

Nonabsorbable intrasellar stent placement for recurrent Rathke cleft cyst: illustrative case

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BACKGROUND Rathke cleft cyst (RCC) has a recurrence rate of 10% to 22%, and preventing recurrence is challenging. For patients who experience persistent recurrence of RCC, placement of steroid-eluting bioabsorbable intrasellar stents has been rarely described. However, recurrences are often delayed, suggesting that dissolvable stents may not be successful long-term. The release of steroids in close proximity to the pituitary gland may also unintentionally influence the hypothalamic-adrenal-pituitary axis.

OBSERVATIONS The authors present a case of a 66-year-old woman with a persistently recurrent RCC who underwent drainage of her cyst with placement of a nonabsorbable intrasellar stent in the form of a tympanostomy tube. After repeat transsphenoidal drainage of her cyst, a tympanostomy T-tube was placed to stent open the dural aperture. Postoperatively, the patient's condition showed improvement clinically and radiographically.

LESSONS Placement of an intrasellar stent for recurrent RCC has rarely been described. Steroid-eluting bioabsorbable stents may dissolve before RCC recurrence and may have an unintentional effect on the hypothalamic-pituitary-adrenal axis. The authors present the first case of nonabsorbable stent placement in the form of a tympanostomy tube for recurrence of RCC. Additional studies and longer follow-up are necessary to evaluate the long-term efficacy of both absorbable and nonabsorbable stent placement.

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KEYWORDS intrasellar stent; nonabsorbable stent; recurrent Rathke cleft cyst; tympanostomy tube

Rathke cleft cysts (RCCs) arise from the pars intermedia and are present in 4% to 33% of individuals.^{1,2} The most common presenting symptoms are headaches, visual field deficits, and endocrinological abnormalities, and patients frequently require endoscopic, endonasal surgical intervention.^{3,4} Despite surgical intervention, recurrence rates range from 10% to 22%.⁵⁻⁹

It is currently believed that patients with larger cysts, cysts located primarily in the suprasellar region, and cysts with associated metaplasia on histopathology are at a higher risk for recurrence.^{9,10} Studies have suggested that aggressive removal of the entire cyst wall may be associated with lower rates of postoperative recurrence.^{11,12} However, this treatment has also been associated with higher rates of additional postoperative endocrinological abnormalities.^{6,12} Instillation of ethanol within the cyst cavity is frequently performed to cauterize the cavity, but the procedure may be ineffective or even catastrophic if there is a cerebrospinal fluid leak.¹²⁻¹⁴ Marsupialization of the cyst also has been suggested but has not been

consistently validated and may still result in challenging cases of recurrence.^{15,16}

Steroid-eluting bioabsorbable stents have been introduced and approved for the treatment of chronic rhinosinusitis because of their ability to decrease synechia formation and polyposis.¹⁷ Recently, a few cases of bioabsorbable stent placement to prevent the recurrence of cystic sellar lesions, including RCCs, have been reported.¹⁸⁻²⁰ However, we present the first case in the literature of nonabsorbable stent placement (in the form of a tympanostomy tube) for recurrent RCCs.

Illustrative Case

A 66-year-old woman presented with a headache, and a large, cystic sellar lesion was noted on magnetic resonance imaging (MRI). Endocrinological workup did not indicate any significant abnormalities. She underwent a transsphenoidal resection for decompression and drainage and was diagnosed as having an RCC. She presented again 21 months later with recurrent headaches and new

ABBREVIATIONS CT = computed tomography; MRI = magnetic resonance imaging; RCC = Rathke cleft cyst.

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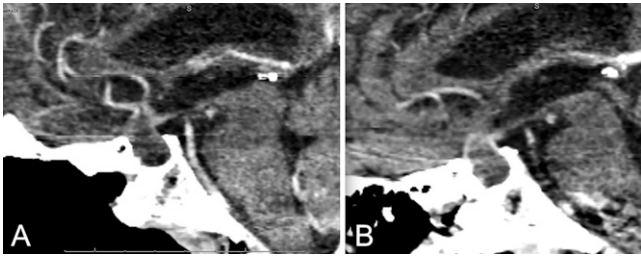


FIG. 1. CT scans with contrast. **A:** Recurrent RCC before the patient's third transsphenoidal operation. **B:** Recurrent RCC after the patient's third transsphenoidal operation.

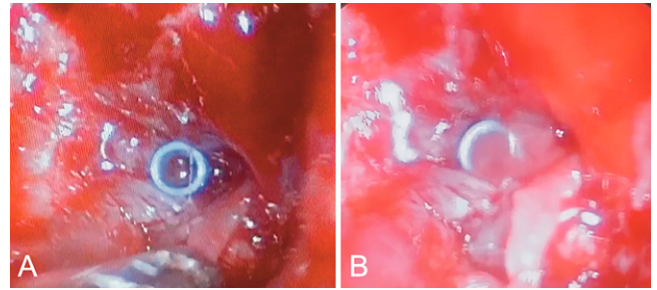


FIG. 2. A and B: Placement of tympanostomy tube within the dural aperture to the sella, with visualized drainage of cystic fluid.

bitemporal hemianopsia. Repeat MRI demonstrated recurrence of her cystic pituitary lesion with suprasellar extension. The patient underwent a transsphenoidal approach for resection, and the cyst was fenestrated to facilitate further drainage and prevent recurrence. Her bitemporal hemianopsia resolved postoperatively, and MRI obtained at her 3-month follow-up visit again demonstrated adequate decompression of the cystic lesion. In the interim, the patient required placement of a cardiac pacemaker, which prevented her from undergoing further MRI. Computed tomography (CT) scans with contrast, which were obtained at 15 months and 2, 3, and 4 years after her second surgery, continued to demonstrate no evidence of recurrence.

Five years after her second operation, the patient developed worsening headaches and mild bitemporal hemianopsia; repeat CT scans demonstrated recurrence. She underwent a repeat transsphenoidal approach for resection and drainage of her RCC. Her

vision and headaches improved, but they worsened again 1 month postoperatively, and recurrence of her cyst was discovered (Fig. 1).

The option of additional surgery was discussed in depth with the patient, including placement of an off-label stent to allow for durable fenestration and drainage of the cyst, with the goal of minimizing recurrence. The patient consented to the procedure. A standard transsphenoidal approach was performed. Milky, mucinous fluid was visualized draining from the cyst, consistent with her previous operation 2 months earlier. After the cyst was fully decompressed, a tympanostomy T-tube was placed to stent open the dural aperture. Fluid was visualized draining from the cyst cavity through the stent into the sphenoid sinus, which confirmed adequate positioning and function of the stent (Fig. 2). Postoperatively, the patient experienced improvement of her headaches and bitemporal hemianopsia (Figs. 3 and 4). CT scans demonstrated successful cyst drainage and placement of her tympanostomy tube stent. Nearly 1 year after

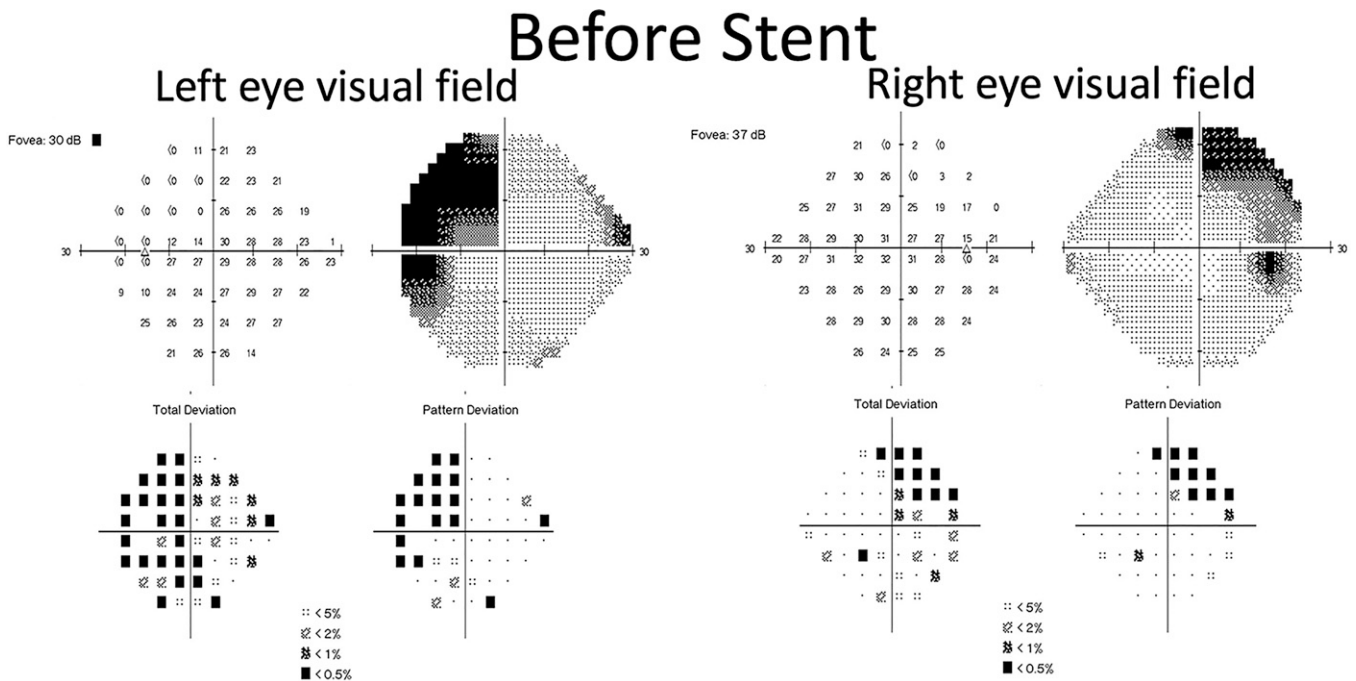


FIG. 3. Visual field schematic demonstrating preoperative bitemporal hemianopsia before intrasellar stent placement.

After Stent

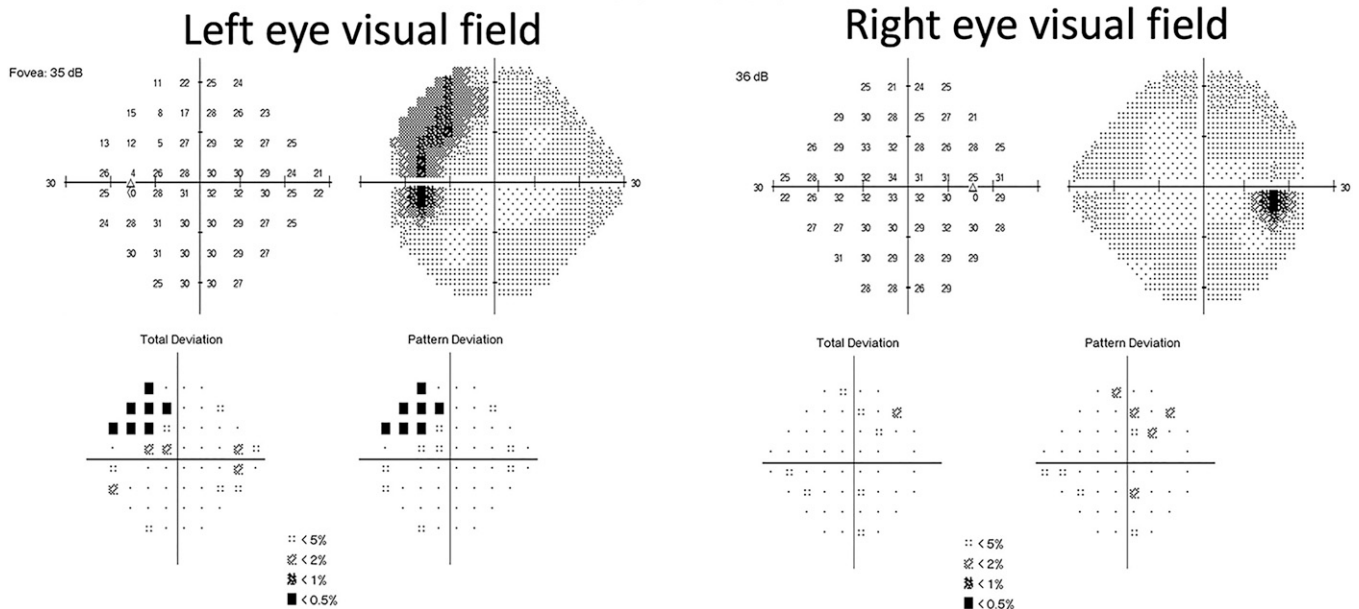


FIG. 4. Visual field schematic demonstrating improvement in bitemporal hemianopia after RCC drainage and placement of intrasellar stent.

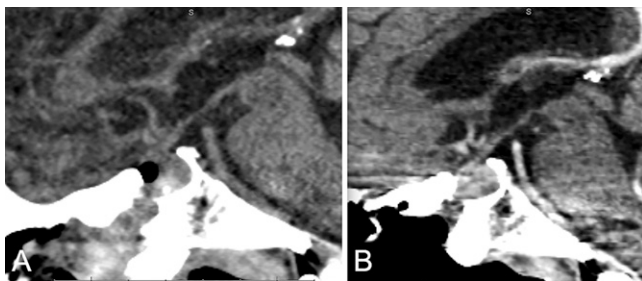


FIG. 5. CT scans with contrast. **A:** No evidence of residual RCC immediately after the procedure. **B:** No evidence of recurrent RCC 6 months postoperatively.

her operation, there was no clinical or radiographic evidence of cyst recurrence (Fig. 5).

Discussion

RCCs are benign lesions that arise from the pars intermedia but may cause debilitating symptoms.^{1,2} In such cases, surgical decompression is recommended; however, the recurrence rate remains between 10% and 22%.⁵⁻⁹ Techniques such as cyst marsupialization, aggressive removal of the cyst wall, and instillation of ethanol into the cyst cavity have all been proposed to lower recurrence rates. Unfortunately, these techniques are associated with risks and have not been clearly shown to be effective.^{6,11-16}

Placement of steroid-eluting bioabsorbable stents has been approved for chronic rhinosinusitis and recently was described as a treatment for RCC.¹⁷⁻²⁰ In those cases, the stent was placed between the

cyst cavity and the sphenoid sinus to facilitate continued drainage of the cyst into the sphenoid sinus and posterior nasopharynx, thus preventing recurrence. No complications have been reported with stent placement for patients who have undergone this procedure, although long-term follow-up results have not been obtained.

Observations

To date, the few cases of stent placement for recurrent RCC have only included bioabsorbable stents. Prior reports have demonstrated that these stents dissolve shortly after the procedure, with evidence of complete dissolution at 3 months.^{18,20} Therefore, although these bioabsorbable stents may be effective at preventing early recurrence, we hypothesize that a nonabsorbable stent may be a more effective option because of the frequency of delayed recurrence. This finding is particularly important because RCCs may recur in either early or delayed fashion.^{6,12,18} Aho et al.⁶ published a retrospective study of 114 patients who underwent complete cyst resection and showed no evidence of residual cyst on 3-month follow-up MRI. Despite this short-term success, 19 (17%) of these patients experienced recurrence in a delayed fashion. Benveniste et al.¹² described 62 patients who underwent RCC resection and noted a recurrence rate of 16% at a mean time of 35.7 ± 49.1 months postoperatively (range 1–166 months). Additionally, steroid-eluting stents have a theoretical risk of affecting the hypothalamic-pituitary-adrenal axis given their proximity to the pituitary gland. Although this risk has not been demonstrated in the few cases of bioabsorbable stents published thus far, chronic exposure of the pituitary gland to steroids may result in significant side effects.

Lessons

RCCs may be challenging to treat and often may recur. Thorough review of the literature reveals many suggestions for treatment but no known durable solutions to prevent recurrence. Furthermore, recurrence may occur in the short and long term, making prevention in these cases even more challenging. A few cases of bioabsorbable stent placement have been described for the purpose of preventing recurrence. However, these stents were fully absorbed within months, which suggests that patients may be at risk of long-term recurrence. The effect of this direct steroid exposure on the hypothalamic-pituitary-adrenal axis is also unknown but may result in significant side effects.

Placement of nonabsorbable stents may provide a durable long-term solution for recurrent RCCs not amenable to other treatment options, and we present the first case describing this procedure. Although the use of nonabsorbable stents is likely to avoid many of the issues described with absorbable stents, they may be associated with a different series of possible complications and side effects. It is possible that nonabsorbable stents have higher rates of infection, inflammation, stent migration, and long-term side effects because of their permanent nature. However, this case study suggests that they can be placed safely and may prevent recurrence of RCC in the short term. Additional studies and longer follow-up are necessary to further evaluate the long-term efficacy of both absorbable and nonabsorbable intrasellar stent placement for recurrent RCC.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Miller. Acquisition of data: Ellens, Miller, Williams, Vates. Analysis and interpretation of data: Williams. Drafting the article: Ellens. Critically revising the article: Ellens, Williams, Vates. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Ellens. Administrative/technical/material support: Vates. Study supervision: Miller, Vates.

Supplemental Information

Previous Presentations

This work was previously presented in poster form at the University of Rochester Schwid Symposium and Poster Session, Rochester, NY, June 19, 2020.

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