Title

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Journal

Otolaryngology, 153(5)

ISSN

0194-5998

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Publication Date

2015-11-01

DOI

10.1177/0194599815573223

Peer reviewed

AMERICAN ACADEMY OF OTOLARYNGOLOGY–HEAD AND NECK SURGERY FOUNDATION

Otolaryngology–Head and Neck Surgery
2015, Vol. 153(5) 822–831

DOI: 10.1177/0194599815573223
http://otojournal.org
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Vestibular Schwannoma Excision in Sporadic versus Neurofibromatosis Type 2 Populations

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No sponsorships or competing interests have been disclosed for this article.

Received June 17, 2014; revised January 8, 2015; accepted January 27, 2015.

Abstract

Objective. To understand the differences in characteristics of neurofibromatosis type 2 (NF2) and sporadic patients with surgically excised vestibular schwannomas in the state of California.

Study Design. Cross-sectional.

Setting, Subjects, and Methods. The records of all patients who underwent vestibular schwannoma excision between 1997 and 2011 were extracted from the California Hospital Inpatient Discharge Databases (CHIDD). NF2 cases were identified using ICD-9-CM diagnosis code 237.72, neurofibromatosis, type 2. All other cases were recoded as sporadic. Trends in total number and population-adjusted rates (per 1 million California residents) of surgery, demographics, hospital case volume, state of residency, complications, length of stay, total charges, expected source of payment, and disposition were examined.

Results. Vestibular schwannoma (VS) excision was performed on 7017 patients, of which 464 patients (6.6%) had NF2. The population-adjusted surgery rate declined from 11.8 to 6.2 ($P < .001$) for sporadic cases and from 0.3 to 0.2 ($P = .01$) for NF2 cases over the study period. NF2 was associated with younger age (mean, 32.9 vs 51.3), higher rate of other complications (8.8% vs 4.4%) and facial nerve complications (32.3% vs 16.8%), higher total charges (median $70,106 vs $46,395), longer stay (median 5 vs 4), and high volume hospitals (80.4% vs 48.8%) (all $P < .001$).

Conclusion. The surgery rates for vestibular schwannoma excision for both sporadic and NF2 patients have declined, but the decline is more prominent for sporadic cases. NF2 patients tend to be younger and have a longer hospitalization and possibly higher corresponding hospital charges compared to patients with sporadic VS.

Keywords

acoustic neuroma, vestibular schwannoma, neurofibromatosis type 2, excision, California Hospital Inpatient Discharge Data

Introduction

Vestibular schwannoma (VS) is a benign tumor of the Schwann cells of the vestibular component of the eighth cranial nerve. Continued growth of the tumor can lead to compression of adjacent neural structures of the cerebellopontine angle. This tumor accounts for 6% of all intracranial tumors, with an incidence rate of approximately 1 in 100,000.[1] While more than 90% of cases are unilateral and sporadic developing between the fourth and fifth decades, bilateral VS occur in less than 5% of cases and are limited to patients with neurofibromatosis type 2 (NF2).[2,3] Recent estimates based on the UK Family Genetic Register Service showed that NF2 is more common than previously observed.[4] In this autosomal dominant syndrome, the most common clinical feature is the development of pathognomonic bilateral VS, occurring in greater than 90% of those affected.[5] Similar to the sporadic type, the natural history of VS in NF2 is progressive tumor growth, with hearing loss as the symptom that has the greatest effect on the patients' quality of life.[6,7]

Improvements in diagnostic modalities have resulted in increased disease incidence, decreased tumor size at time of presentation, and evolved treatment strategies. Management options for these tumors now include watchful observation and stereotactic radiosurgery (SRS), in addition to the conventional

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Presented as poster at the 48th American Neurotology Society Meeting, COSM; April 12-13, 2013; Orlando, Florida

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microsurgical approach.[8] While these advances have allowed for a more individualized treatment strategy, surgical excision continues to be the treatment of choice for progressive or symptomatic VS.[9-11] Microsurgery is favored over radiotherapy in NF2 patients due to the relatively younger age at presentation and overall goal of long-term tumor control.[12]

With the development of high-resolution magnetic resonance imaging (MRI) scans and the treatment options of observation and SRS, the characteristics of patients undergoing microsurgery have transformed. This study seeks to assess the demographics of NF2 patients with surgically excised VS using the California Hospital Inpatient Discharge Data. Likewise, the length of stay, cost of care, outcome, and mortality of these patients are analyzed.

Methods

Data Source

The California Office of Statewide Health Planning and Development (OSHPD) collects data on all inpatients discharged from all licensed hospitals in California. Licensed hospitals include general acute care, acute psychiatric, chemical dependency recovery, and psychiatric health facilities. Data sets are released in form of de-identified records that include patient-level data such as demographics, diagnosis and procedure International Classification of Diseases, 9th edition, Clinical Modification (ICD-9-CM) codes, payment source, admission source, length of stay, and total charges. Information on data collection instruments and methodology is available on the website of OSHPD.[13]

California Hospital Inpatient Discharge Datasets from 1997 to 2011 were merged, and all cases with ICD-9-CM procedure code 04.01, excision of acoustic neuroma, were extracted. NF2 cases were identified using ICD-9-CM diagnosis code 237.72, neurofibromatosis, type 2 (acoustic neurofibromatosis). All other cases were recoded as sporadic. Seventeen cases with unspecified neurofibromatosis codes were excluded due to potential miscoding.

Variables and Definitions

Trends in total number of surgeries and population-adjusted surgery rates (number of surgeries per 1 million California residents), age, gender, ethnicity, hospital case volume, state of residency, complications, length of stay, total charges, expected source of payment, and disposition were examined. Study years were grouped into 5-year periods: 1997-2001, 2002-2006, and 2007-2011. Age was categorized into 4 groups: 1-17 years, 18-34 years, 35-64 years, and 65 years and older. Ethnicity was re-encoded as Caucasian versus non-Caucasian (representing African Americans, Native Americans/Eskimo/Aleut, Asian/Pacific Islanders, and others) because only about 21% of cases had entries, and entries for minorities were very sparse. Hospitals were categorized based on the number of VS excisions performed during the entire study period into low volume (15-90 cases), moderately low volume (105-285 cases), moderately high volume (300-525 cases), and high volume (540 cases or more). The volume classifications were determined to represent the entire span of the data during the study period and followed the previous literature on the topic.[14-16] State of residence was classified into California and other (out-of-state, homeless, or those cases that had an invalid or blank entry) based on the recorded zip codes.

Pertinent patient-level data were gleaned from the database. Cases that utilized auditory brainstem and cochlear implants were identified using the appropriate ICD-9 procedure codes (02.93, 20.96-20.99). Additionally, perioperative complications associated with VS excision were identified through a review of the literature and defined using ICD-9-CM diagnosis and procedure codes (**Table 1**).[14-16] These codes were classified into central nervous system (CNS) complications, facial nerve complications, and other complications.

Total charges rendered the hospital's full-established rates (before contractual adjustments) and included daily hospital, ancillary, and any patient care services. Hospital-based physician fees were not included in these figures. Unknown, missing, or invalid charges were excluded from the analysis. Principal payer was defined as the type of entity or organization expected to pay the greatest share of the patient's bill. Principal payer was recoded into the following groups: (1) Medicare, (2) Medi-Cal (California's Medicaid program), (3) private coverage (ie, HMO or PPO), and (4) other (ie self-pay, worker's compensation, charity, county indigent programs, other government and indigent programs: includes out of state Medicaid). Finally, disposition was recoded into: (1) home residence (routine discharge), (2) further care, (3) other, and (4) death. Further care included any of the following: acute, other, and long-term care within the hospital and acute and other care at another hospital. ''Other'' included skilled nursing/intermediate care, residential care facility, prison/jail, left against medical advice, and home health service. Since the datasets were de-identified, University of California, Irvine Institutional Review Board approval was not required for the analysis.

Statistical Analyses

Population-adjusted surgery rates were calculated as total number of surgeries on California residents per year divided by total population of California during that year. California population estimates were obtained from the US Census Bureau website.[17] The correlation between the population-adjusted rates and time was evaluated using linear regression analysis. Chi-square test was used for analysis of age groups, gender, ethnicity, hospital case volume, state of residency, complications, principal payer, and disposition. Fisher's exact test was used where chi-square assumptions were not met. Data distribution of continuous variables was examined for normality. Mean $\pm$ standard deviation (SD) was calculated for age that had a normal distribution. Median (fifth to ninety-fifth percentiles) were calculated for total charges and length of stay data, which both had significantly positively skewed distributions requiring analysis using nonparametric tests. Kruskal-Wallis H Test was used to compare total charges and length of stay between the 5-

Table 1. Classifications Used for Recoding of Complications.

	ICD-9 CM Procedure Codes	ICD-9 CM Diagnosis Codes
Central Nervous System Complications	01.2 Craniotomy and craniectomy 02.2 Ventriculostomy 02.3 Extracranial ventricular shunt 02.4 Revision, removal, and irrigation of ventricular shunt	320 Bacterial meningitis 331.3 Communicating hydrocephalus 331.4 Obstructive hydrocephalus 348.5 Cerebral edema 349.81 Cerebrospinal fluid rhinorrhea 388.61 Cerebrospinal fluid otorrhea 430 Subarachnoid hemorrhage 431 Intracerebral hemorrhage 432 Other and unspecified intracranial hemorrhage 996.75 Due to nervous system device, implant, and graft (ventricular shunt complication) 997.0 Nervous system complications
Facial Nerve Complications	04.5 Cranial or peripheral nerve graft 04.6 Transposition of cranial and peripheral 04.71-04.79 Nerve anastomoses 08.52 Blepharorrhaphy	351.0 Bell's palsy 351.1 Geniculate ganglionitis 351.8 Other facial nerve disorders 351.9 Facial nerve, unspecified 370.00 Corneal ulcer, unspecified 370.34 Exposure keratoconjunctivitis 370.35 Neurotrophic keratoconjunctivitis 370.40 Keratoconjunctivitis, unspecified 371.40 Corneal degeneration, unspecified 371.49 Other Discrete colliquative keratopathy 374.2 Lagophthalmos 781.94 Facial weakness 951.4 Injury to facial nerve
Other Complications		371.81 Corneal anesthesia and hypoesthesia 371.89 Other corneal disorders 378.53 Fourth or trochlear nerve palsy 378.54 Sixth or abducens nerve 415.1 Pulmonary embolism and infarction 453.4 Venous embolism and thrombosis of deep vessels of lower extremity 453.8 Of other specified veins 453.9 Of unspecified site 787.2 Dysphagia 787.29 Other dysphagia 951.1 Injury to other sympathetic nerve, excluding shoulder and pelvic girdles 951.2 Injury to trigeminal nerve 951.3 Injury to abducens nerve 951.6 Injury to accessory nerve 951.7 Injury to hypoglossal nerve 951.8 Injury to other specified cranial nerves 951.9 Injury to unspecified cranial nerve 998.1 Hemorrhage or hematoma or seroma complicating a procedure 998.5 Postoperative infection

year periods as well as hospital case volume groups. Data analysis was performed using PASW Statistics 18.0 (SPSS, Inc, Chicago, Illinois, USA). A P value of less than .05 was considered significant.

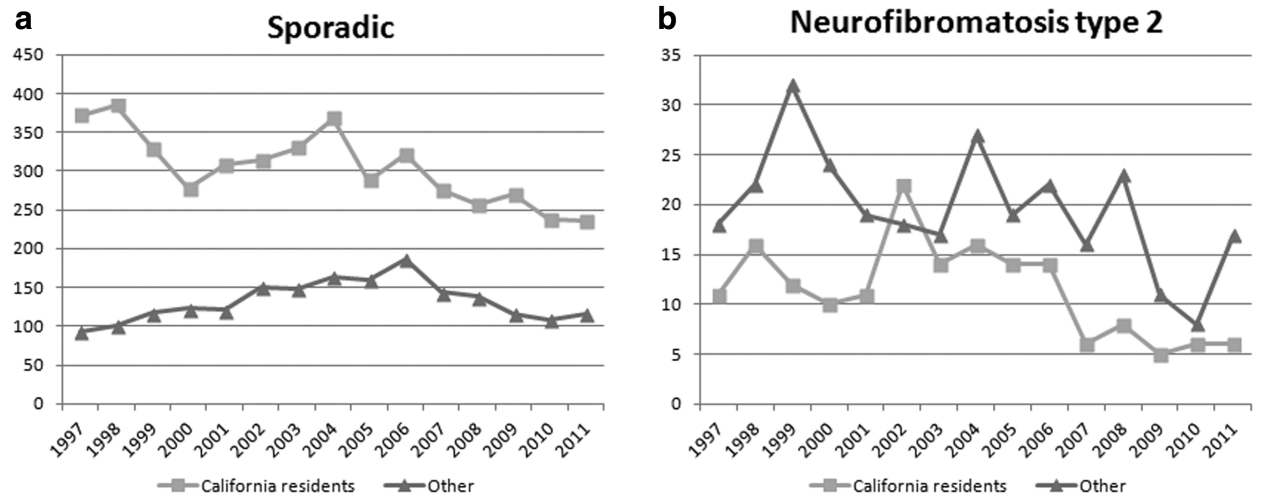

Figure 1. Total number of vestibular schwannoma excisions based on the state of residency: (a) sporadic cases and (b) neurofibromatosis type 2 cases.

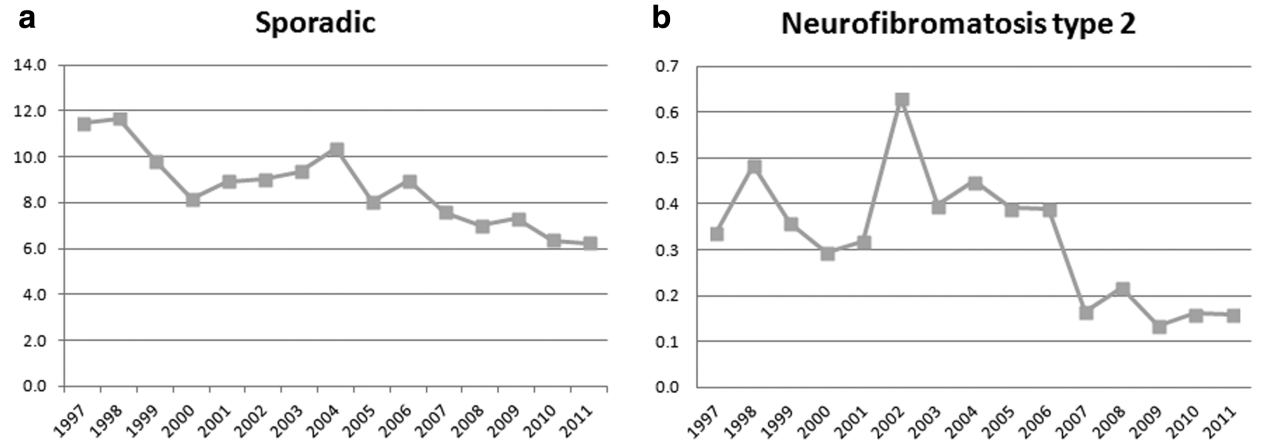

Figure 2. Population-adjusted surgery rate of vestibular schwannoma excisions in (a) sporadic versus (b) neurofibromatosis type 2 per 1 million California residents. Only the patients residing in California are included.

Results

A total of 7017 patients undergoing VS excision were identified in California between 1997 and 2011, of which 4738 (67.5%) were California residents. These surgeries were performed in 125 hospitals across California (115 low volume, 5 moderately low volume, 4 moderately high volume, 1 high volume). NF2 was present in 464 patients (6.6%), of which 171 (36.9%) were California residents. Total number of surgeries for California residents with sporadic disease decreased from 372 to 235 per year from 1997 to 2011 (**Figure 1A**). This translated into a decline in population-adjusted surgery rate from 11.8 to 6.2 per 1 million California residents (adjusted $R^2 = 0.73$, $P < .001$) (**Figure 2A**). Total number of surgeries on California residents with NF2 also showed a decreasing trend, from 11 to 6 per year (**Figure 1B**). This translated into a decline in population-adjusted surgery rate from 0.3 to 0.2 per 1 million California residents (adjusted $R^2 = 0.35$, $P = .01$) (**Figure 2B**).

The mean age of NF2 patients undergoing surgical excision was 32.9 ± 17.1 years compared with sporadic patients who had a mean age of 51.3 ± 14.8 years ($P < .001$). Overall, excisions of VS on NF2 patients were more likely to occur in younger age and high volume hospitals. In addition, NF2 patients were more likely to have other complications, higher total charges, and a longer hospital stay (**Table 2**).

Table 3 shows the trends of VS excisions in NF2 patients over the study period. Investigation of changes in the NF2 subpopulation revealed that the distribution of age groups, gender, ethnicity, and disposition has not changed over time. However, total charges and percentage of private payer increased over time. The hospital with a high volume showed a significant decrease in cases during the study period. Hospitals in the moderately high category demonstrated increased trends from 8.0% to 14.2% during the study period. Moderately low volume hospitals had a significant decrease in volume during the study period. Paradoxically, low volume hospitals had a significant increase in volume.

Table 2. Demographics and Characteristics of Patients Undergoing Sporadic or NF2-Associated Vestibular Schwannoma Excision.

	NF2	Sporadic	*P* Value
Total number of VS excisions	464	6553	
Age group, %			<.001
17 years or less	16.7	0.6	
18-34 years	40.5	9.4	
35-64 years	39.7	76.7	
65 years or older	3.2	13.4	
Gender, %			.6
Male	45.3	48.5	
Female	54.7	51.5	
Ethnicity, %			.9
Caucasian	84.2	82.9	
Non-Caucasian	15.8	17.1	
Hospital case volume, %			<.001
High	80.4	48.8	
Moderately high	8.8	25.7	
Moderately low	5.0	10.0	
Low	5.8	15.5	
State of residence, %			<.001
California	36.9	69.7	
Other	63.9	30.3	
CNS complications, %			.4
Yes	21.8	20.3	
No	78.2	79.7	
Facial nerve complications, %			<.001
Yes	32.3	16.8	
No	67.7	83.2	
Other complications, %			<.001
Yes	8.8	4.4	
No	91.2	95.6	
Length of stay, median; range (mean ± SD)	5; 1-78 (6.4 ± 6.4)	4; 0-131 (5.2 ± 5.2)	<.001
Total charges, median; range (mean ± SD)[a]	$70,106; $6,639-$1,357,941 ($101,213 ± $121,315)	$46,395; $793-$3,609,838 ($79,954 ± $116,845)	<.001
Principal payer, %			<.001
Medicare	8.8	10.7	
Medi-Cal	6.0	3.5	
Private	68.1	76.3	
Other[b]	17.0	9.5	
Disposition, %			.1
Home (routine)	90.5	90.9	
Further care	4.8	3.8	
Other	4.1	5.1	
Death	0.6	0.2	

Abbreviations: CNS, central nervous system; NF2, neurofibromatosis type 2; VS, vestibular schwannoma.
[a]Missing or invalid values, respectively: 25 and 970.
[b]Includes: workers' compensation, county indigent programs, TRICARE, other government, other indigent, self-pay, other payer.

Auditory brainstem or cochlear implant utilization was not significantly different during the study period.

Table 4 compares the characteristics of NF2 patients stratified based on hospital case volume. The CNS complications, facial nerve complications, and other complications were not significantly different for all categories. Length of stay did not depend on case volume. Total charges were statistically different depending on the volume load of the hospital. Most cases in the low group were paid by Medi-Cal. In contrast, higher volume groups (high, moderately high, moderately low) showed that a majority of cases were paid for by private insurers. The

Table 3. Changes in Demographics and Characteristics of Patients with Neurofibromatosis Type 2 with Excised Vestibular Schwannoma during the Studied Period.

	1997-2001	2002-2006	2007-2011	*P* Value
Total number of NF2 cases with excised VS	175	183	106	
Age group, %				.7
17 years or less	16.3	17.8	14.8	
18-34 years	44.2	41.1	33.3	
35-64 years	39.5	35.7	48.2	
65 years or older	0	5.4	3.7	
Gender, %				.4
Male	38.7	44.1	57.1	
Female	61.3	55.9	42.9	
Ethnicity, %				.3
Caucasian	92.9	80.6	76.5	
Non-Caucasian	7.1	19.4	23.5	
Hospital case volume, %				.04
High	83.4	79.8	76.4	
Moderately high	8.0	6.6	14.2	
Moderately low	4.6	7.7	0.9	
Low	4.0	6.0	8.5	
State of residence, %				.03
California	34.3	43.7	29.2	
Other	65.7	56.3	70.8	
CNS complications, %				<.001
Yes	11.4	33.3	18.9	
No	88.6	66.7	81.1	
Facial nerve complications, %				.01
Yes	26.9	35.5	35.8	
No	73.1	64.5	64.2	
Other complications, %				.4
Yes	10.3	9.3	5.7	
No	89.7	90.7	94.3	
Auditory brainstem or cochlear implants, %				.09
Yes	23.4	30.6	19.8	
No	76.6	69.4	80.2	
Length of stay, median; range (mean ± SD)	5; 2-33 (6 ± 5)	4; 1-33 (6 ± 4)	5; 2-78 (8 ± 11)	.005
Total charges, median; range (mean ± SD)[a]	$36,752; $1,391-956,663 ($54,564 ± $76,808)	$89,953; $6,639-$593,231 ($89,953 ± $65,6)	$195,324; $27,893-$1,357,941 ($195,324 ± $184,043)	.03
Principal payer, %				.001
Medicare	7.4	8.7	11.3	
Medi-Cal	4.6	8.2	4.7	
Private	60.6	71.6	74.5	
Other[b]	37.4	11.5	9.4	
Disposition, %				.5
Home (routine)	93.1	90.7	85.9	
Further care	2.9	5.5	6.6	
Other	3.4	3.3	6.6	
Death	0.6	0.5	0.9	

Abbreviations: CNS, central nervous system; NF2, neurofibromatosis type 2; VS, vestibular schwannoma.

[a]Missing or invalid values, respectively: 8, 14, and 3.

[b]Includes: workers' compensation, county indigent programs, TRICARE, other government, other indigent, self-pay, other payer.

Table 4. Effect of Hospital Case Volume on Outcomes of Neurofibromatosis Type 2 Patients with Excised Vestibular Schwannoma.

	High	Moderately High	Moderately Low	Low	*P* Value
Total number of NF2 cases with excised VS	373	41	27	23	
CNS complications, %					.1
Yes	20.1	29.3	17.4	37.0	
No	79.9	70.7	82.6	63.0	
Facial nerve complications, %					.4
Yes	33.5	24.4	21.7	37.0	
No	66.5	75.6	78.3	63.0	
Other complications, %					.2
Yes	8.3	9.8	4.3	18.5	
No	91.7	90.2	95.7	81.5	
Length of stay, median; range (mean ± SD)	5; 2-32 (5.6 ± 3.6)	5; 1-59 (9.32 ± 11.8)	5.7; 3-17 (5.7 ± 3.4)	6; 2-78 (12.4 ± 16.1)	.3
Total charges, median; range (mean ± SD)[a]	$68,282; $9,323-$956,663 ($88,471 ± $79,679)	$101,587; $5,639-$1,357,941 ($198,482 ± $280,726)	$73,722; $33,325-$172,117 ($85,888 ± $48,194)	$82,458.00; $26,781-$982,681 ($163,415 ± $203,577)	.009
Principal payer, %					<.001
Medicare	8.8	12.2	8.7	3.7	
Medi-Cal	2.7	12.2	8.7	40.7	
Private	69.4	70.7	82.6	33.3	
Other[b]	19.0	4.9	0	22.2	
Disposition, %					<.001
Home (routine)	95.4	63.4	78.3	74.1	
Further care	3.2	14.6	4.3	11.1	
Other	0.8	19.5	17.4	14.8	
Death	0.5	2.4	0	0	

Abbreviations: CNS, central nervous system; NF2, neurofibromatosis type 2; VS, vestibular schwannoma.
[a]Missing or invalid values, respectively: 1, 7, 15, and 2.
[b]Includes: workers' compensation, county indigent programs, TRICARE, other government, other indigent, self-pay, other payer.

high volume hospital showed the largest proportion of routine discharges.

Discussion

Over the past several decades, there has been an apparent increase in the incidence of VS attributed to the increased detection of incidental tumors.[18] A previous study showed that the mean tumor size at diagnosis registered in a national database decreased from 30 mm in 1976 to 10 mm in 2008.[19] Paradoxically, our study found that the number of patients undergoing surgical resection for NF2 VS cases has decreased from 1997 to 2011. The decreasing trend agrees with previous studies on national data for current management trends of VS.[20,21]

Interestingly, our study shows that there is a proportionally larger decrease in excision rate of sporadic VS when compared to NF2-associated VS. Since sporadic VS patients are typically older than NF2 patients, Gal et al[22] argued that increasing age is significantly associated with conservative management. This supports our finding that sporadic VS might be treated more conservatively with ''watchful waiting'' or stereotactic radiosurgery. Additionally, our results indicate that NF2 patients who underwent microsurgery for VS are significantly younger than patients with sporadic disease. We found that the average age of NF2 patients in our study is 32.9 years old, corresponding to the data from the NF2 Natural History Consortium.[23,24] Furthermore, our data show that the population-adjusted surgery rate for VS excision in NF2 patients declined from 0.3 to 0.2 per 1 million California residents. General treatment guidelines for VS in NF2 patients advocate for surgical resection of the VS when the tumor is still small (<1.5 cm), in hopes of hearing preservation.[11,25,26] Thus, NF2-associated VS patients are treated if new symptoms arise or there are signs of tumor growth. Additionally, case reports have shown that malignant transformation of VS in NF2 patients occur more often than sporadic VS. Despite these recommendations, some neurotologists treat NF2 patients with SRS. Indeed, a previous study by our group found that 64% of neurotologists perform SRS for NF2 patients.[27]

Stereotactic radiosurgery and ''watchful waiting'' have become increasingly used to manage VS when they are small in size or asymptomatic. The literature surrounding the decrease in resection rates of VS is limited and not all in agreement. Lau et al[20] utilized the Surveillance, Epidemiology and End Results Program (SEER) to evaluate current trends in treatment of VS and came to the conclusion that surgical resection for tumors specifically less than 2 cm in size have decreased in recent years in favor of SRS therapy while the overall rate for observation has remained low and stable from 2004 to 2007. On the other hand, Patel et al[21] also utilized SEER data but came to the conclusion that microsurgery decreased from 92.7% to 53.4%, while the use of radiosurgery/radiotherapy increased from 5% to 24.2%, and observation increased to 22.4%. Thus, it is apparent that the etiology for the decline in surgery rates for VS is due to increased use of SRS and observation, but it is currently difficult to determine the amount each option has contributed to the overall decline in surgery rates. Further research on the trends of SRS on sporadic and NF2 patients is necessary to determine how these tumors are treated. A cross-sectional investigation of the current treatment trends might be useful in the long-term follow-up of these patients.

As NF2 is predominantly inherited in an autosomal dominant manner, it can present in early childhood with cataracts and multiple intracranial tumors. CNS complications, such as cerebrovascular accident, can occur in patients with NF2 due to larger tumor sizes in these patients and the presence of multiple intracranial tumors that can lead to a more complicated resection.[28] Although our findings showed a higher incidence of CNS complications in NF2 patients, this did not reach statistical significance. We also discovered that other serious systemic complications, such as pulmonary embolism, hemorrhage, hematoma, and postoperative infection, were more common in patients with NF2 than in sporadic patients (**Table 2**), which has not been reported in the literature. These associated complications may occur because patients with NF2 who undergo surgery have more medical comorbidities compared with sporadic cases and have a longer hospital stay. Many of the sporadic disease patients with multiple comorbidities elect to have SRS rather than microsurgery. Additionally, facial nerve complications after VS excision occur more frequently in the NF2 tumors than sporadic VS (**Table 2**). This may be due to the more vascular nature of the NF2 tumors or previous irradiation when compared to sporadic VS. In general, the literature has shown that morbidity and mortality are significantly lower when complex surgical procedures are performed at high volume hospitals. For example, Slattery et al[14] and Barker et al[16] reported that the hospitals or providers with the highest volumes of VS surgeries had lower total charges and average charges per day than lower volume hospitals. Our study confirms the inverse relationship of volume and total charges.

In a comparison of NF2 and sporadic patients, NF2 patients stayed in the hospital longer than patients with sporadic disease (median 5 vs 4), with an associated increase in hospital charges. Our results are similar to results from other centers, including in other countries. In 1 series, the average duration of hospital stay for patients with sporadic VS undergoing surgical resection was 6.4 ± 3.42 days (range, 2-36 days).[29] Again, these differences may occur because patients with NF2 who undergo surgery have more medical comorbidities compared with sporadic cases, and sporadic patients with comorbidities tend to choose SRS over microsurgical resection.

In California, private insurance is the principal payer for the majority of NF2 and sporadic VS excisions at 68.1% and 76.3%, respectively (**Table 2**). The primary difference is related to the difference in the ''other'' category and may be due to a higher group of out-of-state patients in the NF2 group. Our data show that the trend for hospitalization charges associated with VS resection in NF2 patients has increased from a median of $35,785 in 1997-2001 to $195,324 in 2007-2011 despite stable median hospitalization stays. Even when accounting for an average hospital-based inflation rate of 232% from 1991 to 2011, the average charge for NF2 patients is more than double the amount that would be expected ($83,021).[30] Although new technologies may be a factor in the increase in cost, we found that there has been no increase in utilization of auditory brainstem or cochlear implant utilization in the NF2 population throughout the study period (**Table 3**). An additional area to explore is a cost analysis of NF2 patients undergoing excision of VS because of their relatively young age and other comorbidities.

An analysis of the state of residence of NF2 patients suggested that the majority of patients resided outside California. One explanation is that California has several high volume hospitals and subspecialty ear clinics, with 3 neurotology fellowship programs that attract out-of-state patients.

Our data suggest that there are significant differences in median charges depending on volume (**Table 4**). Complicated patients are often referred to the higher volume centers. Therefore, it is possible that the higher volume centers achieved the same level of charge for a more complicated average patient. Furthermore, hospitals with high volumes for treating NF-2 and vestibular schwannomas were associated with a higher frequency of routine discharges and lower frequencies of complications that may further decrease cost. Interestingly, we found that moderately high volume hospitals had the highest median total charges ($101,587). One explanation would be that these hospitals treat the complex cases, resulting in high costs. While this number is relatively higher, since charges are determined by each hospital, it is unclear if these hospitals have a higher baseline charge for the same services or if there is a higher utilization that results in the higher level of charges.

Findings of the current study should be interpreted with the understanding of its limitations. The generalizability of the data may be limited to states that are similar in demographics and a larger, more diverse population like California. States with multiple academic medical centers and neurotology fellowships may demonstrate similar trends as well. Nonetheless, findings from a statewide database are more likely to represent

national trends in comparison to single institutional studies. Furthermore, specific characteristics of cases such as tumor size or location, preoperative radiation, surgical approach, or extent of resection that influence the surgical outcome for VS excisions were not captured by the data sets. Additionally, NF2 patients might have had surgeries on each side on different years, or surgery on one ear and radiation on the other, and there was no way account for these in the data sets. Furthermore, analysis of the charges based on the data set should be treated with caution. Annual changes in the hospital's charge master and fee codes could directly affect the total charges for a given patient. Charges are based on negotiated fees between the hospital and insurance companies and create huge discrepancies. Although we found that most of these cases were paid for by federal, state level, or other programs, private insurers have different fee schedules for the same procedures and services. Thus, these factors may explain the discrepancies between expected trends and actual results from the data. Finally, as with any other large data set analysis, the accuracy of the findings is dependent on the accuracy of data entry, which could not be evaluated.

Conclusion

Total number and population-adjusted rates of surgery for VS excision in both sporadic and NF2 patients have declined in California from 1997 to 2011. Sporadic excisions rates, however, have declined more substantially than NF2-associated excision rates. About 6.6% of VS excisions were performed on NF2 patients, who tend to have a younger age, more systemic complications, longer stay, higher total charges, and governmental source of payment in comparison to sporadic cases.

Author Contributions

Hossein Mahboubi, writing/data analysis; **Marlon M. Maducdoc**, writing/data analysis; **Amy Y. Yau**, writing/concept; **Kasra Ziai**, writing/data analysis; **Yaser Ghavami**, writing/data analysis; **Karam W. Badran**, writing/data analysis; **Majid Al-Thobaiti**, analyzed data, wrote article, revised article; **Bryan Brandon**, analyzed data, wrote article, revised article; **Hamid R. Djalilian**, concept and writing.

Disclosures

Competing interests: None.

Sponsorships: None.

Funding source: None.

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