Management of Ranula: 9 Years' Clinical Experience in Pediatric and Adult Patients

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Ranulas are extravasation mucoceles that arise from the sublingual gland (SLG) owing to a ruptured main duct or ruptured acini after obstruction.[1] The term *ranula* is the Latin word for a small frog, and it is used for the condition because it causes croaking speech and an oral ranula resembles a frog's belly. In plunging ranulas, the mucous collection extends into the neck, with or without associated intraoral swelling.[2] A congenital predisposition to the development of plunging ranula has been suggested based on the association with anatomic anomalies in the mylohyoid muscle, the occurrence of plunging ranula among siblings, and the observation of a larger number of ranulas in some ethnic groups.[3-5] Extensions of the

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Table 1. MAIN DEMOGRAPHIC DATA FOR PATIENTS TREATED FOR RANULA FROM JANUARY 2001 THROUGH DECEMBER 2010

	Patients (n)	Mean Age (yr)	Men (n)	Women (n)	Right Side (n)	Left Side (n)	Adult (n)	Pediatric (n)
Oral ranula	55	35	32	23	27	28	44	11
Plunging ranula	10	26	8	2	5	5	8	2
Total	65	33	40	25	32	33	52	13

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ranula into the neck can occur along the deep lobe of the submandibular gland between the mylohyoid and hypoglossal muscles or from congenital dehiscence in the mylohyoid muscle itself, with part of the SLG projecting into it. This finding is seen in up to one third of normal individuals and corresponds to the Gaughran mylohyoid boutonniere.[6] The diagnosis of oral and plunging ranula may be based on the clinical presentation, but ultrasonography, computed tomography, magnetic resonance imaging, and fine-needle aspiration have also been used for differential diagnosis, particularly in patients with plunging ranulas.[7] Several surgical approaches and techniques for oral and plunging ranulas in pediatric and adult patients have been proposed, with different results. Among these, excision of the SLG is considered the most effective form of treatment, although it is associated with a certain risk of complications. These include injury to the lingual nerve or Wharton duct, with possible stenosis, leading to obstructive sialadenitis and ductal laceration, causing salivary leakage.[8]

The present report summarizes the experience in the authors' department in the management of patients with oral or plunging ranulas over a period of 9 years.

Materials and Methods

A retrospective analysis was performed in the Department of Otorhinolaryngology–Head and Neck Surgery at the University of Erlangen-Nuremberg, including data for all pediatric and adult patients treated for a final diagnosis of oral or plunging ranula from January 2002 through December 2010. The study was approved by the local ethics committee.

The diagnosis was based on clinical and ultrasound features and was confirmed after histologic evaluation of the surgical specimens. All images were obtained using a state-of-the-art ultrasound system (Acuson Antares; Siemens Medical Solutions USA, Inc, Ultrasound, Mountain View, California) with a VF10-5 linear scanner (5 to 10 MHz; Siemens Medical Solutions USA, Inc). Two patients with final histologic findings showing lymphangioma and dermoid cyst were excluded from the analysis, as were 5 patients with small oral ranulas who declined surgery. Sixty-five patients were included in the study. The case notes were evaluated to obtain the main demographic, clinical, diagnostic, and therapeutic data. These included gender; age at presentation; lesion type, side, and size; previous surgery; diagnostic management; surgical treatment; type of anesthesia; treatment for recurrences; and follow-up period. The patients had follow-up examinations with a clinical evaluation followed by ultrasonography after 1 week, 1 month, and 6 months after surgery, and any complications were noted. All patients were contacted by telephone in July 2011 and asked about any current symptoms and any postoperative complications not previously reported.

Statistical analysis was carried out using GraphPad Prism (http://www.graphpad.com/quickcalcs/CatMenu.cfm). A nonparametric Fisher exact test was used to analyze the variance among the different types of treatments and the relative statistical significance of the differences. A nonparametric binomial exact test was used to compare the results in the treatment of ranula with the results published in a recent meta-analysis by Harrison.[1]

Results

The main demographic, clinical, and therapeutic data are listed in Table 1. The patients' mean age at the time of surgery was 33 years (range, 6 to 72 yr); 13 were children (<16 years old) and 40 were male (61.5%). The locations of the ranulas were evenly distributed on both sides (32 on the right, 33 on the left), and their median size at the preoperative ultrasound evaluation was 26 × 16 mm. The clinical appearance of an oral ranula is shown in Figure 1.

Ten patients (15.4%), 2 of whom were children (18.2%), had plunging ranulas; the mean age in this subgroup of patients was 26 years (range, 8 to 48 years). B-mode and color-coded duplex ultrasonography was performed in all patients during the preoperative evaluation. Ultrasound showed a homogeneous, ovoid, and thin-walled hypoechoic lesion, with the classic features of a cyst, intimately related to the mylohyoid muscle and the SLG in every case (Figs

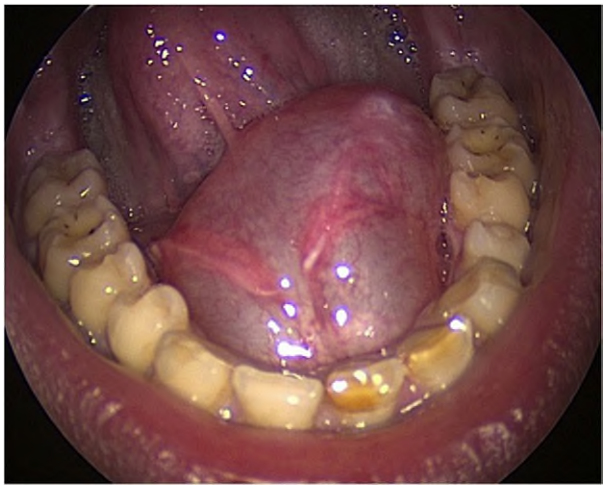

FIGURE 1. An oral ranula on the floor of the mouth on the left side, causing deviation of the tongue and crossing the midline.

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2, 3). Dehiscence of the mylohyoid muscle was noted in some cases. There was an incidental finding of a hilar stone in the ipsilateral submandibular gland in 2 patients who underwent ultrasound examinations for oral ranula. Sixteen patients (24.6%) had previously undergone related surgery or treatments, including ranula marsupialization (n = 3); sialodochotomy in the submandibular duct alone owing to duct stenosis (n = 3) or in combination with transoral removal of submandibular gland stones (n = 5); partial excision or biopsy of the SLG (n = 3); excision of leukoplakia on the floor of the mouth (n = 1); and extracorporeal shock wave lithotripsy (n = 1). Informed consent was obtained from all patients preoperatively.

Surgery was performed with the patients under local anesthesia (n = 44) or general anesthesia (n = 21) using transnasal intubation, depending on the clinical features of the lesion, the patient's age, and the surgical approach preferred by the surgeon. General anesthesia was chosen in 3 cases because the patients required another head and neck procedure. Various treatments were performed (Fig 4): marsupialization (n = 21); excision of the ranula alone (n = 11) or in combination with partial (n = 11) or total (n = 18) removal of the ipsilateral SLG; and excision of the SLG alone, without the ranula (n = 4). Treatment for plunging ranulas (10 patients) was also based on 2 different surgical approaches—transoral or transcervical (3 cases; Figs 5, 6).

All surgical specimens were sent for histopathologic examination, with the final reports showing chronic inflammation of the SLG and cystic extravasation mucoceles. The follow-up period was 6 to 112

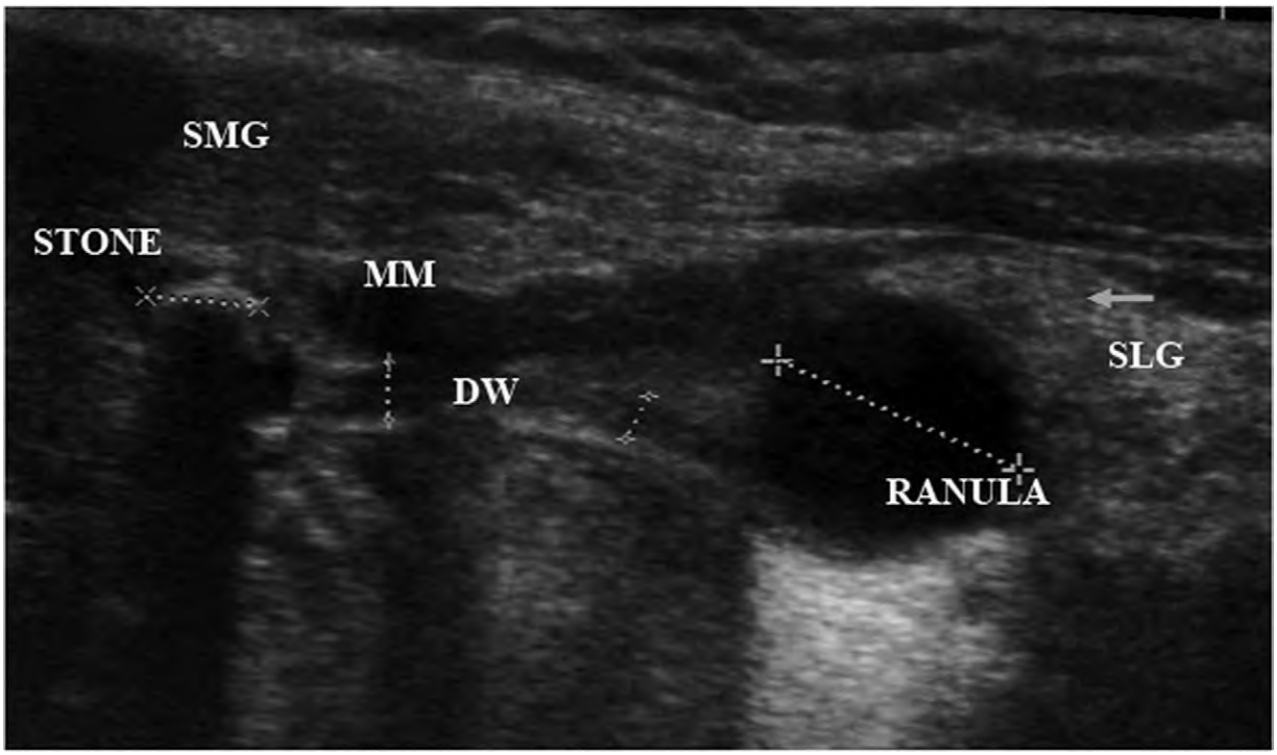

FIGURE 2. A sagittal oblique ultrasound view of an oral ranula lying over the mylohyoid muscle (MM) in association with a stone in the submandibular gland (SMG). DW, Wharton's duct; SLG, sublingual gland.

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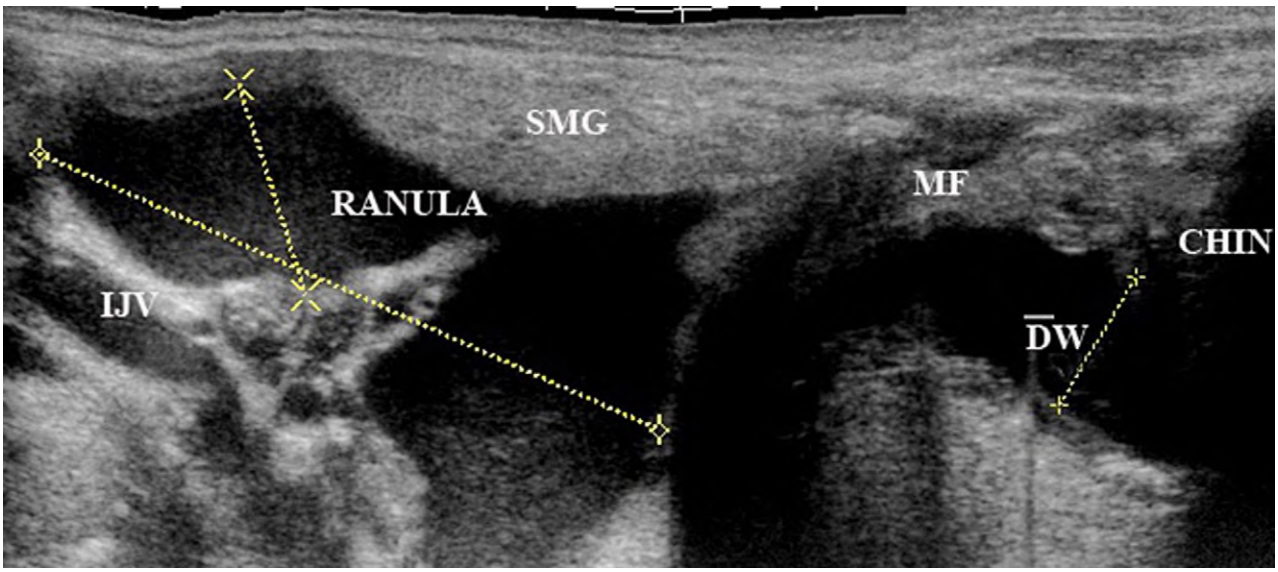

FIGURE 3. A panoramic sagittal oblique ultrasound view of a large plunging ranula occupying the submental, submandibular, and parapharyngeal spaces. DW, Wharton duct; IJV, internal jugular vein; MF, floor of the mouth; MM, mylohyoid muscle; SLG, sublingual gland; SMG, submandibular gland.

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months (mean, 52 mo). Nine patients (13.8%) had recurrences that developed within 6 months of the operation. Ultrasound examinations in these patients showed cystic structures, indicating the extent of the lesions. The other patients had unremarkable ultrasound features on the floor of the mouth during the entire follow-up period. No differences were observed in recurrences between patients with a history of trauma or possible iatrogenic injury and other patients. Treatment for recurrences was based on complete excision of the SLG in combination with ranula excision (n = 5) or alone (n = 1) and marsupialization (n = 1). A wait-and-see strategy was adopted in a 4-year-old child with a recurrent intraoral ranula in view of her age, but the patient missed the follow-up appointment and telephone contact was unsuccessful. Another patient with a recurrence was awaiting treatment at the time of writing this report. There have been no other recurrences after revision operations.

Treatments involving complete excision of the SLG were associated with the lowest rate of recurrence (3.6%), followed by partial SLG excision (9.1%), marsupialization (13.6%), and ranula excision (36.7%). Table 2 presents the statistically significant difference (P = .01) in the recurrence rates between complete SLG excision and ranula excision alone. Complications were noted in 7 patients (10.8%) and included a sensory deficit in the tip of the tongue (n = 4), which resolved within 2 weeks in 1 case, and obstructive recurrent sialadenitis (n = 3). No statistically signifi-

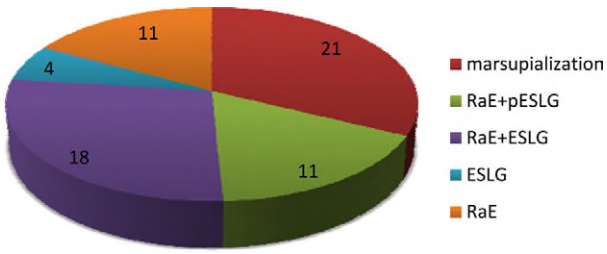

FIGURE 4. Surgical treatment for oral and plunging ranulas in 65 pediatric and adult patients from January 2001 through December 2010. ESLG, total sublingual gland excision; pESGL, partial sublingual gland excision; RaE, ranula excision; SLG, sublingual gland.

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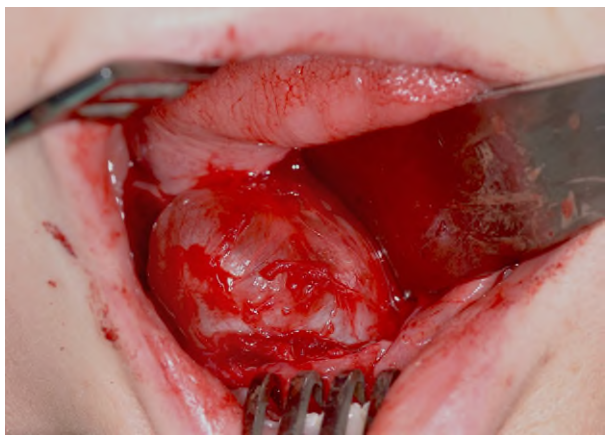

FIGURE 5. Transoral surgical approach for ranula treatment.

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FIGURE 6. Transcervical surgical approach for treating plunging ranula.

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cant differences in complication rates were observed among the different treatments.

Discussion

Several surgical approaches and techniques have been described for the management of oral and plunging ranulas, and the results have shown that SLG excision is associated with the lowest rate of recurrent disease. The present results support this view. The data were compared with those in a recent meta-analysis by Harrison[1] in Figure 7. Complete SLG excision was successful in all but 1 case (3.6%) in the present group of patients, confirming the low recurrence rate of about 2.1% with this procedure. Recurrences result from incomplete excision, with persistence of part of the SLG in a mylohyoid muscle hiatus.[1] Simple marsupialization and excision of the ranula alone were associated with higher recurrence rates, as has been reported in previous studies.[1] However, the data for marsupialization appear to be more favorable in the present study, with a recurrence rate of 13.6% ($P < .001$, binomial test).

Partial excision of the SLG together with ranula excision was also performed in the authors' department, with only 1 recurrence (9.1%). McGurk[9] also described this procedure in 12 cases, with a success rate of 100%. The procedure depends on the correct identification of the secretory unit supplying the ranula and on the technique used; decompression of the ranula was performed before excision. Other conservative approaches have been suggested in the literature in a few case series but have never been used in the authors' department; these include variations in marsupialization, intracystic injection of the sclerosing agent OK-432 (picibanil), and administration of botulinum toxin type A.[9-12]

Plunging ranulas can be managed surgically with a transcervical or transoral approach.[1,13] In the present study, including 10 plunging ranulas, the transoral approach was preferred in 7 cases (70%). The submandibular incision is associated with potential injury to the marginal mandibular branch of the facial nerve, cervical fistula formation, and cervical scarring.[2] Removal of the SLG with this method is considered difficult because it requires division of the mylohyoid muscle and dissection up to the level of the mucosa of the floor of the mouth.[6] For these reasons, an intraoral approach is preferred by many surgeons.[7,14]

Complications associated with procedures for the treatment of ranula include injury to the Wharton duct, bleeding or hematoma, wound dehiscence, and sensory deficits in the tongue.[15] Excision of the SLG is associated with a major risk of injury to the lingual nerve and injury to the Wharton duct.[8] In the present series, a persistent sensory deficit in the tongue, consisting of paresthesia or numbness at the tip of the tongue, was seen in 3 patients; episodes of recurrent obstructive sialadenitis, possibly related to Wharton duct injury causing stenosis, were seen in another 3. Only 1 of these complications appeared after simple marsupialization. However, there was no documentation of any injury to these structures in the surgical notes.

Table 2. DIFFERENT TREATMENTS FOR PATIENTS WITH RANULA IN THE DEPARTMENT OF OTORHINOLARYNGOLOGY IN ERLANGEN, GERMANY

	Treatments (n)	Recurrences (n)	Recurrence Rate (%)	Complications (n)	Previous Surgery (n)
Marsupialization	22	3	13.6	2	7
Complete sublingual gland excision	28	1	3.6*	3	9
Partial sublingual gland excision	11	1	9.1	0	6
Ranula excision alone	11	4	36.4*	2	2
Total	72	9	12.5	7	24

*There was a statistically significant difference ($P = .01$) in recurrence rates between complete sublingual gland excision and ranula excision alone.

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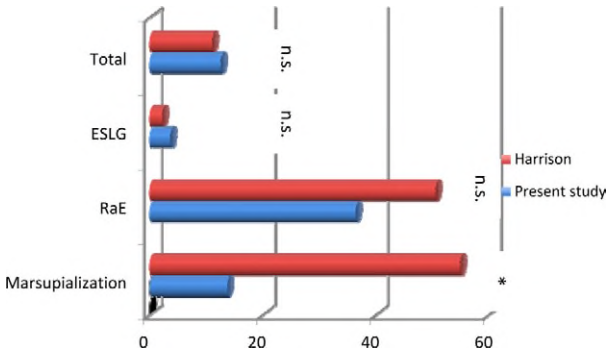

FIGURE 7. Recurrence of ranula (percentage) after different treatments in the present study compared with a recent review of the literature by Harrison.[1] *$P < .001$ (binomial test). ESLG, surgical treatment with complete excision of the sublingual gland; n.s., not statistically significant; RaE, surgical treatments with ranula excision alone.

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Sixteen patients (24.6%) in the present case series had previously undergone surgery or treatment of the floor of the mouth, which in most cases consisted of sialodochotomy procedures for stenosis or transoral removal of submandibular gland stones. Three of these patients developed a plunging ranula. There is clinical evidence that ranula can have a traumatic etiology. In an earlier histologic study, McGurk et al[16] noted a ruptured minor sublingual duct (Rivinus duct) in all 8 cases of oral ranula observed, indicating an origin from part of the lesser SLG after trauma to the floor of the mouth. Other studies have also reported an iatrogenic etiology for ranula. Ranulas were observed in approximately 5% of patients undergoing submandibular duct relocation for the management of uncontrollable sialorrhea.[8] Zhi et al[2] carried out a retrospective study on plunging ranula and noted previous surgery in 34.9% of patients, with procedures consisting of incision and drainage for submental or submandibular abscesses, submandibular gland excision, incomplete excision of the SLG, and surgery for presumed dermoid cysts.

The diagnosis of ranula was made on clinical and ultrasonographic grounds. The histologic findings confirmed, as other studies have shown,[6,17] that ranulas are extravasation mucoceles that arise from the SLG. Clinical examination provides most of the diagnostic information.[8] Ultrasound is an inexpensive, safe, and reliable method that can add further details, especially in patients with plunging ranula, and can help with differential diagnoses such as other types of cystic lesion and benign tumors and with monitoring the patients during the follow-up. Jain et al[18] investigated the value of ultrasound in the diagnosis and management of plunging ranula. The investigators reported that high-resolution ultrasound could determine the extent of plunging ranula and to confirm the cystic nature of the lesion, assess the status of the mylohyoid muscle (a defect was shown in 100% in the case series), and evaluate the SLG for rupture or herniation. In the present retrospective study, it was difficult to evaluate the incidence of defects in the mylohyoid muscle from the images recorded, and this was not well documented in all patients' files. Magnetic resonance imaging and computed tomography have also been used for differential diagnosis in some cases of plunging ranula. The presence of a "tail" is pathognomonic for plunging ranula.[19] Aspiration of mucus from the lesion and laboratory testing for amylase content should also make the diagnosis of ranula clear.[8] These findings may be useful, but they do not appear to modify the indication for surgery.

In conclusion, 9 years' surgical experience in the authors' department confirms the variety of treatments offered for oral and plunging ranulas in pediatric and adult patients. Total SLG excision is the preferred treatment method and is associated with the lowest rate of recurrent disease, as reported previously[1]; a statistically significant difference ($P = .01$) was observed in the present study compared with ranula excision alone. However, marsupialization, with a success rate higher than 85%, may be considered an alternative treatment, particularly in patients who wish to have less invasive procedures. This procedure is fast and easy to perform, with the patient under local anesthesia, and does not appear to cause complications if a second operation is needed. Effective involvement of the patients in the final surgical decision-making process is therefore recommended, with information being provided about the strengths and weaknesses of these 2 valid surgical options. Ranula excision alone should be avoided in view of the high rate of recurrences, and it is not recommended along with partial SLG excision to avoid scar formation. The latter could potentially increase the difficulty of a second operation, although no statistically significant differences were observed in the present study in relation to complications after revision surgery.

Ultrasound is recommended as the preferred diagnostic tool in patients with ranula. It provides additional information about the relation between the pseudocyst on the floor of the mouth and the SLG and mylohyoid muscle, and it helps with the differential diagnosis in patients with plunging ranula. In addition, it is an inexpensive and repeatable examination method that is useful during follow-up. Less easily accessible and more expensive diagnostic methods do not appear to alter the surgical management.

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