

## The middle ear of cleft palate patients in their early teens: a literature study and preliminary file study

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**Abstract.** *The middle ear of cleft palate patients in their early teens: a literature study and preliminary file study.*  
**Objectives:** Middle ear disease is a frequent problem in young children with cleft palate (CP). Less is known about otological status in the adolescent CP population. The aim of this study is to provide an overview of current knowledge in the literature concerning the aetiology of middle ear disease in CP patients and the middle ear status of older children with CP, and to compare the situation in our centre to this background through an assessment of the otological status of patients in our CP population.

**Material and methods:** A literature review was conducted to summarise current knowledge about middle ear status in CP patients. A retrospective analysis was performed of the medical records of 20 CP patients between the ages of 10 and 15 who were treated and followed at the University Hospitals Leuven. The available otological data, otoscopic findings, information about hearing and surgery performed was collected for each patient when aged three and above ten years.

**Results:** Current knowledge indicates that middle ear function improves as CP patients get older. In our study, otoscopic appearance was acceptable in 63% of ears of children aged over ten years of age. Otitis media with effusion (OME) was far less frequent above this age than around the age of 3, with a decrease from 50% to 13% of all ears. Above the age of 10, tympanic perforations were present in 13% of ears and retraction of the tympanic membrane in 23%.

**Conclusions:** Despite a very high incidence of OME in young CP patients, the long-term otological prognosis is not necessarily sinister. A favourable natural evolution, diligent otological follow-up, early diagnosis and treatment of middle ear disease with the use of tympanostomy tubes are the likely contributors to the acceptable otological result in older CP patients.

### Introduction

High incidences of OME, negative middle ear pressures and conductive hearing loss in infants with CP have long been reported.<sup>1-5</sup> The incidence of OME decreases sharply in the general population above the age of 6 years.<sup>1</sup> Less is known about the prevalence and natural course of middle ear problems in older children with CP.

The aim of this study is therefore double: first to provide an overview of the current knowledge in the literature about the aetiology of middle ear disease in CP patients and the middle ear status of older children with CP. Secondly, this knowledge is compared to the situation in the patients treated at the University

Hospitals Leuven through a retrospective analysis of the medical records of 20 randomly selected CP patients between the ages of 10 and 15 years.

### PART 1 – Literature review

#### The aetiology of middle ear disease in cleft palate patients

Eustachian tube (ET) dysfunction is the primary cause of middle ear disease in CP patients.

Most authors describe a functional obstruction, a failure in the normal active opening mechanism of the ET, rather than an anatomical, mechanical obstruction. Deficient dilation of the ET because of anatomical and struc-

tural abnormalities of the tubal cartilage and the paratubal musculature as well as a hypercompliant tube may be to blame for this.<sup>2,6,7</sup>

The tubal cartilage of children below the age of 7 contains a higher density of chondrocytes compared to adults. The difference is even more pronounced in children with CP, although not to a statistically significant extent. This reflects cartilage immaturity and possibly explains the greater compliancy of the ET.<sup>6</sup> The hinge portion of the ET, the area between the lateral and medial lamina, has been shown to contain less elastine in CP children than in normal children. Reduced elasticity in the hinge portion of the ET leads to easier collapse and impaired return to the normal

position of the cartilage after dilation of the ET.<sup>8</sup>

The tensor veli palatini muscle (TVP) is the most important and necessary dilator of the ET. The muscle is hypoplastic and its insertion into the lateral lamina of the tubal cartilage is abnormal in CP patients, probably leading to the reduced transmission of the dilating force of the TVP.<sup>2,4,5</sup> The transversal orientation and fan-shaped insertion into the aponeurosis palatina of the levator veli palatini muscle is lost in CP patients. The muscle can only contract isometrically, and the increase in diameter is therefore less. As a result, the support it provides for dilation by elevating the media lamina of the ET is disrupted.<sup>9</sup>

A closing failure of the ET has been found in a high percentage of children with middle ear effusion with and without CP. The tube is incapable of withstanding physiological pressure variations in the nasopharynx such as those that occur during sniffing or crying. Passive opening of the tube allows air to be evacuated directly from the middle ear. This tends to lock the tube. Additional functional obstruction in the CP population leads to an inability to evacuate the resulting negative pressure and therefore to middle ear problems. The elastic properties of the ET and its surrounding tissue are probably to blame for the deficient closing capacity of the ET.<sup>10,11</sup>

Abnormalities of the skull base may also affect the otological status of CP patients.<sup>2,5,12</sup> The muscle vectors on the tube are affected, reducing the effectiveness of the dilation. A more horizontally oriented ET allows an easier retrograde flow of secretions and pathogens from the nasopharynx

to the middle ear. A shorter ET will also probably afford less protection against retrograde flow.

Children with CP may be more susceptible to middle ear infections.<sup>13,14</sup> The easier retrograde flow through the ET is especially unfortunate in CP patients before palatal repair because of the free reflux of feeds, secretions and pathogens from the oral to the nasal cavity. Enlarged adenoids may play a role. However, adenoid removal is generally contra-indicated because they help to seal off the velopharyngeal sphincter area.<sup>5</sup> Because of feeding difficulties, CP patients are often not breastfed, denying them the protective qualities of antimicrobial components in human milk.<sup>13</sup>

#### **Current knowledge about the otological status of CP patients in their early teens**

Analysing the available data about the otological status of older children with CP is no easy task as different researchers employ different definitions of what constitutes hearing loss, abnormal otoscopy and other important parameters. The composition of study populations varies widely and the populations are often split into subgroups in different ways, for instance depending on how cleft types are categorised. Once split into categories, many studies have small populations and therefore low-powered conclusions. It is of interest to note that the frequency of occurrence of OME in isolated cleft lip patients is approximately the same as in their age-matched counterparts.<sup>1,5</sup> The information provided here therefore relates only to CP patients.

In CP patients, there is an improvement in the parameters of

middle ear function with increasing age,<sup>1,7,15-17</sup> paralleling ET function improvements. ET function eventually recovers in the majority of cleft plate patients.<sup>7</sup> In a retrospective study by Smith *et al.*<sup>17</sup> of 81 CP patients, 85 ears were in follow-up for at least 6 years after palatoplasty. By the age of 12 years, 79% of these ears recovered, attaining normal ET function, defined as the presence of a type A tympanogram (Figure 1). Before palate repair or tympanostomy tube placements, ET dysfunction was found in 100% of ears. Palatoplasty was performed at a mean age of 19.5 months. The average time to recovery of ET function was 6.0 years (range 12 months to 10.3 years) after palatoplasty, at a mean absolute age of 7.9 years (range 1.5 to 17.3 years).<sup>17</sup> Between the ages of 10 and 15, normal middle ear pressures are reported in around 50 to 80% of CP patients.<sup>16,18-20</sup>

As can be expected from this ET function improvement, hearing also improves with age.<sup>15,16,21</sup> Handzic-Cuk *et al.*<sup>15</sup> used tonal audiometry to evaluate hearing levels in 243 CP patients between the ages of 1 and 34 (median age of 6 years). An age-related decrease in the prevalence of hearing loss at speech frequencies was found. A hearing loss of more than 20 dB was found in 51% of ears in the age group of 1-3-year-olds and in 16 and 18% for the age categories 10-12 and 13-15 years respectively (Figure 2).<sup>15</sup> The reported incidences of permanent, long-term hearing loss range from 0% to 90%, with an average of around 50%.<sup>2,4,19</sup> Many studies date from a time when tympanostomy tubes were not routinely used. More recent studies indicate that there is no longer necessarily such

**Eustachian Tube function**

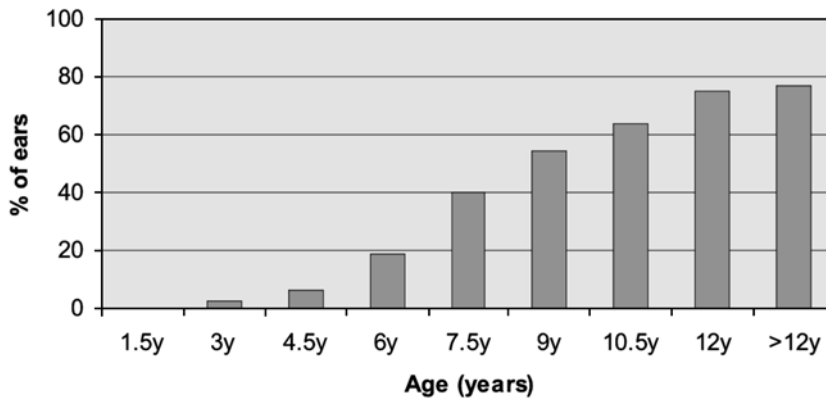


Figure 1

Percentage of ears with normal Eustachian tube function according to age. Note the gradual increase of ears with normal tubal function over time.<sup>17</sup>

**Hearing levels according to age**

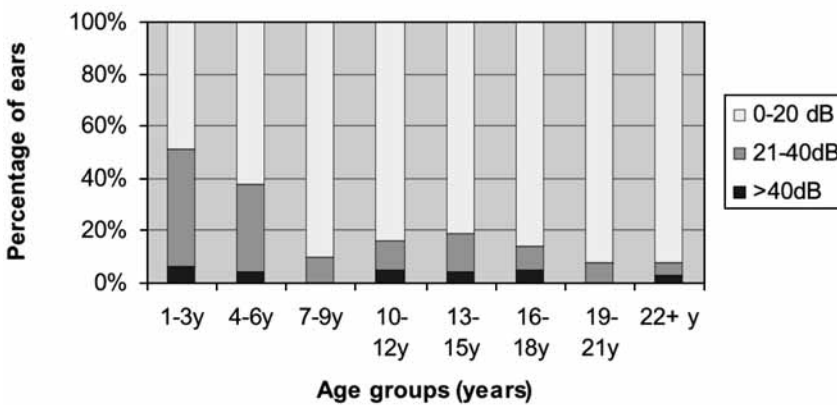


Figure 2

Hearing levels according to age.<sup>15</sup>

Number of ears per age group: 1-3y: 108, 4-6y: 138, 7-9y: 78, 10-12y: 40, 13-15y: 31, 16-18y: 23, 19-21y: 28, + 22y: 40.

a high incidence of middle ear problems.<sup>17</sup> Hearing loss is reported in 16 to 24% of CP children around and above the age of 10, with an average of about 20%.<sup>15,18,20</sup> As expected, this is usually a conductive hearing loss.

Improved ET function is also reflected in improving otoscopic findings with age.<sup>16</sup> Møller<sup>16</sup> conducted a prospective study of 149 CP patients between the ages

of 1 month and 20 years. The results for otoscopy are shown in Table 1.<sup>17</sup>

Studies mention normal otoscopy in around half of all CP patients above the age of 10. Tympanosclerosis is reported in up to 20% of patients and often regarded as an abnormal otoscopic appearance even though it is generally associated with very little hearing loss and normal middle

ear pressures. It is also not considered to be a risk factor for progression to chronic otitis media.<sup>3,14,18</sup> In older children with CP the incidence of OME is reported to be 10% or less, with figures as low as 2%. Perforations are reported in around 5% and tympanic retractions of different degrees in around 20% of cases.<sup>3,16,18,20</sup>

Abnormal otoscopic findings, middle ear pressure and hearing thresholds are statistically related.<sup>3,16,20,22</sup> The incidence of middle ear problems appears to be independent of the severity or type of CP.<sup>3,7,14,19</sup>

There is no universal agreement about the effect of palate repair on otological outcome. We know that the middle ear status of CP patients improves with age, with an increased tendency towards normalisation from the age of 10 upward. This improvement continues for many years after palate repair and there is no sudden improvement shortly after the repair. Evidence suggesting that the age or technique of palate repair are influential is lacking, as are control groups who have not undergone palate repair, for obvious reasons.<sup>14,17,19,23</sup>

The protracted and variable course of the improvement in middle ear status suggests that there are several influencing factors at play, such as age-related craniofacial growth, changes in ET function and a reduced frequency of upper respiratory infections.<sup>5,14,17</sup>

Opinions vary about the use of tympanostomy tubes in the CP population. Many authors advocate their aggressive use, with routine placements at times of surgery and intensive follow-up with replacement of tympanostomy tubes when OME is present.

Table 1

Otoscopic findings in a study by Møller.<sup>16</sup> The figures indicate the percentage of ears per age group. Normal otoscopy includes slight scarring of the tympanic membrane, with normal tympanic membrane mobility and an air-filled middle ear. Atelectasis of the eardrum was defined as retraction to the incudostapedial joint or to the promontory. OMA = acute otitis media

Age (years)	n	Normal otoscopy	Tympano-sclerosis	Atelectasis	Perforation	OME	OMA
≤5	124	12	2.4	0	0.0081	83	0.016
7-10	56	41	23	8.9	7.1	20	0
11-14	32	34	31	2.2	9.4	3.1	0
15-20	152	70	13	7.9	5.3	2.6	0.0066

The aim is to achieve immediate improvements in hearing and to avoid possible long-term effects on speech and language development, as well as to prevent persistent OME and its possible complications.<sup>14,18,24</sup> Others advocate a more conservative approach. They point out that long-term hearing thresholds, and speech and language outcomes, do not seem to be better in groups undergoing routine placements of tympanostomy tubes. They also fear the possible otological complications of tube placements such as tympanosclerosis and perforations.<sup>14,20,25</sup> Many authors mention the fact that there is a positive correlation between abnormal otoscopic appearance and the number of tympanostomy tube placements in the past. It is important to realise that this can be interpreted in different ways. Both a high number of tympanostomy placements and otoscopic abnormalities can be seen as a reflection of greater underlying middle ear disease and ET dysfunction which might have led to even greater long-term problems had tympanostomy tubes not been placed. On the other hand, some researchers believe that tympanostomy tubes are the cause of the abnormal otoscopic findings and therefore argue against their use.

## PART 2 – The situation in the University Hospitals Leuven: retrospective analysis

### Materials and methods

The material for this study consisted of 20 medical files selected at random from the list of CP patients between the ages of 10 and 15 under treatment and in follow-up at the University Hospitals Leuven. Patients with syndromic forms of CP, isolated cleft lip and sensory deafness were excluded. A retrospective analysis of these files was made. Six patients had an isolated CP, 12 a unilateral, and 2 a bilateral cleft lip and palate. There were 11 boys and 9 girls in the study. The mean age was 13 years and 4 months at the last follow-up appointment; the youngest patient was 11 years and 1 month and the eldest 15 years and 3 months. The available otological data, otoscopic findings, subjective comments on hearing and audiometry results were collected and summarised for each patient at the age of 3 and above the age of 10. All surgical procedures, including tympanostomy tube placements and the date and the age at which they were performed on the patients, were noted. Lip closure was performed according to Millards and

Tennison at a mean age of 5 months. Closure of the soft palate took place at a mean age of 1 year and 2 months, using the supraperiosteal retroposition technique described elsewhere in this report by Vander Poorten *et al.*<sup>26</sup> in one surgical step for CP-only patients with a hard palate cleft measuring up to 1 cm, in two surgical steps for CP up to the foramen incisive or for cleft lip and palate patients. In the latter patients, closure of the hard palate was performed at a mean age of 6 years and 2 months.

Audiometry performed in our hospital was considered to be normal when the pure tone average (PTA, mean hearing threshold at 500, 1000 and 2000 Hz, Fletcher index) was 20 decibels or less. Such audiometry was performed when the medical testing circuit active in schools referred children, or when there were subjective complaints about hearing. In Belgium, all children are subjected to compulsory medical monitoring at school at two-year intervals between the ages of 4 and 15, with hearing tests at the ages of 5, 11 and 15. When tests suggest abnormalities, the child is referred for further testing, and we proceed with audiometry. If school hearing tests are normal and if there are no subjective complaints or otoscopic

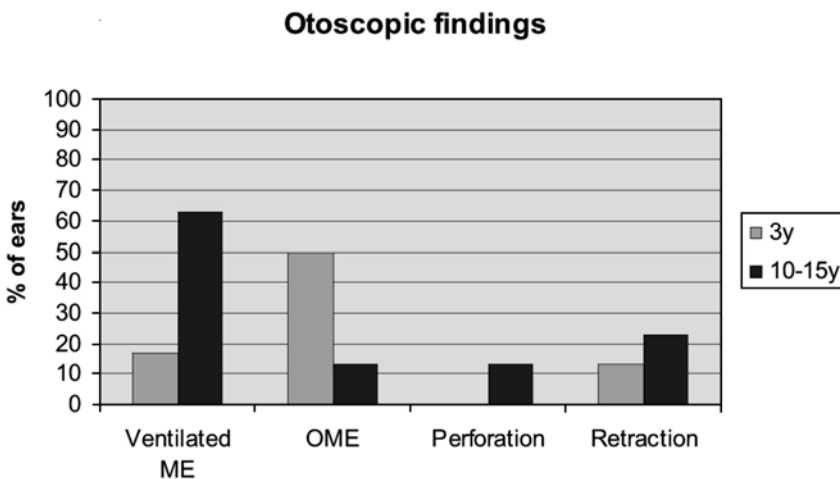


Figure 3  
Otosopic findings. TM = tympanic membrane

abnormalities, audiometry is not usually repeated. A hearing loss is considered to be present if the PTA exceeds 20 decibels.

**Results**

*Otосcopy (Figure 3)*

At the age of three, a normal tympanic membrane was seen bilaterally in 3 patients and unilaterally in 3 patients. Between the ages of 10 and 15, half of all patients had a normal tympanic membrane bilaterally and 1 unilaterally at some point during follow-up. At the age of three, tympanosclerosis was seen only in one ear. By the time the patients were between 10 and 15, 12 patients had tympanosclerosis, 10 bilaterally and 2 unilaterally. In 8 patients this was the only otoscopic abnormality ever seen between the ages of 10 and 15, in both ears for 7 patients and unilaterally for 1. In total, 63% (25 ears) had a normal tympanic membrane or only tympanosclerosis and a ventilated middle ear throughout follow-up between the ages of 10 and 15.

Three different pathological otoscopic findings were men-

tioned in the files: OME, tympanic membrane perforations and retraction of the tympanic membrane. OME was far less frequent above the age of 10 than at around the age of 3. There was a decrease from 50% at the age of 3 (8 patients bilaterally, 4 patients unilaterally) to 13% between 10 and 15 (1 patient bilaterally, 3 patients unilaterally). At the age of 3 no tympanic membrane perforations were seen. Above the age of 10, 13% of all ears had a perforated tympanic membrane (5 patients, always unilaterally). During the course of follow-up only 1 perforation healed spontaneously, 3 were present at the last consultation, and 1 was not reassessed because of cerumen. Tympanic retraction was seen in 13% of ears (1 patient bilaterally, 3 patients unilaterally) at the age of 3. Above the age of 10 this percentage was 23% (3 patients bilaterally, 3 patients unilaterally).

*Hearing above the age of 10*

Audiometry was obtained in our hospital for 9 patients above the age of 10. One patient had bilateral hearing loss, an additional 5 had

only unilateral hearing loss and 3 patients had a normal audiogram bilaterally. The remaining 11 patients did not have subjective complaints and did not have a referral note following systematic school-based hearing monitoring. In 8 of these patients the sense of hearing was noted to be subjectively good. No information about the sense of hearing, subjective or objective, was at hand for 3 patients.

*Tympanostomy tubes*

The mean number of tympanostomy tubes per ear for all ears was 2.9. Per otoscopic category above the age of 10 this number was 2.6 for ears with a normal tympanic membrane, 2.3 for ears with tympanosclerosis, 2.0 for ears with tympanic perforations, 5.6 for ears with OME and 4.2 for ears with tympanic retraction.

**Discussion**

A normal otoscopy became far more frequent as our CP patients got older. Tympanosclerosis was seen more often at older ages. This makes sense since it is an acquired condition. As mentioned above, tympanosclerosis is generally associated with little or no hearing loss and normal middle ear pressures, so we see it as an acceptable, non-pathological finding.<sup>15</sup> This means that, in total, 63% of ears in our 20 patients have an acceptable otoscopic appearance. This corresponds well with findings in the literature of normal otoscopy in about 50% and tympanosclerosis in around 20% of cases.<sup>3,16,18,20</sup>

OME was far less frequent above the age of 10 than at around the age of 3. There was a decrease

from 50% to 13% for all ears. These findings are similar to findings in the literature, where the spectacular decrease in the incidence of OME to around 10% or less is related to a better ET function and normal middle ear pressures.<sup>16,18,20</sup>

Thirteen per cent of all ears above the age of 10 had a tympanic perforation. It seems that, when present, perforations tend to persist. The percentage here is slightly higher than figures of around 5% mentioned in the literature. The occurrence of retraction in 23% of ears is similar to percentages of around 20% stated in the literature.<sup>16,18,20</sup>

In our study a hearing loss was present in 21% of ears for which information about the sense of hearing was available. This matches the figures of around 20% mentioned in the literature.<sup>15,18,20</sup>

The low average number of tympanostomy tube placements in ears with tympanosclerosis and tympanic perforations indicates no apparent correlation between these otoscopic findings and tympanostomy tube placements. Tympanostomy tubes therefore do not necessarily seem to be the cause of these otoscopic abnormalities. The high average number of tympanostomy tube placements in ears with OME and tympanic retractions, on the other hand, seems to be a reflection of greater underlying middle ear disease in these ears, as it is very improbable that tympanostomy tubes are the cause of these abnormalities.

The number of patients included in this study is small but serves its purpose as a sample survey to provide a general idea of the otological situation in the adolescent CP population at our centre.

Audiometric or tympanometric findings were not routinely obtained. These tests were mainly performed on patients with subjective or objective suspicions of middle ear disease. A prospective study with routine otoscopy, audiometry and tympanometry in all patients would obviously yield a statistically more precise result and is an objective for the future.

### Conclusion

Despite a very high incidence of OME in young CP patients and multiple tympanostomy tube placements, the long-term otological prognosis is not necessarily sinister. The majority of ears have a tympanic membrane in a good condition and normal hearing after the age of 10 is reached. A certain favourable natural evolution, without any clear temporal link to the moment of palate repair or the number of tympanostomy tube placements, seems to contribute to the otological result in older CP patients. We feel that diligent otological follow-up, with early diagnosis and treatment of middle ear-disease, mainly by using tympanostomy tubes, is the key to minimising long-term hearing problems.

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