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Pediatric Endoscopic Cholesteatoma Surgery

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Abstract

Objectives. (1) To describe and review a single center's pediatric endoscopic cholesteatoma experience, including surgical and audiologic outcomes. (2) To assess the most common locations of residual cholesteatoma following endoscopic removal.

Study Design. Case series with chart review.

Setting. Tertiary otologic referral center.

Subjects. Patients <19 years of age who underwent cholesteatoma removal with either endoscopic or microscopic visualization.

Methods. In a comparison of patients who underwent total endoscopic ear surgery (TEES), combined endoscopic-microscopic surgery, or microscopic surgery, analyzed outcomes included locations and incidence of recurrent and residual cholesteatoma, complications, and audiometric testing.

Results. Sixty-six patients (mean age, 10.9 years; range, 4-18 years; 43.4% female) with 76 ears met inclusion criteria. The average overall follow-up was 18.8 months (range, 6.7-48.3). Forty-seven (61.8%) ears underwent microscopic removal of cholesteatoma; 29 (38.1%) ears underwent combined endoscopic-microscopic removal; and 8 (10.5%) ears underwent TEES removal. Significantly more mastoidectomies were completed in microscopic cases as compared with endoscopic cases (P = .049). Though second-look procedures occurred in 15 (51.7%) endoscopic cases and 10 (21.3%) microscopic cases (P = .006), the rate of residual disease was 20.0% and 40.0% in endoscopic and microscopic cases, respectively (P = .38). When controlling for preoperative hearing, only the air-bone gap for TEES demonstrated significant improvement (P = .009). No complications were noted.

Conclusion. The present report describes our experience with pediatric endoscopic cholesteatoma surgery, demonstrating similar hearing outcomes, rates of recurrence and residual disease, and complication rates as compared with traditional microscopic techniques.

Keywords

pediatric, cholesteatoma, ossiculoplasty, tensor fold, endoscopic ear surgery

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The application of endoscopes to visualize the middle ear was first described >4 decades ago, but recent technological advances have led to greater use of endoscopes in middle ear surgery.[1] Increasingly, endoscopes are being used to diagnose and treat middle ear pathology.[1-3] In select cases, endoscopes have been used as the only means of visualization for the entirety of a procedure, without assistance from a microscope.[4] As applications for endoscopic ear surgery evolve, the challenges of operating with 1 hand, additional training, loss of depth perception and binocular vision, repeated endoscope cleaning, and cost remain to be definitively addressed.[5]

Nonetheless, some evidence suggests that endoscopes could improve outcomes in middle ear cholesteatoma surgery. In the absence of endoscopic visualization, studies have reported residual cholesteatomas rates of 10% to 43% during second-look procedures following canal wall up tympanomastoidectomies.[6-8] Reinforcing this finding, endoscopic reports have noted that adjunctive use of the endoscope during primary cholesteatoma surgery identifies otherwise unseen disease in about 40% of patients, with reports ranging from 16% to 76%.[9,10] In 1993, Thomassin et al compared outcomes with and without the endoscope in patients undergoing canal wall up tympanomastoidectomies

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for middle ear cholesteatomas.[3] With all patients undergoing second-look operations, they found that cholesteatomas were noted in 47% of cases when only the microscope was used, as compared with 6% in cases when the original surgery was supplemented with the endoscope.[3] Reduced residual disease rates with endoscopic assistance have been reproduced by other studies.[11-13]

Most of the early endoscopic ear surgery reports failed to differentiate children and adult outcomes. With various reports over the years suggesting increased incidence of residual disease in children, few studies exist that assess only endoscopic ear surgery in children.[8,14-17] Thus, we sought to review our pediatric endoscopic cholesteatoma surgery experience, analyzing surgical and audiologic outcomes.

Methods

Following institutional review board approval (141631), we conducted a retrospective chart review of all cholesteatoma operations in children aged $\leq$18 years between January 2012 and June 2015. All subjects were included if they underwent primary surgery, with or without the use of endoscopes for cholesteatoma removal, and all subsequent operations. Exclusion criteria were applied to remove subjects $>$18 years of age, those who had follow-up $<$6 months, and those who underwent canal wall down tympanomastoidectomies. Subjects were divided into 2 groups: group 1 consisted of subjects who underwent cholesteatoma surgery with use of a microscope, while group 2 comprised subjects who underwent cholesteatoma surgery with use of an endoscope at any point during the procedure. This latter group was further divided into 2 subgroups: combined endoscopic-microscopic group and total endoscopic ear surgery (TEES) group. ''Combined'' cases consisted of endoscopic removal of disease, with the microscope used to drill the mastoid if contracted and/or if disease extended beyond the antrum. TEES cases consisted of transcanal cases completed with exclusive endoscopic cholesteatoma removal without microscopic assistance.

Preoperative variables assessed included age at the time of initial surgery, sex, pure tone average (PTA), air-bone gap (ABG), and word recognition (WR) score, when recorded. Intraoperative variables of interest included cholesteatoma location, tensor fold identification and status, ossicular chain involvement and continuity, ossicular chain reconstruction, whether the mastoid was drilled, intraoperative complications, length and reason for hospital stay, and documentation whether another procedure was planned at the time of primary surgery. Postoperative variables reviewed included length of follow-up, presence of residual or recurrent disease, PTA, ABG, WR score, and complications. Each subject's postoperative audiometric testing was conducted after ossicular reconstructions were performed (either primary or secondary procedure), when the ossicular chain was eroded, or after a primary procedure, when the ossicular chain was intact.

As outlined by the Hearing Committee of the American Academy of Otolaryngology—Head and Neck Surgery, the PTA was obtained by averaging the audiometric thresholds at 0.5, 1, 2, and 3 kHz, while calculation of an ABG was made by subtracting the bone conduction thresholds from the air conduction thresholds at 0.5, 1, 2, and 4 kHz and averaging the differences.[18]

Complete cholesteatoma removal with a TEES approach was attempted if preoperative computed tomography temporal bone imaging was available and not suggestive of middle ear disease extending past the dome of the lateral semicircular canal. If disease extended past the lateral semicircular canal on preoperative imaging or if disease was unable to be removed completely with use of endoscopes and angled instruments, a postauricular incision was made approximately 0.5 cm posterior to the auricular sulcus, followed by periosteal cuts and completion of a mastoidectomy.

To simplify the reporting of cholesteatoma locations, the middle ear was divided into the following component parts: retrotympanum (including the sinus tympani), hypotympanum, epitympanum, mesotympanum, and the protympanum. Residual cholesteatomas were defined as cholesteatoma identified during the second look under an intact tympanic membrane, while recurrent cholesteatomas were defined as non self-cleaning tympanic membrane retractions following surgery.

Statistical analysis was performed with SPSS 21 (IBM, Armonk, New York). Continuous features were summarized with means, medians, and ranges; categorical features were summarized with frequency counts and percentages. The number of subjects who underwent second-look procedures was used as the denominator in each group to calculate residual rates of disease. Student's *t* test was used to compare means between 2 groups, with all tests 2-sided and $P < .05$ considered statistically significant, while a 1-way analysis of variance was used to compare means with $\geq$3 groups. An analysis of covariance was used to analyze postoperative audiometric outcomes, controlling for preoperative differences. Fisher's exact test was used to compare nominal variables. Pearson's correlation calculated the correlation between subject age and presence of cholesteatoma in $>$1 site.

Results

Sixty-six subjects met inclusion criteria, 10 of which had bilateral surgical interventions. Therefore, 76 cases were identified: 29 in group 1, the endoscope group, and 47 in group 2, the microscope group. When the endoscope group was subdivided, 8 cases were fully endoscopic (TEES), and 21 were combined endoscopic-microscopic. The mean overall age of all participants was 10.9 years (range, 4-18), with 43.4% female. There were 38 right ears (50.0%) in total. The mean follow-up time in the endoscopic group was 14.2 months (range, 6.7-26.2), 15.3 months (range, 6.7-26.2) in the TEES group, and 21.7 months (range, 6.8-48.3) in the microscopic group, with a mean overall follow-up time of 18.8 months (**Table 1**). Preoperative audiometric results are

Table 1. Demographics.

	Microscope Group		Endoscope Group		Combined		TEES		Total	
	n	%	n	%	n	%	n	%	n	%
Sex										
Male	25	53	18	62	14	67	4	50	43	57
Female	22	47	11	38	7	33	4	50	33	43
Ear										
Right	21	45	17	59	13	62	4	50	38	50
Left	26	55	12	41	8	38	4	50	38	50
Age groups, y										
<5	1	2	0	0	0	0	0	0	1	1
6-10	24	51	17	59	13	62	4	50	41	54
11-15	16	34	9	31	7	33	2	25	25	33
16+	6	13	3	10	1	5	2	25	9	12
Total	47	62	29	38	21	72	8	28	76	100
Mean (range)										
Age, y	10.7 (4-18)		11.1 (5-17)		11.0 (6-16)		11.6 (5-17)		10.9 (4-18)	
Follow-up (range), mo	21.7 (7-48)		14.2 (7-26)		13.8 (7-24)		15.3 (7-26)		18.8 (7-48)	

Abbreviation: TEES, total endoscopic ear surgery.

Table 2. Pre- and Postoperative Audiometric Results.[a]

	Preoperative Mean Values			Postoperative Mean Values					
	PTA, dB HL	ABG, dB HL	WR, %	PTA, dB HL	*P* Value	ABG, dB HL	*P* Value	WR, %	*P* Value
Microscope	33.2	25.2	96.5	26.6	.054	21.7	.090	98.0	.182
Endoscope	31.8	29.7	98.1	22.6	**.007**	21.5	.409	98.4	.756
P value	.60	.39	.28	.20		.75		.85	
Combined	33.9	31.0	98.5	24.3	**.047**	24.6	.123	98.0	.816
TEES	27.4	27.0	97.5	16.7	**.011**	14.6	**.009**	100.0	.423
P value	.30	.50	.50	.28		.33		.66	

Abbreviations: ABG, air-bone gap; PTA, pure tone average; TEES, total endoscopic ear surgery; WR, word recognition.
[a]Bold font indicates $P < .05$.

listed in **Table 2**. All groups had similar preoperative audiometric values with no significant differences.

Intraoperative Findings and Ossicular Chain Involvement

Cholesteatoma was identified in all locations in both groups: protympanum, hypotympanum, mesotympanum, epitympanum, retrotympanum, and mastoid. Overall, 72.4% cases had cholesteatoma involving >1 site, likewise in 75.9% and 70.2% of endoscopic and microscopic cases, respectively (*P* = .79). **Table 3** lists the cholesteatoma locations, along with other intraoperative findings. No complications were reported.

More than two-thirds of all endoscopic procedures also had a mastoidectomy completed, which was significantly less than in microscopic cases (*P* = .049). Regarding ossicular disease, 55.3% of all ears had ossicular cholesteatoma involvement, with an equal proportion between the 2 main patient groups (**Table 3**). All endoscopic cases, as well as TEES cases, did not significantly preserve the ossicular chain any more than the microscope group.

While it was not documented in the microscope group and thus not included in the analysis, the tensor fold was identified in 16 subjects (55.2%) in all endoscope cases, which was obstructed in 12 (75.0%) subjects. Of the 12 subjects, 9 (75.0%) had at least some disease in the epitympanum. In the analysis of the status of the ossicular chain when the tensor fold was recorded as open, 75.0% had an intact ossicular chain, as opposed to 25.0% when the tensor fold was closed, which was not significant (*P* = .12).

In regard to operative time, microscopic cases lasted a mean of 134.6 minutes (range, 57-222), while all endoscopic cases lasted a mean of 247 minutes (range, 103-435); combined endoscopic-microscopic cases, a mean of 262 minutes (range, 143-435); and TEES cases, a mean of 208

Table 3. Intraoperative Findings.[a]

	Microscope		All Endoscopic		*P* Value	Combined Endoscopic		*P* Value	TEES		*P* Value
	n	%	n	%		n	%		n	%	
Sites											
Protympanum	0	0.0	1	3.4		0	0.0		1	12.5	
Hypotympanum	11	23.4	13	44.8		9	42.9		4	50.0	
Mesotympanum	36	76.6	24	82.8		16	76.2		8	100.0	
Epitympanum	31	66.0	15	51.7		11	52.4		4	50.0	
Retrotympanum	8	17.0	13	44.8		9	42.9		4	50.0	
Mastoidectomy	43	91.5	21	72.4	**.049**	21	100.0	.30	0	0.0	N/A
Ossicular chain status											
Intact	22	46.8	12	41.4		10	47.6		2	25.0	
Compromised	25	53.2	17	58.6		11	52.4		6	75.0	
Reconstructed	17	68.0	5	29.4	**.03**	2	18.2	**.01**	3	50.0	.64
PORP	9	52.9	4	80.0		1	50.0		3	100.0	
TORP	5	29.4	1	40.0		1	50.0		0	0.0	
Tensor fold status											
Open			4	25.0		3	27.3		1	20.0	
Closed			12	75.0		8	72.7		4	80.0	
Second looks	10	21.3	15	51.7	**.01**	12	62.0	.23	3	37.5	.18
Residual disease	4	40.0	3	20.0	.38	2	16.7	.35	1	33.3	N/A
Mean (range)											
Intraoperative time, mo	135 (57-222)		247 (103-435)		< **.001**	262 (143-435)		<**.001**	208 (103-286)		<**.001**

Abbreviation: N/A, not applicable; PORP, partial ossicular replacement prosthesis; TEES, total endoscopic ear surgery; TORP, total ossicular replacement prosthesis.
[a]Bold font indicates $P < .05$.

minutes (range, 103-286 minutes)—all of which were significantly different from microscopic cases (all $P < .001$; **Figure 1**).

Recurrent or Residual Disease

In the entire endoscopic group, a second-look procedure was performed in 15 subjects (51.7%); 1 (6.7%) required a third procedure. Of the 8 cases who underwent TEES, 3 (37.5%) required a second look: 1 for ossicular chain reconstruction and 1 each for inferior and superior retractions. In the microscope group, 10 (21.3%) subjects underwent second-look procedures, 2 (20.0%) of which required a third procedure. The microscope group underwent significantly fewer second-look procedures as compared with all endoscopic cases (P = .01), though there was no significant difference between second-look procedures for microscopic cases and TEES (P = .18).

In terms of the risk of recurrence, in the entire endoscope group, 3 (10.3%) subjects had recurrent disease, located in the epitympanum (n = 3), mesotympanum (n = 2), retrotympanum (n = 1), and hypotympanum (n = 1). In the microscope group, 4 (8.5%) subjects had recurrent disease, located in the epitympanum (n = 4), mesotympanum (n = 3), mastoid (n = 2), and the sinus tympani and facial recess in 1 subject each. These rates were not significantly different (P = .47).

In regard to residual disease, 3 (20.0%) subjects were noted to have residual disease in all endoscopic cases, involving the epitympanum (n = 1), sinus tympani (n = 1), and stapes capitulum (n = 1). In the microscope group, another 4 subjects (40.0%) had residual disease, involving the mesotympanum (n = 4), epitympanum (n = 2), and mastoid (n = 2). These rates were not significantly different (P = .38).

Five (17.2%) endoscopic subjects underwent ossicular chain reconstruction at the time of the primary surgery. Seventeen (36.1%) microscopic subjects underwent ossicular chain reconstruction during the primary surgery, which was not significantly different (P = .12).

Postoperative Hearing Results

Postoperative audiometric results are summarized in **Table 2**. All groups, except the microscopic group, demonstrated significant PTA improvements as compared with the preoperative condition. However, when compared with each other after controlling for the preoperative hearing differences, there were no statistical differences in PTA between groups. In addition, though the TEES group showed significant ABG improvement postoperatively (P = .009), when compared against the microscope and combined endoscopic groups, there were no statistical differences. When prostheses used for ossicular chain reconstruction were assessed, there was no

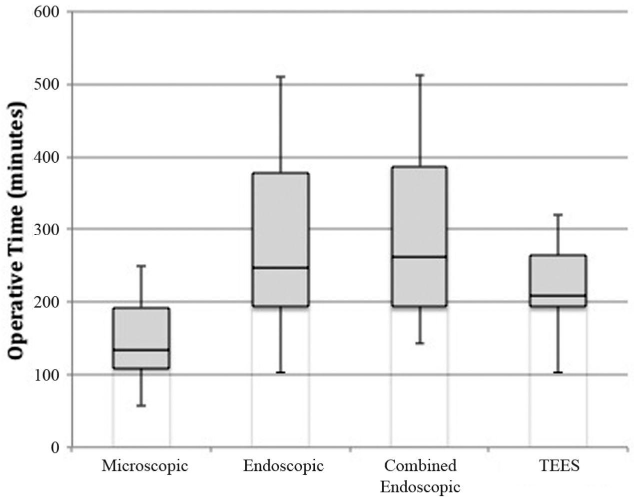

Figure 1. Operative times. TEES, total endoscopic ear surgery.

significant difference between the endoscope and microscope groups in terms of PTA, ABG, and WR, controlling for the prosthesis type. There were no long-term complications in either group.

Discussion

Several reports have demonstrated that the addition of endoscopes to cholesteatoma surgery can reduce the incidence of residual disease.[3,11,12,14,19-22] Several surgeons even avoid postauricular incisions, endoscopically removing cholesteatomas through the external canal and tympanic membrane.[4,6,23] Nonetheless, even with endoscopic assistance, residual disease is still encountered postoperatively. As with microscopic-only operations, the most common sites of residual disease following combined endoscope-microscope cases are the sinus tympani, facial recess, and undersurface of the scutum.[12,19]

It has been suggested that children are at a higher risk to develop residual disease as compared with adults.[17,24,25] Tarabichi first reported on his experience in treating 73 attic cholesteatomas with only endoscopic visualization, noting that only 6.8% required revision for recurrent or residual disease.[4] Interestingly, 2 of his 5 failures occurred in children <8 years of age.[4] Likewise, Barakate et al indicated that 3 of 7 children between the ages of 5 and 8 years had cholesteatoma noted at second-look procedures.[13] Though early endoscopic studies included patients of all ages, recent reports have begun focusing only on children.[14-16,26] Most recently, Marchioni et al compared cholesteatoma recurrence rates in pediatric patients who underwent TEES with pediatric patients who underwent canal wall up microscopic approaches without endoscopes, noting recurrence rates of 12.9% and 17.2%, respectively.[16] With a mean follow-up of 36 months, residual disease was present following 19.3% of TEES and 34.4% canal wall up cases, with no significant difference. Our series is similar to that of Marchioni et al, with recurrent rates of 10.0% in the endoscope group and 6.6% in the microscope group ($P = .709$) and with residual rates of 22.2% in endoscopic cases against 36.4% in microscopic cases ($P = .69$).[16]

We report on 76 consecutive pediatric cholesteatoma cases, 29 of which were conducted with endoscopic visualization. Both groups were noted to have similar cholesteatoma locations and preoperative audiologic measurements. During primary surgery, there were no significant differences between groups at preserving the ossicular chain, with rates better, if not comparable, to previous published pediatric endoscopic series.[4,6,11,16] But several differences were also appreciated. While mastoidectomies were significantly more likely to be performed in microscopic cases as compared with combined endoscopic cases, endoscopic cases, combined and TEES, lasted significantly longer, which we attribute to our early experience and anticipate this decreasing over time. Nonetheless, no significant differences in recurrence or residual rates were noted between microscopic or endoscopic cases, though we recognize that microscopic cases underwent significantly fewer second looks, which we attribute to surgeon experience and preference, as well as the desire of the endoscopic surgeons to honestly document their residual disease rate.

While there is controversy and debate regarding the role and existence of ventilation pathways within the middle ear, it is hypothesized by some that attic cholesteatomas can develop when the tensor fold is obstructed and the isthmus is blocked.[27] Thus, we sought to identify and analyze any relationship that the tensor fold may have with the presence of cholesteatoma within the epitympanum. The tensor fold status was observed in 55.2% of endoscopic cases, 75% of which noted complete closure. Of interest, 75% of subjects who had a closed tensor fold also possessed disease within the epitympanum. In assessment of ossicular chain integrity, 75% of subjects who had a closed tensor fold had ossicular erosion, as compared with 25% of subjects who had an eroded ossicular chain when the tensor fold was open ($P = .12$). Though suggestive, the existence of blocked ventilation pathways and the possible role that they may play should be explored in future prospective studies.

Audiometrically, within-group comparisons demonstrated statistical significant improvements in PTA in only the endoscope groups. Thus, that endoscopic cases underwent significantly more second-look procedures and significantly fewer primary ossicular chain reconstructions as compared with microscopic cases may suggest that endoscopic ossicular chain reconstruction during second-look procedures may lead to improved audiometric outcomes. However, after controlling for preoperative differences, there were no significant differences between groups with PTA, ABG, or WR score. While further studies should compare audiometric outcomes between microscopic and endoscopic techniques, possible theories that could contribute to improved hearing results include (1) stabilization of the tympanic membrane at the time of prosthesis placement during second looks, (2) stabilized middle ear mucosa allowing improved middle ear space aeration and drum mobility, (3) minimal elevation of the drum due to endoscopic use during second look, and (4) the ability to endoscopically observe the prosthesis coupling distally and medially until the tympanomeatal flap is returned to its natural position.

Despite our outcomes, we recognize the criticisms of endoscope ear surgery and believe that they are pertinent for discussion. Of significant note, most detractors complain that 1-handed surgery is a significant limitation, as concerns regarding the safety of removing disease from the stapes superstructure, footplate, and/or dehiscent facial nerve with 1 hand seems counterintuitive. Moreover, operating 1 handed prevents the ability to simultaneously dissect and suction the operative field. While further data regarding the safety of excessive heat dissipation from the endoscope tip is forthcoming, other critics have voiced concerns regarding the length of the operations.[28,29] Nonetheless, many admit that with increased familiarity with the equipment and setup, as well as manipulation within the ear, time is negligible between TEES and microscopic techniques.

There are several limitations to our study. As a retrospective review, we are beholden to the accuracy of the electronic medical record. Alternatively stated, failure to adequately document operative findings based on surgeon practice must be considered. Though a similarly dated consecutive patient sample acted as a control group, the paucity of second-look procedures with relative short follow-up in the both groups probably underestimates the actual incidence of residual and recurrent disease. In addition, a selection bias will always exist in TEES cases, as the endoscope can be used exclusively only in disease of limited extent. Furthermore, though we tried to control for cholesteatoma disease severity by demonstrating similar locations and ossicular chain integrity between endoscopic and microscopic groups, we recognize that this is not foolproof; thus, outcomes could also be due to differences in disease severity that we were unable to account for. In addition, not all microscopic cases can be performed completely endoscopically, since endoscopes cannot access disease extension to the mastoid; as such, the TEES group is by definition going to have cholesteatomas limited to the middle ear and antrum. Therefore, until a prospective study is completed comparing outcomes following microscopic versus endoscopic middle ear cholesteatoma removal, complete comparisons will be difficult.

Conclusion

Reviewing 76 pediatric cholesteatoma cases, we found a 10.3% recurrence rate and a 20.0% residual disease rate in TEES and endoscopically assisted cases. With previous evidence to suggest that pediatric cholesteatomas tend to have higher residual disease rates as compared with those of adults, our results demonstrate that our recurrence and residual disease rates in endoscopic cholesteatoma removal are not significantly different from those of the microscopic removal. Audiometrically, there were no significant PTA, ABG, and WR score differences between groups after controlling for preoperative differences.

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Author Contributions

Jacob B. Hunter, data collection, data analysis, data interpretation, drafting of manuscript, revising manuscript, final approval of manuscript; **M. Geraldine Zuniga**, data collection, data analysis, drafting of manuscript, revising manuscript, final approval of manuscript; **Alex D. Sweeney**, data interpretation, revising manuscript, final approval of manuscript; **Natalie M. Bertrand**, data collection, final approval of manuscript (the author reviewed, corrected, revised, and ultimately provided final approval of the manuscript); **George B. Wanna**, data interpretation, revising manuscript, final approval of manuscript; **David S. Haynes**, data interpretation, revising manuscript, final approval of manuscript; **Christopher T. Wootten**, data interpretation, revising manuscript, final approval of manuscript; **Alejandro Rivas**, data interpretation, revising manuscript, final approval of manuscript

Disclosures

Competing interests: George B. Wanna, consultant for MED-EL, Advanced Bionics, Cochlear, Grace Medical, and Oticon; David S. Haynes, consultant for MED-EL, Advanced Bionics, Cochlear, Grace Medical, Stryker, and Synthes; Alejandro Rivas, consultant for MED-EL, Advanced Bionics, Cochlear, Grace Medical, Stryker, and Olympus.

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