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Systematic Review

Outcomes and Complications in Superior Semicircular Canal Dehiscence Surgery: A Systematic Review

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Objective: Superior semicircular canal dehiscence (SSCD) represents a rare condition that may be associated to some particular symptoms as vertigo, autophony, and Tullio phenomenon. In those patients who present severe symptoms surgical treatment is required. Middle fossa craniotomy and transmastoid approaches are both described. Concerning repairing techniques, plugging and/or resurfacing are typically used to close the defect. Our aim was first to analyze the overall outcomes and complications of this surgery. Our second aim was to make a comparison between the different surgical modalities to investigate the eventual advantages and disadvantages.

Review Methods: A search through Ovid MEDLINE was organized in January 2015 to include all eligible articles. A statistical analysis of the obtained data was performed.

Results: Twenty studies comprising a total of 150 procedures were included. Four modalities of canal repair were described (plugging, capping, resurfacing, plugging with resurfacing). The overall rate of success resulted in 94% (95% confidence interval: 87%-97%). No statistically significant differences were observed among the different modalities of canal repair concerning both success rate and surgical complications. Differences observed between the middle fossa approach and transmastoid approach in terms of outcome were not statistically significant.

Conclusions: This review showed that surgical treatment for SSCD represents a safe option for those patients with severe symptoms of this condition. Differences observed in terms of success rate or complications were analyzed between the different surgical techniques with results that were not statistically significant.

Key Words: Superior semicircular canal, plugging, resurfacing, transmastoid approach, middle fossa approach.

Level of Evidence: NA

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INTRODUCTION

An opening in the thin bone margin of the superior semicircular canal creates a direct interface between the membranous canal and the overlying dura. This opening is also called the third window, in reference to the oval and round windows, the two physiological openings of the otic capsule.[1] Superior semicircular canal dehiscence (SSCD) syndrome is a condition characterized by the presence of sound- and/or pressure-induced vertigo or oscillopsia due to the dehiscence of bone overlying the superior semicircular canal.[2] Vestibular activation in response to acoustic stimulation is termed the Tullio phenomenon, whereas vestibular activation elicited by pressure changes in the external auditory canal or Valsalva maneuver is referred to as Hennebert's sign.[3] Hearing loss may also be associated with SSCD and is often characterized by a low-frequency air-bone gap with a bone-conduction threshold that can be supranormal (more than 0 dB).[4] Vestibular-evoked myogenic potential responses have been shown to be useful in evaluating patients suspected of having the symptoms and signs of SSCD.[5] However, the diagnosis can only be confirmed by high-resolution temporal bone computed tomography (CT).[6]

In some patients, the symptoms of SSCD can be controlled by avoiding provocative stimuli, but for patients who are debilitated by the vestibular and audiological manifestations, surgery may represent the only treatment possible. Resurfacing, capping, or plugging of the superior semicircular canal has been shown to be effective in achieving long-term resolution of vestibular symptoms and signs.[5] All these techniques have been reported to alleviate symptoms of superior canal dehiscence, in particular sound- and pressure-induced vertigo.

In plugging, the dehiscent canal is obliterated with bone paté or other materials. The goal of the resurfacing and capping technique is to cover the SSCD without occluding the canal. Bone, cartilage grafts, fascia, or hydroxyapatite

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cement are placed over the dehiscence to achieve a continuous separation between the membranous labyrinth and the dura.

Some authors have suggested that the plugging technique achieves superior results and symptomatic control as compared with the resurfacing technique.[7] Nevertheless, canal plugging theoretically carries a higher risk of damage to the vestibular and cochlear structures, because it is more invasive to the labyrinth.[8] Concerning the surgical approach, middle fossa and transmastoid routes represent two different possible ways to perform this surgery. The first operations were performed through the middle fossa approach,[9] because this approach offers a direct visualization of the dehiscence and the correct placement of the grafts is easier. However, over the last few years, because most ear surgeons are more familiar with the anatomy of the mastoid than the middle fossa, the transmastoid approach has become increasingly popular.[1]

So on the basis of the actual knowledge, numerous variants exist to surgically treat SSCD syndrome. For this reason the purpose of our study was to carefully investigate the outcomes and complications related to these different surgical techniques in those patients who have undergone surgery for SSCD.

MATERIALS AND METHODS

Search Methods for Identification of Studies

In January 2015, a computerized MEDLINE search was performed using the PubMed service of the U.S. National Library of Medicine and the following search string was run: "Semicircular Canals/abnormalities"[Mesh] OR "Semicircular Canals/diagnosis"[Mesh] OR "Semicircular Canals/injuries"[Mesh] OR "Semicircular Canals/pathology"[Mesh] OR "Semicircular Canals/radiography"[Mesh] OR "Semicircular Canals/surgery"[Mesh] OR "Semicircular Canals/therapy"[Mesh].

The initial search returned a total of 862 results. Abstracts and titles obtained were screened independently by two of the authors (F.M.G., M.R.), who subsequently met and discussed disagreements on citation inclusion.

Inclusion criteria for citations were articles dealing with superior semicircular canal surgery. Exclusion criteria were articles written in languages other than English and clear unrelated types of pathologies than superior semicircular canal dehiscence. Among the 862 articles, 31 met the initial inclusion criteria according to both authors (F.M.G., M.R.), so they were obtained and reviewed in detail by the same two authors, who met and discussed disagreements on article inclusion.

Inclusion criteria for full-text articles and single patients identified were: articles comprising cohorts of patients with a well-defined diagnosis of SSDC syndrome and articles showing precisely both outcomes and complications related to surgery. Exclusion criteria were articles focusing only on hearing outcomes, articles focusing only on vestibular outcomes, lack of sufficient information about surgical complications, lack of information defining exactly the performed surgical approach (transmastoid or middle fossa approach), lack of information defining exactly the repairing technique (plugging, resurfacing, or capping), articles including duplicate data, and review articles.

A further manual check was performed on the references included in the articles, and one additional study was identified that met the inclusion criteria through a review of references and a concurrent PubMed search. The final number of articles included in the present review was identified, and the main information was extracted and summarized.

Statistical Methods

We performed analysis for patients stratified by approach group. χ^2 test or Fisher exact test for significance were carried out to evaluate the differences. We performed univariate logistic regression for the surgical outcome variable, where we compared the event failure versus successful event of the operation. Univariate logistic regression models were used to determine the effect of the technique. The models did not reach statistical significance among the different techniques. The basis of odds ratio calculations was the plugging technique. A limitation in our calculation of odds that needs to be noted is the difference in the number of observations among the groups. The calculation ratios refer to the specific dataset with regard to the type of surgical intervention. An odds ratio of 1 means that both groups had the same odds of event success, and therefore the technique of operations probably does not increase the risk of failure. An odds ratio of greater than 1 means that the technique of operations may increase the risk of failure, and an odds ratio of less than 1 means that the event success may reduce the risk of failure.

We performed proportional meta-analysis with metaprop, which is a command implemented in the statistical program Stata (StataCorp LP, College Station, TX) used to perform proportional meta-analyses. This command, metaprop, implements procedures that are specific to binomial data and allows computation of exact binomial and score test-based confidence interval (CI). It provides appropriate methods for dealing with proportions close to or at the margins of where the normal approximation procedures often break down, by use of binomial distribution to model the within-study variability or by allowing Freeman-Tukey double arcsine transformation to stabilize the variances. By using metaprop, no studies with 0% or 100% proportions were excluded from the meta-analysis.

We fit the logistic-normal random-effects model to the data. With this model, there is no worry about studies with cure rates close to 0% or at 100% in some studies, because we use the exact method. The CIs for the individual studies also are computed with exact method. We used the updated command metaprop one, which requires Stata 13 to fit the generalized linear mixed model. Statistical analysis was carried out using statistical software Stata 13 (StataCorp LP). Statistical significance was set at $P < .05$.

RESULTS

After an initial check, full-text retrieval, and manual cross-checking of references included in the articles, 20 studies comprising a total of 142 patients and 150 procedures clearly met the inclusion criteria and were chosen for analysis (Fig. 1). The characteristics of these selected studies are included in Table I. All studies were performed with a retrospective cohort design. The average length of follow-up was reported in only 14 studies, resulting in 18.7 months and ranging from 1 to 42 months. Among the 150 surgical procedures, 45 (30%) were performed with a transmastoid approach, whereas a middle fossa approach was used in 105 (70%) cases. Canal repairing was obtained with plugging (90 cases, 60%), resurfacing (16 cases, 10.6%) or capping (25 cases, 16.6%), and plugging together with resurfacing (19 cases, 12.6%). We considered as success all

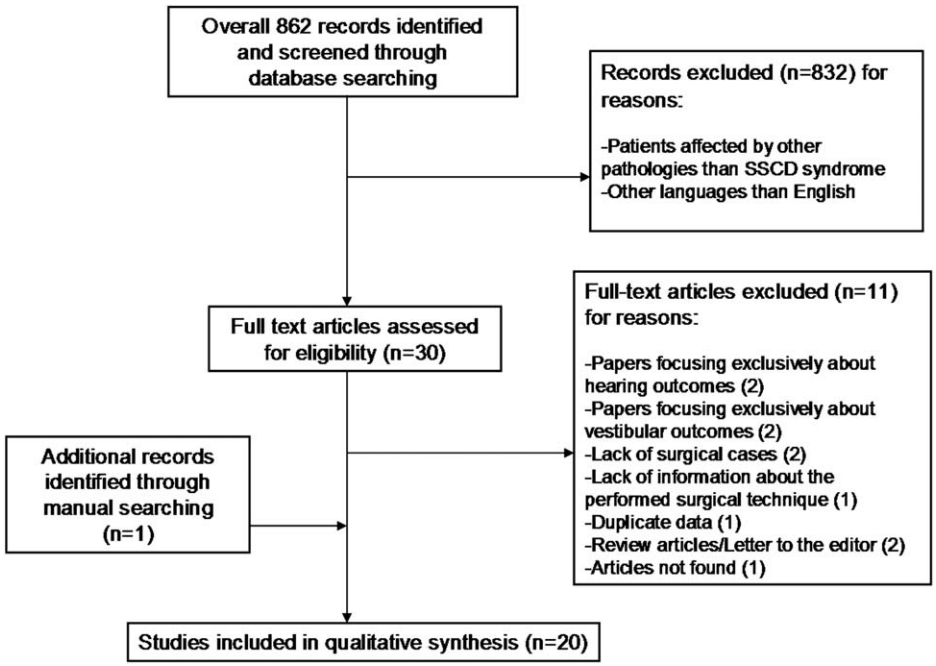

Fig. 1. Flow diagram of article selection. SSCD = superior semicircular canal dehiscence.

the 140 (93.3%) operations achieving total or partial disappearance of patients' symptoms, whereas 10 (6.7%) procedures that did not produce any improvement were considered as failures. The overall surgical outcomes and the results on the basis of different repair modalities and surgical approaches are summarized in Table II. Multiple comparisons between the repair modalities did not demonstrate any statistically significant difference in terms of success rates.

Overall, there were 11 cases of minor complications that occurred during the postsurgical period. Respectively, they were transient sensorineural hearing loss (three cases), benign paroxysmal positional vertigo (five cases), and transient important disequilibrium (three cases). Major complications resulted in permanent sensorineural hearing loss (six cases) and facial nerve palsy (three cases).

The respective complication rates were 13/90 (14%) for plugging, 4/25 (16%) for capping, 1/16 (6%) for resurfacing, and 2/19 (11%) for plugging with resurfacing. The observed difference was not statistically significant (P = .873). The respective complication rates were 7/45 (16%) for transmastoid approach and 13/105 (12%) for middle fossa approach. The observed difference was not statistically significant (P = .607).

The results of the univariate logistic regression were not statistically significant (Table III).

Meta-analysis

The overall success rate for the 150 procedures is illustrated in Figure 2. This figure represents the study-specific proportions of a successful procedure with 95% exact CI for each study and overall pooled estimate with 95% CI with logit transformation and back transformation, χ^2 statistic of likelihood ratio (LR) test comparing the random- and fixed-effects model, the estimated between-study variance, and significance testing if the estimated proportion is equal to zero. Analyzing together all the operations performed with both transmastoid and middle fossa approach, results were homogeneous, and the pooled percentage of success rate was 94% (95% CI: 87%-97%); LR test = 0.46 (df =1), P = .25; test of ES = 0, z = 6.17, P = .001. The P value for the LR is .25, indicating presence of not statistically significant heterogeneity. Between-study variance estimation is 0.28, indicating presence of not statistically significant heterogeneity.

The pooled percentage (95% CI) rate for possible complications was 12% (95%CI: 5%-25%). Results were homogeneous and the LR test = 10.01 (df = 1), P = .00 (statistically significant); test of ES = 0, z = −4.33, P = .00 (Fig. 3). Figure 3 presents the study-specific proportions of complications with 95% exact CI for each study and overall pooled estimate with 95% CI with logit transformation and back transformation, χ^2 statistic of LR test comparing the random- and fixed-effects model, the estimated between-study variance, and test of significance testing if the estimated proportion is equal to zero.

DISCUSSION

On the basis of the results observed in our analysis. it appears that surgical treatment for SSCD syndrome ensures a high rate of success and is associated to a low number of complications that often are only temporary during the postsurgical period.

Concerning the possible modalities of canal repair, four main techniques were identified in this review. Resurfacing of the superior canal or, more commonly, plugging represent the two main described techniques, but recently some authors introduced canal capping as a possible alternative. A fourth option was identified by

TABLE I.
Main Characteristics and Data of Included Studies.

Authors	Year	Country	No. of Patients	No. of Procedures	Mean Follow-up (mo)	Repairing Technique	Seal Material
TM approach							
Brantberg et al.[15]	2001	Sweden	2	2	4.5	Plugging	Fascia
Kirtane et al.[2]	2009	India	1	1	12	Plugging	Bone paté; bone wax; cartilage
Crovetto et al.[16]	2008	Italy	1	1	N.A.	Resurfacing	Bone graft; fascia
Teixido et al.[17]	2008	U.S.A.	5	5	N.A.	Plugging	Bone wax; perichondrium
Deschenes et al.[14]	2009	U.S.A.	2	3	3.5	Plugging	Bone paté; bone wax
Fiorino et al.[18]	2010	Italy	6	6	N.A.	Plugging + resurfacing	Bone dust; fibrin glue; cortical bone
Amoodi et al.[8]	2011	Canada	4	5	36.5	Resurfacing	Bone paté; fascia
Teixido et al.[19]	2011	U.S.A.	5	5	9	Resurfacing	Tragal perichondrium
Beyea et al.[6]	2012	Canada	16	16	N.A.	Plugging	Bone paté; fibrinogen glue
Dang et al.[20]	2014	U.S.A.	1	1	36	Plugging + resurfacing	Temporalis fascia; bone pate
MF approach							
Martin et al.[21]	2004	U.S.A.	1	1	12	Resurfacing	Bone graft; temporalis fascia
Mikulec et al.[4]	2005	U.S.A.	11	11	1	Plugging (10)/ resurfacing (1)	Bone wax; bone graft
Watters et al.[22]	2006	U.S.A.	1	1	18	Plugging + resurfacing	Bone wax; temporalis fascia; bone graft
Hillman et al.[23]	2006	Australia	13	16	N.A.	Capping	Hydroxyapatite bone cement; temporalis fascia
Wilkinson et al.[24]	2008	U.S.A.	1	1	36	Plugging	Temporalis fascia
Peterson et al.[3]	2008	U.S.A.	3	3	N.A.	Resurfacing	Bonesource
Niesten et al.[25]	2012	U.S.A.	32	34	28.7	Plugging	Bone wax; temporalis fascia; bone graft
Carter et al.[26]	2014	U.S.A.	5	5	4	Plugging + resurfacing	Bone wax; bone graft
Goddard and Wilkinson[27]	2014	U.S.A.	23	24	19.9	Plugging (18)/plugging + resurfacing (6)	Bone wax; bone graft
Mueller et al.[1]	2014	Switzerland	9	9	42	Capping	Hydroxyapatite

MF = middle fossa; N.A. = not available; TM = transmastoid.

some surgeons who started to perform both plugging with resurfacing during the same operation to improve their surgical success. On the basis of our statistical analysis, the difference in terms of success rate between the four modalities of repair was not statistically significant, and when analyzed in consideration of the associated surgical approach (transmastoid or middle fossa) they did not show any statistically significant difference.

Several authors consider plugging to be more effective than resurfacing in reducing symptoms of SSCD,[4,10] because of the risk of dislocation or absorption of the graft material used in the resurfacing technique. The failures of canal resurfacing reported in literature are mainly due to the dislocation or absorption of the graft material.[1] However, between the procedures performed with exclusive canal resurfacing, we found only one case of failure (success rate 94%), whereas seven failures (success rate 92%) were observed among the patients treated with canal plugging. These data appear in contrast with the concept mentioned above, but it must be noted as the total number of patients forming the resurfacing group is very limited in comparison to the plugging group (n = 16 vs. n = 90). This consideration may partially explain these unexpected results. A similar result in terms of success rate (92%) was observed for capping carried out in 25 surgical operations. From a surgical point of view, resurfacing and capping may also be considered as two variants of the same procedure, because in both these techniques the dehiscent canal roof is repaired covering the defect without the insertion of any material in the canal lumen. Interestingly, from data obtained in our analysis it should be noted that performing canal plugging associated with canal resurfacing ensures excellent results in terms of definitive symptom disappearance (success rate 100%). Whereas single canal plugging showed a worse success rate (92%). However, it should also be noted that the number of procedures performed with an exclusive canal plugging was higher than the number of operations describing plugging with resurfacing (n = 90 vs. n = 19).

Concerning hypothetical postsurgical complications, it may be affirmed that resurfacing and capping promise to guarantee a major preservation of the structures forming the internal ear in comparison to plugging. In accordance with some authors,[7] the risk of rupture of the membranous canal seems to be higher in plugging than in resurfacing and capping, because manipulation of the membranous canal is more violent in plugging.

TABLE II.
Outcomes on the Basis of Different Repair Modalities and Surgical Approaches.

Surgical outcome	Plugging	Resurfacing	Plugging + Resurfacing	Capping	Total
Overall results on the basis of different repair modalities					
Success	83	15	19	23	140
Failure	7	1	0	2	10
Total	90	16	19	25	150
Results observed with transmastoid approach					
Success	25	11	7	—	43
Failure	2	0	0	—	2
Total	27	11	7	—	45
Results observed with middle fossa approach					
Success	58	4	12	23	97
Failure	5	1	0	2	8
Total	63	5	12	25	105

The respective success rates were 83/90 (92%) for plugging, 15/16 (94%) for resurfacing, 23/25 (92%) for capping. and 19/19 (100%) for plugging with resurfacing. The observed difference found was not statistically significant ($P = .657$). For the transmastoid approach, canal plugging was performed in 27 ears, canal resurfacing in 11, whereas seven underwent plugging with resurfacing of the superior semicircular canal. The respective success rates were 25/27 (93%) for plugging technique, 11/11 (100%) for resurfacing, and 7/7 (100%) for plugging with resurfacing operations. The comparison among the different repair techniques showed a difference not statistically significant ($P = .498$) in terms of success rates. For the middle fossa approach, canal plugging was performed in 63 ears, canal resurfacing in five, whereas 12 underwent plugging with resurfacing and 25 were treated with capping of the superior semicircular canal dehiscence. The respective success rates were 58/63 (92%) for plugging technique, 23/25 (92%) for capping, 4/5 (80%) for resurfacing, and 12/12 (100%) for plugging with resurfacing. The observed differences were not statistically significant ($P = .553$).

The eventual rupture of the membranous canal should be related to permanent/transient sensorineural hearing loss or important disequilibrium. So we expected to find a greater amount of hearing loss, dizziness, and benign paroxysmal positioning vertigo among patients who underwent plugging in comparison to those treated with exclusive resurfacing. On the other hand, capping could theoretically show complications caused by the hydroxyapatite cement, which is widely used in neurosurgery. The most important long-term complication observed with this material is local infection, especially when the cement is in contact with the frontal sinus. Seroma and foreign body reactions have also been reported.[11,12]

However, from our analysis we could not find any statistically significant relation between the reported complications and different repairing techniques. In the patients treated with exclusive plugging, there were four cases of permanent sensorineural hearing loss. Interestingly, two cases of permanent sensorineural hearing loss were also observed among patients who underwent capping or resurfacing. Conversely, no cases of hearing loss were noted in any of the 19 subjects who underwent plugging with resurfacing.

Another debated question about SSCD surgery is regarding which is the better approach to perform a surgical repair of the superior semicircular canal. Middle fossa craniotomy with resurfacing and/or plugging of the defect has been the traditional treatment for patients with debilitating vestibular or auditory symptoms of SSCD.[13] However, although the results of the middle fossa approach may be favorable, a middle fossa craniotomy carries with it the risk of serious complications. Temporal lobe retraction puts the patient at risk for both transient and permanent adverse events such as contusion, ischemia, infarction, seizure, the development of epilepsy, and aphasia.[14] In addition to the risk of complications related to the middle cranial fossa approach, patients undergoing this procedure typically endure an inpatient recovery period of at least several days, including 1 day in an intensive care setting following their craniotomy. This can be avoided completely via the transmastoid approach, because it allows occlusion of the canal anterior and posterior to the dehiscence while avoiding dural elevation and risk to the membranous labyrinth.[13] However, when resurfacing is performed through the transmastoid approach, exposure of the dehiscence with the mirror is sometimes impossible,[13] and the correct placement of the bone, cartilage, or fascia material can only be verified postoperatively by means of CT.[1] On the basis of our analysis, the respective complications rates for the transmastoid approach and the middle fossa approach did not show a statistically significant difference.

It must be noted that our study presents some weaknesses. First, in our meta-analysis it was impossible to consider the role played by different materials employed to perform resurfacing or plugging. In fact, the large majority of authors did not use the same material in all the procedures performed. Second, in some of the included articles the mean follow-up was relatively short and even lacking in some others. Moreover, concerning

TABLE III.
Results of Univariate Logistic Regression for Surgical Outcomes.

Surgical Outcome	OR	95% CI
Overall		
Plugging	Reference category	
Resurfacing	0.79	0.09–6.89
Plugging + resurfacing	1	Omitted
Capping	1.03	0.20–5.30
Middle fossa approach		
Plugging	Reference category	
Resurfacing	2.90	0.26–31.14
Plugging + resurfacing	1	Omitted
Capping	1.01	0.18–5.57

Univariate logistic regression determined that the resurfacing technique had a 0.79 odds ratio, compared to the repair mode of successful surgical outcome (plugging); a value of odds ratio for the plugging with resurfacing technique was omitted because there were no events, and a value of 1.03 odds ratio was obtained for the capping technique. The results of the univariate logistic regression are not statistically significant. For middle fossa approach, the univariate logistic regression determined that the resurfacing technique had a 2.90 odds ratio, compared to the repair mode of successful surgical outcome (plugging); a value of odds ratio was omitted for the plugging with resurfacing technique, because there were no events, and a value of 1.01 odds ratio was obtained for the capping technique. For the transmastoid approach, the logistic regression was not performed, because there were no events. The results of the univariate logistic regression are not statistically significant.

CI = confidence interval; OR = odds ratio.

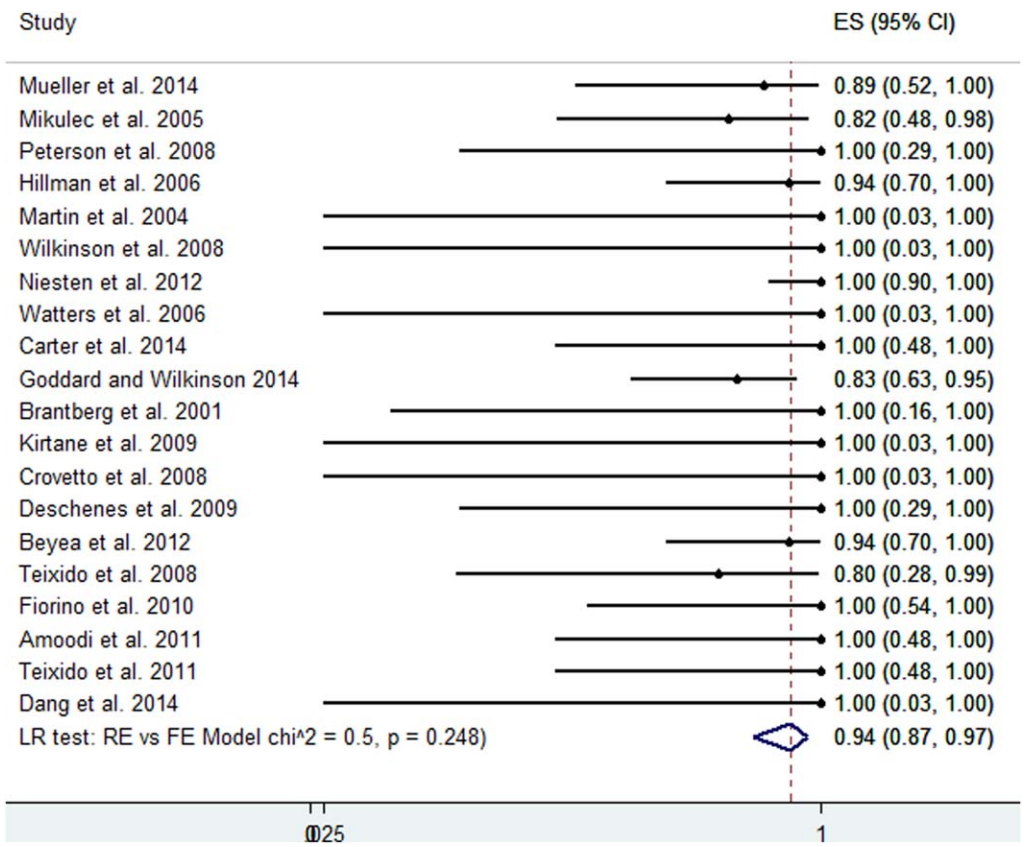

Fig. 2. Proportional meta-analysis of the included studies with procedure success rate and 95% confidence interval (CI). Pooled proportion of patients with success procedure. Size of data markers is proportional to sample size. The combined data marker indicates the DerSimonian-Laird combined proportion of all studies. CI indicates confidence interval. Output generated by the Stata procedure metaprop. ES = effect size; LR = likelihood ratio; RE = random effect; FE = fixed effect. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

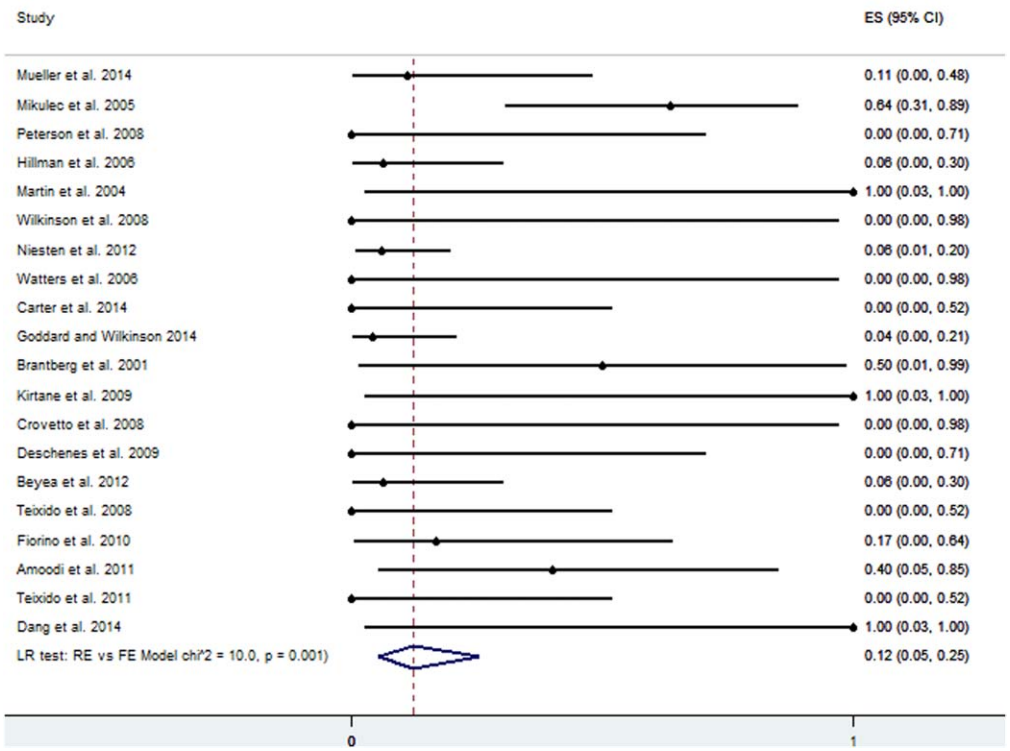

Fig. 3. Proportional meta-analysis of the included studies with procedure complications rate and 95% confidence interval (CI). Pooled proportion of patients with success procedure. Size of data markers is proportional to sample size. The combined data marker indicates the DerSimonian-Laird combined proportion of all studies. CI indicates confidence interval. Output generated by the Stata procedure metaprop. ES = effect size; LR = likelihood ratio; RE = random effect; FE = fixed effect. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

the correct definition of *success*, it must be considered that among the patients forming the success group, subjects are also present who reported only partial symptomatic relief. Another important issue to consider is the small number of patients forming this review. This last limitation is related to the rare incidence of SSCD syndrome.

Finally, concerning statistical analysis, a limitation in our calculation of odds is due to the difference in the number of observations between the technique subgroups. The calculated ratios refer to the specific dataset; however, data selection, with regard to the type of surgical intervention, was not biased.

CONCLUSION

This review showed that surgical treatment for SSCD represents a safe option for those patients who complain of severe symptoms of this condition. Overall, among the four different techniques proposed to perform superior semicircular canal repair, we could not find any statistically significant difference in terms of success rate and complications. Nevertheless, in our opinion, the high rate of success obtained performing canal plugging together with resurfacing, even though this technique was applied in only a few cases, must be stressed. Future studies on a wider cohort of patients treated with this modality could confirm if this option represents the best choice in terms of symptom disappearance. Moreover, concerning the two approaches described to visualize the superior semicircular canal during this surgery, our analysis did not show significant differences in terms of possible complications between the middle fossa approach and transmastoid route.

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