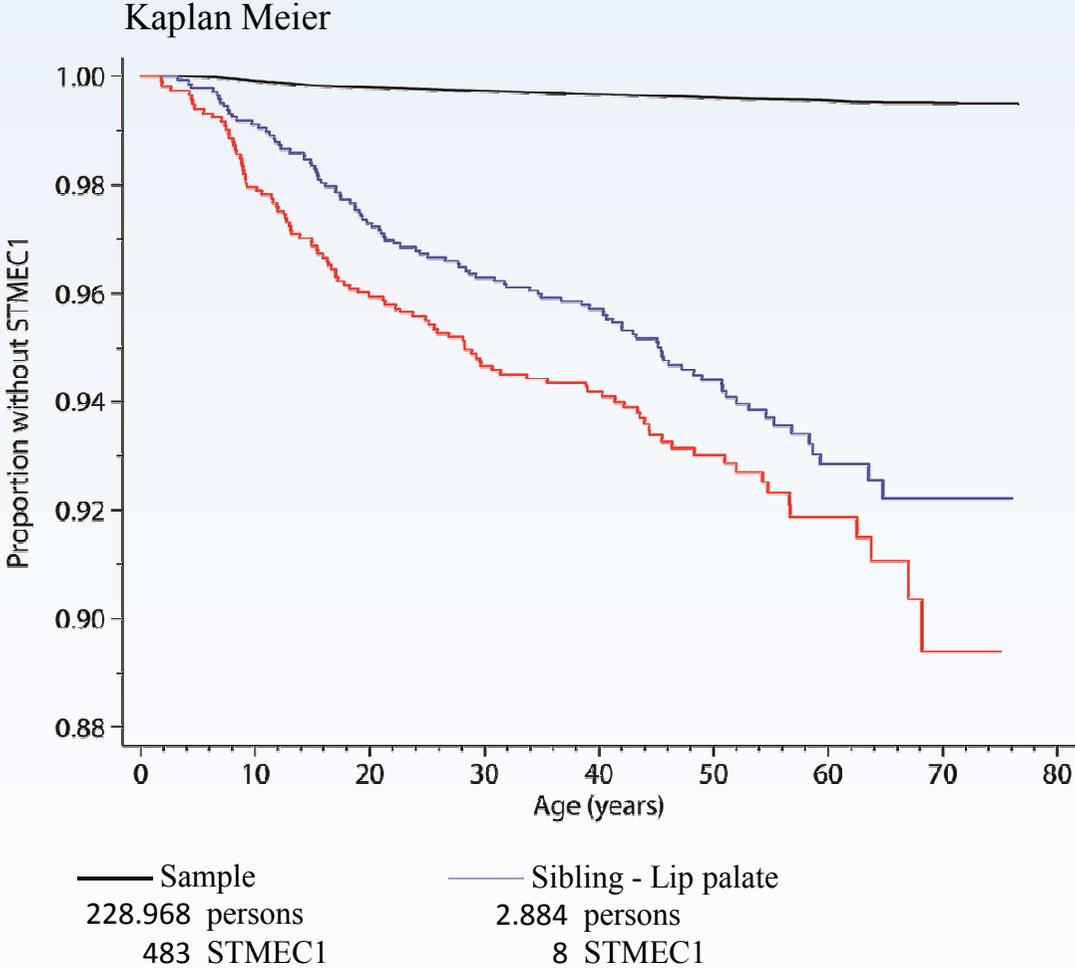
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<b>All</b>		
Cleft lip and palate	14	12-18
Cleft palate	19	16-24
<b>Children</b>		
Cleft lip and palate	11	7-16
Cleft palate	19	14-27
<b>Adults</b>		
Cleft lip and palate	16	13-22
Cleft palate	19	15-26

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STMEC1: First-time surgically treated middle ear cholesteatoma

HR: Hazard rate

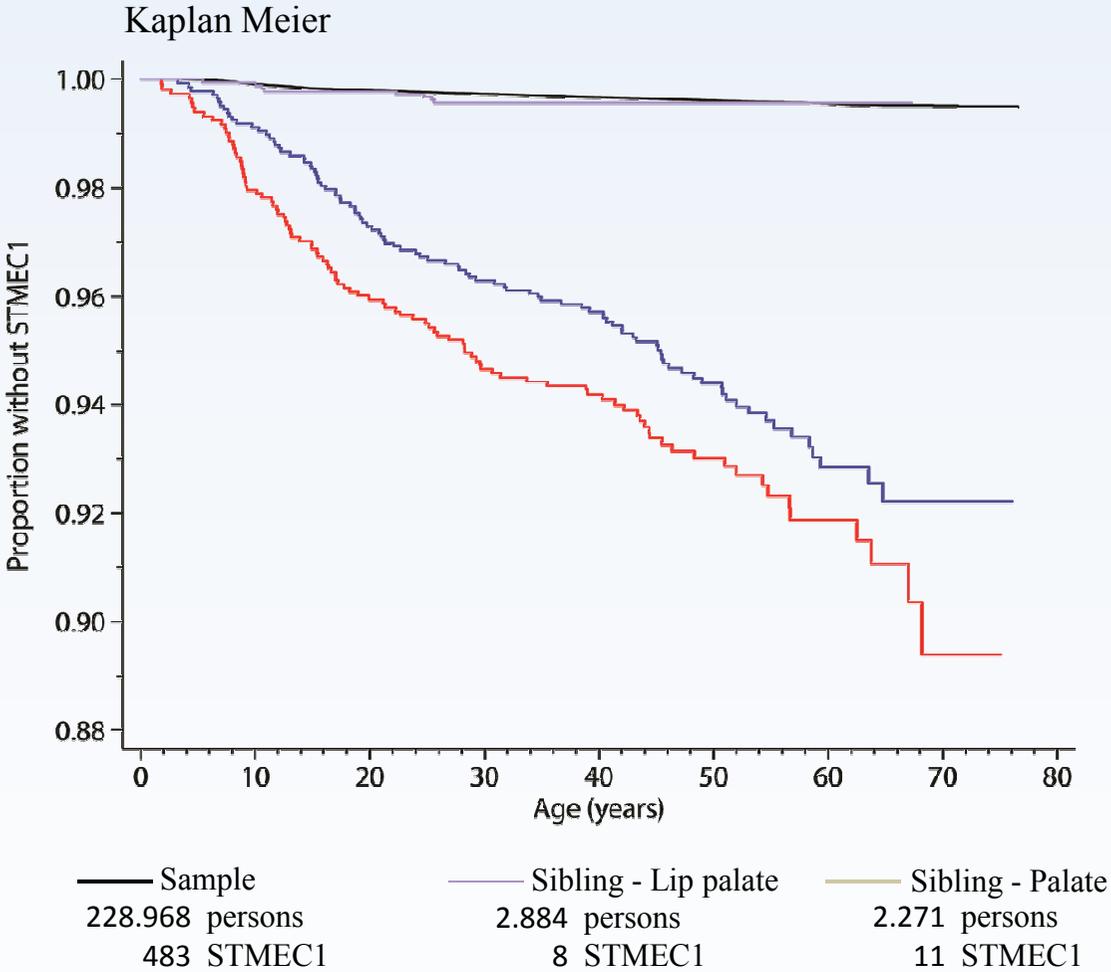
# Results



STMEC1: First-time surgically treated middle ear cholesteatoma

HR: Hazard rate

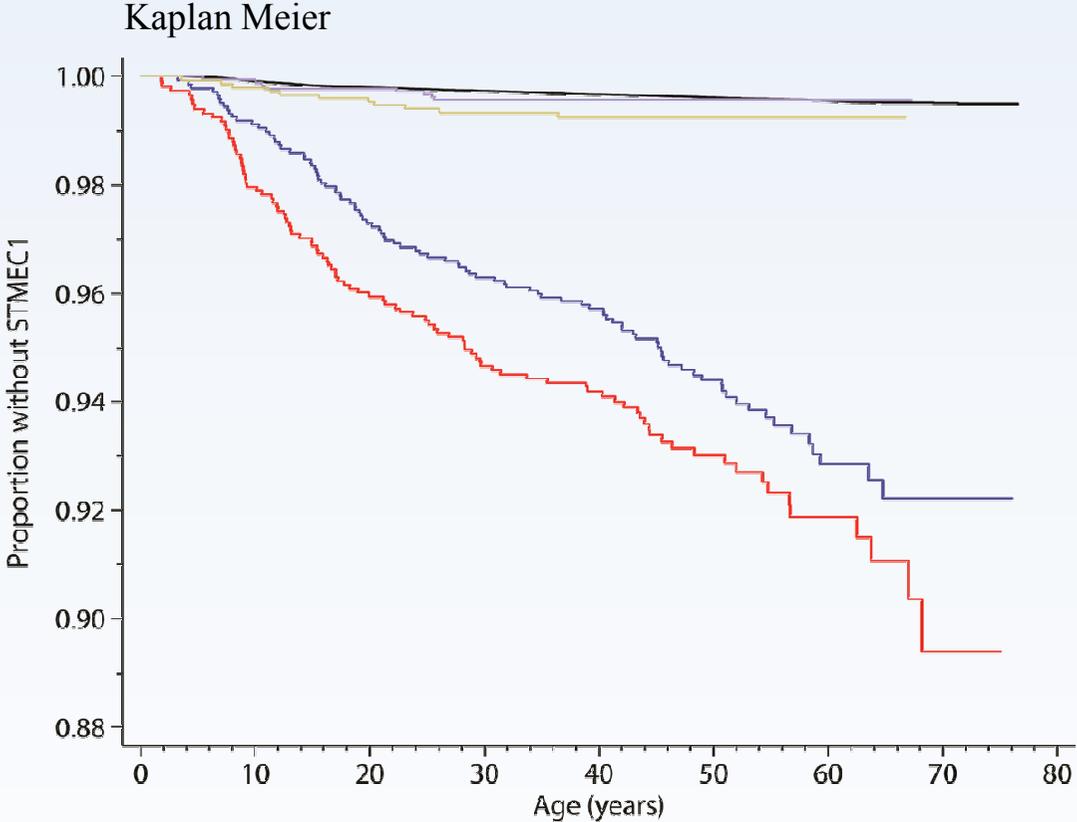
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STMEC1: First-time surgically treated middle ear cholesteatoma

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HR for STMEC1 by cleft-type using Cox regression analysis

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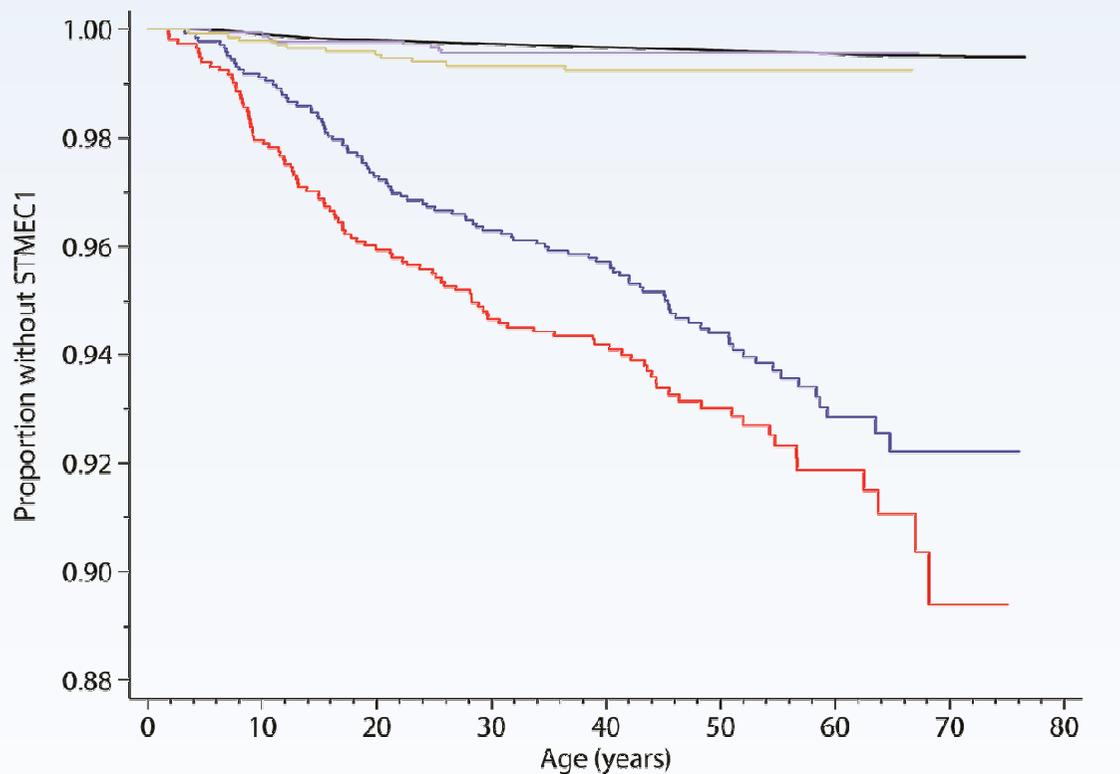
— Sample	— Sibling - Lip palate	— Sibling - Palate
228.968 persons	2.884 persons	2.271 persons
483 STMEC1	8 STMEC1	11 STMEC1

STMEC1: First-time surgically treated middle ear cholesteatoma

HR: Hazard rate

# Results

Kaplan Meier



— Sample	— Sibling - Lip palate	— Sibling - Palate
228.968 persons	2.884 persons	2.271 persons
483 STMEC1	8 STMEC1	11 STMEC1

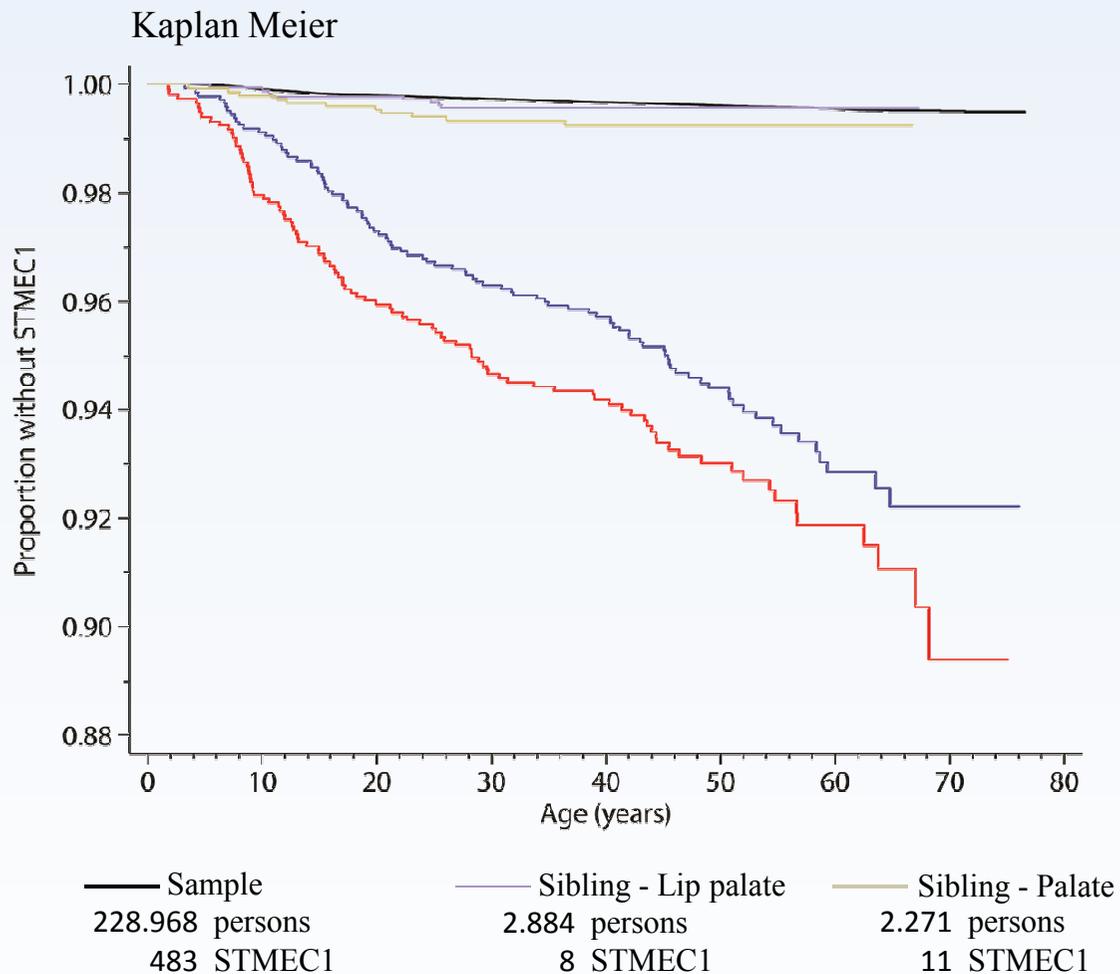
STMEC1: First-time surgically treated middle ear cholesteatoma

HR for STMEC1 by cleft-type using Cox regression analysis

	HR	95% CI
<b>All</b>		
Sibling with		
Cleft lip and palate	1.3	0.7-2.7
Cleft palate	2.3	1.3-4.2
	P-value 0.007	

HR: Hazard rate

# Results



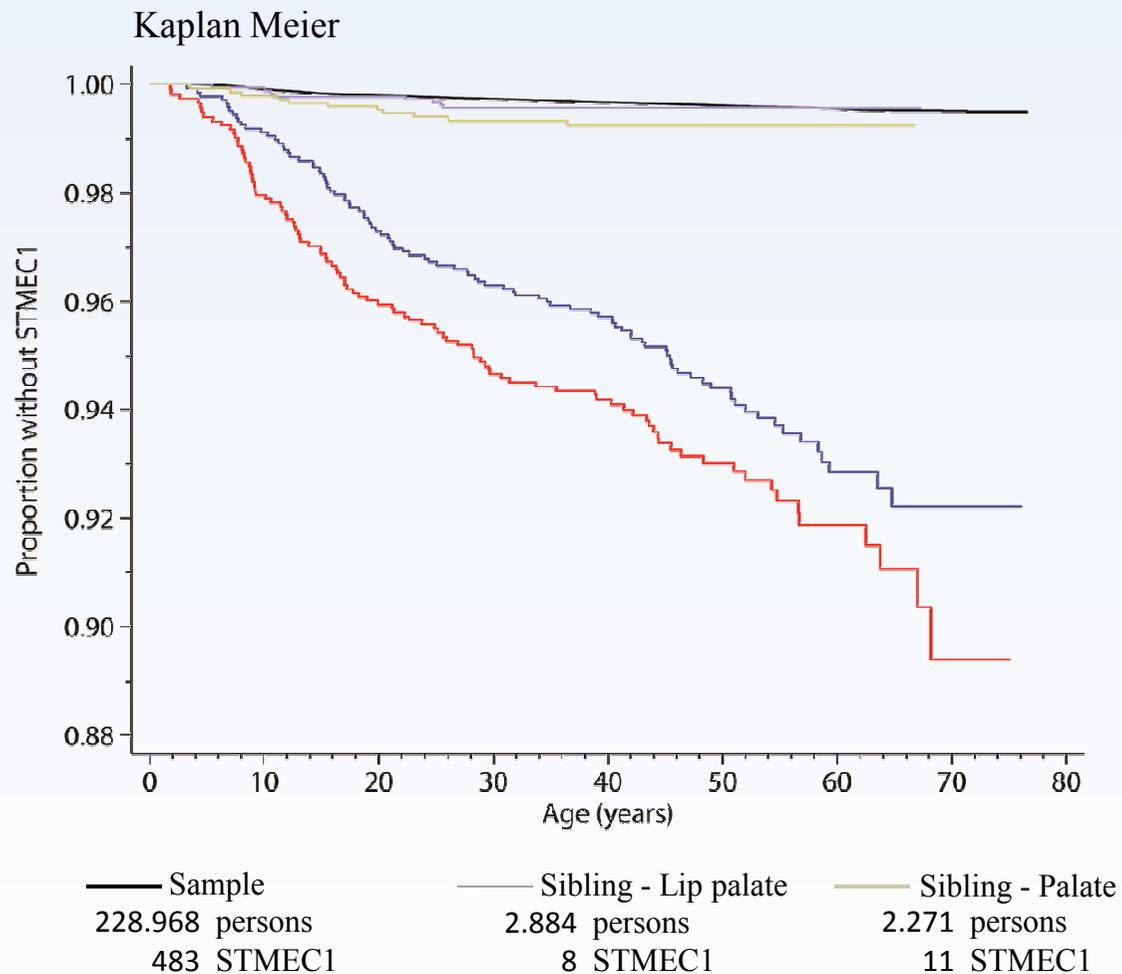
HR for STMEC1 by cleft-type using Cox regression analysis

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<b>All</b>		
Sibling with Cleft lip and palate	1.3	0.7-2.7
Cleft palate	2.3	1.3-4.2
		P-value 0.007
<b>Children</b>		
Sibling with Cleft lip and palate	1.3	0.5-3.4
Cleft palate	2.3	1.02-5.2
		P-value 0.046

STMEC1: First-time surgically treated middle ear cholesteatoma

HR: Hazard rate

# Results



STMEC1: First-time surgically treated middle ear cholesteatoma

HR for STMEC1 by cleft-type using Cox regression analysis

	HR	95% CI
<b>All</b>		
Sibling with Cleft lip and palate	1.3	0.7-2.7
Cleft palate	2.3	1.3-4.2
		P-value 0.007
<b>Children</b>		
Sibling with Cleft lip and palate	1.3	0.5-3.4
Cleft palate	2.3	1.02-5.2
		P-value 0.046
<b>Adults</b>		
Sibling with Cleft lip and palate	1.4	0.5-3.7
Cleft palate	2.3	0.9-5.6
		P-value 0.068

HR: Hazard rate

# Summing up

~ 8,000 patients with orofacial cleft

~ 200,000 sample persons without cleft

Additional number corresponding siblings

Cleft palate: 20-fold risk of cholesteatoma

Cleft lip: as background population

Cleft lip and palate: Less than cleft palate

Cumulative proportion with surgically treated cholesteatoma at 16 years of age:

3.3% in cleft palate

2.0% in cleft lip and palate

Siblings to cleft palate: 2-fold risk of cholesteatoma