

Management of two osteomas in the frontoethmoidal region using an osteoplastic flap, endoscopic approach and 3D CT imaging

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Abstract: The aim of this study was to demonstrate the application of preoperative three-dimensional (3D) computed tomography (CT) reconstruction in the surgical management of paranasal sinus (PNS) osteomas. Osteomas are the most common benign PNS tumors and can lead to significant symptoms, including chronic rhinosinusitis, frontal pressure, and orbital or intracranial complications. Surgical excision is recommended for symptomatic cases or those with potential complications, with endoscopic and external approaches being the primary techniques. A 74-year-old patient presented with symptoms of chronic rhinosinusitis, including nasal obstruction and anosmia. CT revealed osteomas in the left ethmoid and frontal sinuses, measuring 27 × 21 × 28 mm and 15 × 17 mm, respectively. The patient was qualified for surgical excision of lesions. Preoperative planning included 3D CT reconstruction to assess the osteomas' size, location, and surgical access, particularly for a planned osteoplastic flap (OPF). During surgery, an endoscopic approach was initially utilized, resulting in frontosphenoidectomy and removal of ethmoid osteoma. The other osteoma extended from the anterior to the posterior wall of the frontal sinus and required conversion to an external approach using the preplanned OPF. The surgery was successfully completed, with intraoperative findings being consistent with imaging. Postoperative complications were limited to frontal sinus outflow stenosis, managed endoscopically during follow-up. PNS osteomas may require complex surgical planning, especially when endoscopic and external approaches are combined. Preoperative 3D CT reconstruction provides valuable insights into tumor characteristics and aids in precise surgical planning. This case highlights the utility of 3D imaging in safe and effective management of challenging PNS osteomas.

Keywords: paranasal sinus osteoma, 3D computed tomography, osteoplastic flap, endoscopic sinus surgery, frontal sinus surgery, surgical planning, sinus tumor.

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Introduction

Osteomas are benign bone tumors and with prevalence rate of 3% in patients presenting with chronic rhinosinusitis, they constitute the most common benign tumors of the paranasal sinuses. The frontal and ethmoid sinuses are, respectively, the first and the second most frequent location of osteomas of the paranasal sinuses. [1]

The first symptoms of paranasal sinus (PNS) osteoma usually appear in the third to sixth decade [2] and vary depending on size and localization. Osteomas of the frontoethmoidal region are usually associated with earlier occurrence of symptoms. Possible manifestations involve frontal pressure, headache, chronic rhinosinusitis, mucocoele, facial deformities, proptosis, periorbital pain and epiphora. Most osteomas are, however, an incidental asymptomatic finding.

Surgical excision is recommended for symptomatic osteoma and in certain cases also for asymptomatic osteoma. [1] Possible surgical methods of excision comprise endoscopic approach, external approach and combination of both. [3, 4] The decision concerning the approach is made based on characteristics of the osteoma, its location and direction of extension, as well as experience of the surgeon. [1, 3] Osteoplastic flap is a well-established technique belonging to external approaches. However, due to significant morbidity its use is nowadays limited and endoscopic procedures are preferred method of management of PNS osteoma in most patients with favorable anatomy. [5] The imaging methods, including three-dimensional 3D reconstruction, provide a more precise characterization of osteoma's size, location and relation to the surrounding tissue, playing an important role in planning appropriate surgical approach. [4, 6]

Multiple osteomas might be associated with Gardner's syndrome, an inherited autosomal dominant condition, a variant of familial adenomatous polyposis (FAP) [6] with the incidence estimated to be 1 in 8300 to 16000 in American population. [2] The manifestations of Gardner's syndrome include cutaneous and soft tissue tumors, osteomas, supernumerary or unerupted permanent teeth and multiple intestinal polyps with propensity for malignant transformation. [7]

Development of colon polyps begins in the second decade, with the average age at diagnosis being 22 years. Without treatment, the risk of malignant transformation reaches almost 100% in affected patients and is usually observed in individuals aged 30 to 50 years. [8] Development of clinically significant osteomas precedes the diagnosis of FAP by an average of 17 years. As an early diagnosis and surgical treatment prevent the malignant transformation of the polyps, it is beneficial for patients with osteomas to undergo assessment for exclusion of the Gardner's syndrome. [2]

The aim of the study is to demonstrate the application of preoperative 3D planning with the use of 3D computed tomography reconstruction in removal of ethmoid and frontal sinus osteomas via endoscopic and osteoplastic flap approach.

Case presentation

A 74-year-old man presented with symptoms suggestive of chronic sinusitis, including nasal congestion and anosmia. Patient reported no gastrointestinal symptoms. Findings from CT imaging performed 1 year previously were indicative of ethmoid and frontal sinus osteomas. Nasal examination was significant for obstruction and polyps in the left nasal cavity. No pathological discharge was found. CT scan of the head was performed and revealed osteomas in the left ethmoid and frontal sinus, measuring $27 \times 21 \times 28$ mm and 15×17 mm, respectively (Fig. 1, 2). The CT scan description suggested osteomas' probable association with Gardner's syndrome. Patient's

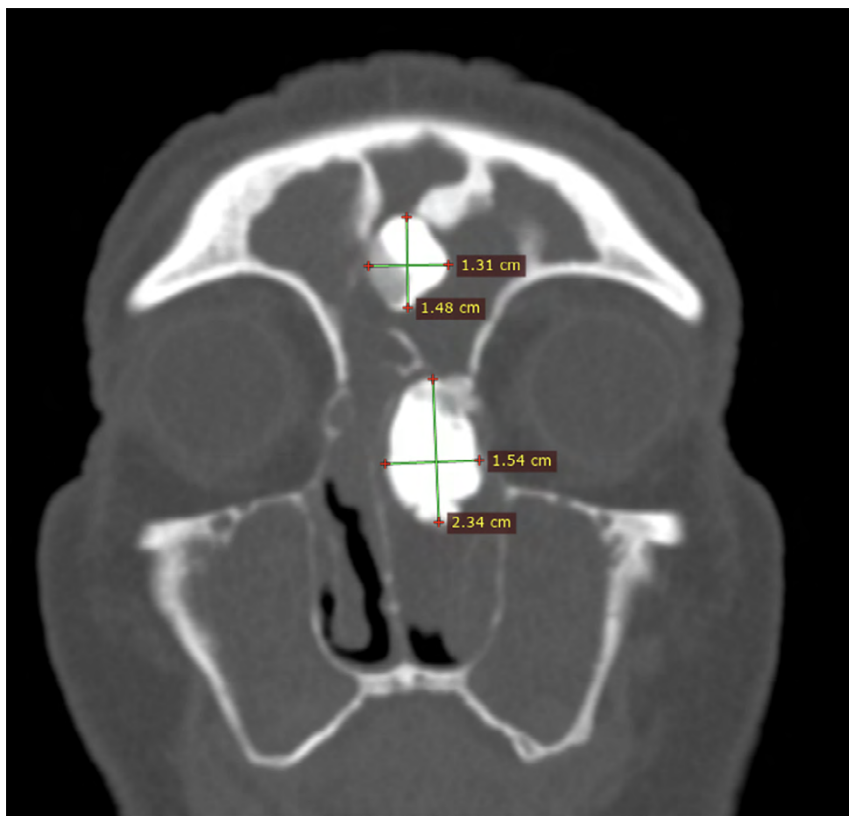


Fig. 1. Computed tomography image of osteomas in ethmoid and frontal sinus with diameter of the osteomas measured in coronal projection.

history was remarkable for bilateral polypectomy and bilateral ethmoidectomy performed, respectively, 26 and 17 years previously.

The patient was qualified for surgical excision of lesions in ethmoid and frontal sinus. The pre-surgical operation planning first involved generating a three-dimensional computed tomography (3D CT) reconstruction, using dedicated software (RadiAnt). Based on it, the team of operators determined and visualized the image of the planned anterior osteoplastic flap with its exact measurements (Fig. 3, 4). Operators also used 3D CT reconstruction images to plan the optimal external access to the frontal sinus osteoma in case the lesion was not amenable to an endoscopic approach.

Using the endoscopic approach, polypectomy was performed in the left nasal cavity, enabling the identification and dissection of oval-shaped osteoma, with dimensions consistent with those found in the CT. Considering the diameter of the lesion, it proved necessary to cleave it into 2 fragments for safe extraction through the left nostril. Frontosphenoidectomy was performed. Examination of the frontal sinus revealed the other osteoma, consistent with the CT scan description. The nasal septum was dislocated. The endoscopic examination of the right nasal cavity and the sinuses included polypectomy, followed by frontoethmoidectomy. The endoscopic approach



Fig. 2. Computed tomography image of osteomas in ethmoid and frontal sinus with diameter of the frontal sinus measured in sagittal projection.

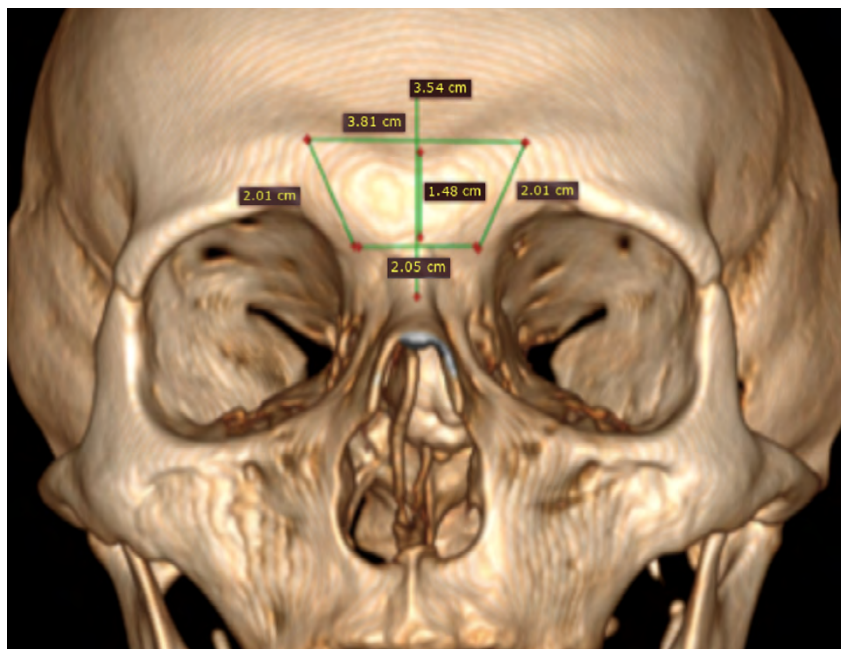


Fig. 3. The three-dimensional computed tomography reconstruction showing the frontal osteoplastic flap which was planned to be removed during surgery, including its exact measured diameter.

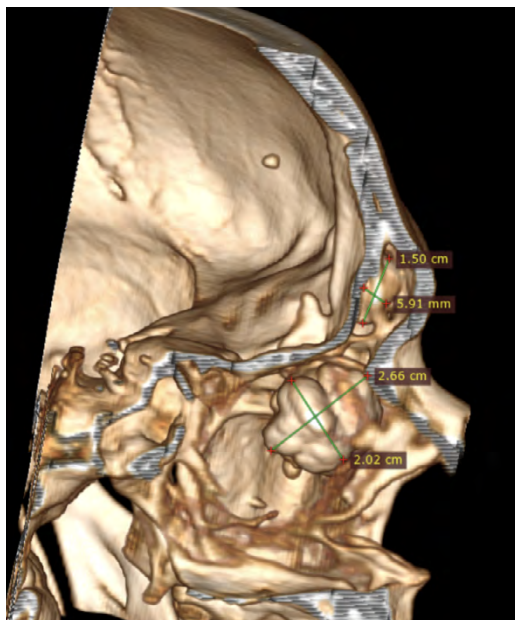


Fig. 4. The three-dimensional computed tomography reconstruction showing the ethmoid and frontal sinus osteomas with measured diameters in sagittal projection.

provided bilateral assessment of the frontal ostium leading to the decision to extract the frontal sinus osteoma using the external approach.

T-shaped incision above the frontal sinuses was made and followed by the removal of the trapezoidal bone flap in accordance with the preoperative plan (Fig. 5, 6), which provided access to the frontal sinuses. Examination of the frontal sinuses revealed the osteoma located in the median line, measuring approximately 30×20 mm, connected to both anterior and posterior wall. Using the

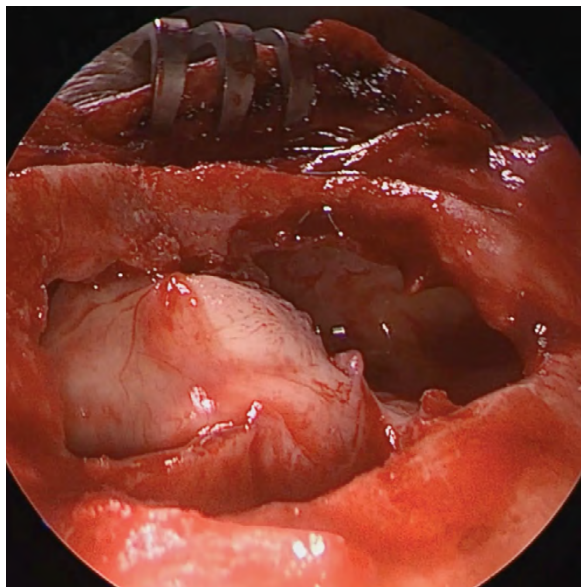


Fig. 5. Intraoperative image of the frontal bone including exposed frontal sinuses and osteoma after the removal of the frontal bone flap.

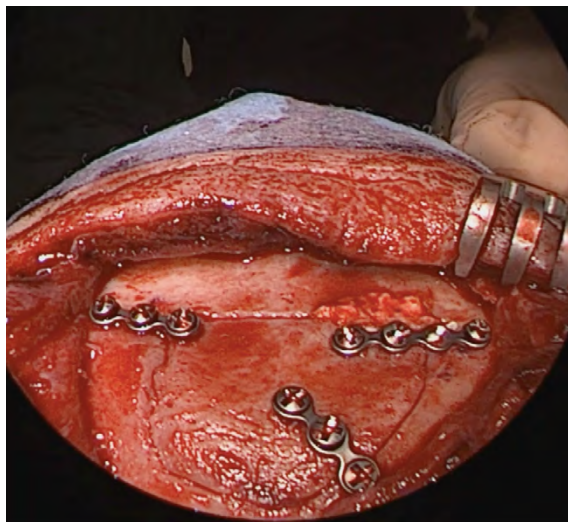


Fig 6. Intraoperative image of the frontal bone after securing the frontal bone flap in place with plates and screws.

cutting burr, the lesion was separated from the anterior wall, allowing it to be extracted. Polyps in the frontal sinus were exposed and removed. The openings of frontal sinus connecting frontal sinuses to the nasal cavity were widened as in the endoscopic modified Lothrop procedure, providing appropriate drainage. The bone flap was replaced and secured in place with plates and screws. The wound drainage and the anterior nasal packing with steroid were kept for 72 and 16 hours, respectively.

Pathological examination confirmed the lesion removed from the frontal sinus was an osteoma. At the follow-up visits 2 weeks and 4 months after the surgery endoscopic examination revealed obstruction on the left side of the frontal sinuses opening. The opening was widened, no pathological discharge was detected. The follow-up examination was otherwise insignificant. The patient was referred to a gastroenterologist for further examination regarding the suspicion of Gardner's syndrome.

Discussion

Osteoma is the most common benign neoplasm of the nose and paranasal sinuses [3] and the most common primary bone tumor in the craniofacial region. Apart from the paranasal sinuses, osteomas develop in maxilla, mandible, mastoid sinus, external auditory canal, the cranial vault, [9] and rarely outside the head and neck region. [3] Aksakal *et al.* [10] calculated the prevalence rate of paranasal sinus (PNS) osteoma in patients who underwent paranasal sinus, maxillofacial computed tomography (CT) and brain CT angiography to be nearly 1%. The frontal and ethmoid sinuses were the most frequent locations. The study conducted by Erdogan *et al.* [9] determined the prevalence rate PNS osteoma in patients undergoing CT scans for sinus symptoms to be 3% with frontal and ethmoid being, respectively, the first and second most frequent location. Semerci *et al.* [11] in a recent study estimated the prevalence of PNS osteomas in patients undergoing cone beam CT tomography for various dental indications to be 7%, with ethmoid sinus constituting the most frequent location, followed by the sphenoid and frontal sinus in decreasing order. In the forementioned studies the size of osteomas varied from 1.15 mm to 50 mm. [9, 10, 11]

The spectrum of clinical manifestations of PNS osteomas is wide and depends on size, location and direction of extension. As most PNS osteomas are asymptomatic, they are frequently identified incidentally in imaging studies or other clinical procedures, [1] similarly to bony defects of the posterior and middle cranial fossa [12] and other lesions in the sinonasal and orbital region. [13] The literature states that only 4 to 10% of all osteomas produce clinical symptoms, with frontal pressure, headache and chronic rhinosinusitis being the most common. Other manifestations include mucocele, external deformities and symptoms resulting from orbital or intracranial extension. Slow growth of osteomas, however, usually precludes development of manifestations other than headache and rhinosinusitis. [1] The peak incidence of PNS osteoma varies in literature and is stated to be in the third [9] and seventh [14] decade.

Multiple osteomas are rare. Aksakal *et al.* [10] reported that multiple osteomas were found in 24 out of 176 patients with at least one osteoma. Erdogan *et al.* [9] stated that in 4 out of 57 cases osteomas were found in more than one sinus. Presence of multiple osteomas might be associated with Gardner's syndrome, an autosomal dominant disease genetically indistinct from familial adenomatous polyposis (FAP), characterized by a triad of symptoms comprised of osteomas, mostly of the mandible and PNS, cutaneous and soft tissue tumors, and intestinal polyposis with propensity for malignant transformation. The literature suggests that development of clinically significant osteomas precedes the diagnosis of FAP by 17 years. Thus, it is crucial for patients diagnosed with PNS osteomas to undergo further examination for Gardner's syndrome. [2]

In this case, 71-year-old patient presented with two PNS osteomas, one located in the left ethmoid sinus and the other in the left frontal sinus, in the median line. The sizes were stated in the description of the CT scan as $27 \times 21 \times 28$ mm and 15×17 mm, respectively. The symptoms present on admission involved nasal congestion, more pronounced on the left side, and anosmia. The symptoms were suggestive of chronic sinusitis and therefore consistent with the most common symptoms of osteomas described in the literature. [1] The patient did not, however, report headache or frontal pressure, which are considered the most common symptoms. The literature states that in individuals affected with Gardner's syndrome the risk of developing colon cancer is nearly 100% by the age of 40. [3] In this case, the patient was 74 at the time of admission to the hospital and presented with no symptoms associated with colon cancer. Nonetheless, the patient underwent further gastroenterological evaluation, and the diagnosis of Gardner's syndrome patient was ruled out.

Indications for surgical excision of an osteoma include symptomatic osteoma (following the exclusion of other explanations for the symptoms), osteoma associated with current complication, and under certain conditions asymptomatic osteoma. [1, 3] As far as asymptomatic osteoma is concerned, Georgalas *et al.* [1] recommends excision of a growing osteoma, an osteoma extending more than 50% of the frontal sinus and an osteoma associated with imminent complications, involving complete obstruction of the frontal recess and intraorbital or intracranial extension. In a more recent study, Sofokleous *et al.* [3] extended those indications to include all osteomas located in the ethmoid sinus, in proximity to frontal recess or in the sphenoid sinus, regardless of their size and lack of symptoms. In this case, the patient presented with symptoms suggestive of chronic rhinosinusitis, associated with two osteomas located in frontal and ethmoid sinus and was therefore qualified for surgical resection of both osteomas.

Surgical resection of an osteoma of the frontoethmoidal region can be performed via an external (open) approach, endoscopic approach, and combined external and endoscopic approach. Since endoscopic approach provides an excellent visualization of the operating field, including the posterior wall of the frontal sinus, anterior ethmoid sinus and orbital walls, [15] and open approach

exposes the patient to longer recovery course and higher complication rates, the endoscopic approach is advisable whenever applicable. [3] Thus, throughout the years attempts have been made to determine the criteria for identification of frontal sinus osteomas amenable to exclusively endoscopic approach. [1] A grading system proposed by Chiu *et al.* [16] is widely used in literature. The authors recommend endoscopic approach for management of only grade I and II osteomas, which include osteomas medial to a virtual sagittal plane through the lamina papyracea with base of attachment posterior-inferior along the frontal recess, and external or combined approach for management of grade III and IV osteomas. Grade III osteomas are defined as those with base of attachment located anteriorly or superiorly within the frontal sinus, or as tumor extending lateral to a virtual sagittal plane through the lamina papyracea. Grade IV osteomas are referred to as tumors filling the entire frontal sinus. According to the literature grading system by Chiu *et al.* [16] has now been surpassed by the current practice, and the limitations of exclusively endoscopic approach are still being explored. [1, 3] In a recent study Sofokleous *et al.* [3] proposed a grading system that classifies grade I and II tumors as manageable through an endoscopic approach and leaves the feasibility of endoscopic resection of grade III tumors at the discretion of the surgeon, as grade III tumors require appropriate training and experience. Grade I tumors according to Sofokleous *et al.* [3] involve former grade I and II tumors, grade II corresponds to former grade III, grade III involves former grade IV tumors and those with a point of attachment in far-lateral aspect of a well-pneumatized frontal sinus. Grade IV tumors are referred to as osteomas with certain characteristics, which makes them currently unamenable to exclusively endoscopic approach. As far as ethmoid osteoma is concerned, anterior extension is considered a limitation of the endoscopic approach. Additionally, extension anteriorly to the nasolacrimal duct and under the skin requires a combined approach. [1]

Endoscopic management of frontal sinus diseases, including osteoma, can be achieved through procedures first described by Draf, [17] which allow opening the frontal sinus outflow more widely. The procedures vary in extensiveness, with Draf III being the most extensive and involving expansion of the frontal recess, superior septectomy and establishing common frontal sinus cavity. Draf III can be a method of choice in management of severe polyposis, benign or malignant tumors and conditions related to trauma or in patients with specific anatomic variations. [18] On the other hand, osteoplastic flap (OPF) technique, belonging to external approaches, for decades was considered the gold standard in the management of frontal sinus diseases. Since the popularization of endoscopic sinus surgery, the role of OPF is mostly reduced to managing frontal sinus diseases in patients with unfavorable anatomy or complications due to previous surgeries. Frontal OPF can be performed with or without obliteration of the frontal sinus. The latter usually includes not only abstaining from occluding the frontal recess, but also widening it to provide drainage to the nasal cavity, restoring physiological function of the sinus. [5]

In this case, three-dimensional (3D) computed tomography (CT) reconstruction was used during preoperative planning for assessment of the frontal sinus osteoma and the anatomy of the frontal sinus. Considering osteoma's anterior attachment within the frontal sinus, the frontal sinus osteoma could be characterized as at least grade III according to Chiu *et al.* [16] or grade II according to Sofokleous *et al.*, [3] classifying it as amenable to endoscopic approach, provided the surgeon had appropriate experience and training. Given osteoma's both anterior and posterior attachment to the frontal sinus and patient's anatomy, a decision was made to perform left frontosphenoidectomy and resection of the left ethmoid osteoma, followed by intraoperative assessment of the frontal ostium to determine if conversion to osteoplastic flap was needed.

3D CT reconstruction was used to determine appropriate size, location and precise measurements of planned frontal OPF. Intraoperative bilateral assessment of the frontal ostium resulted in conversion to the external approach, which was performed according to preoperative 3D reconstruction. The frontal OPF was performed without obliteration of the frontal sinus and appropriate drainage from the sinus to the nasal cavity was provided during the endoscopic part of the surgery.

Despite being a well-established procedure, which is in use for more than 40 years, OPF technique is associated with substantial morbidity. The literature lists dural injury, bone flap fracture and orbital fat exposure as the most common intraoperative complications of OPF. [5] The most significant postoperative complications involve frontal sinus outflow stenosis, mucocele, frontal pain, paresthesia, anesthesia and impaired cosmesis. [1, 5] In a study conducted by Pagella *et al.* [5] frontal sinus outflow stenosis was present in about 36% of patients. Both frontal outflow stenosis and mucocele may be managed with endoscopic endonasal salvage procedure. Similarly, the most common complication related to Draf III procedure is frontal sinus outflow stenosis. Its likelihood might be reduced by regular debridement and intranasal steroid use. [18] In this case, no intraoperative adverse events occurred. Endonasal examination during follow-up visits 2 weeks and 4 months after the surgery revealed left frontal sinus outflow stenosis, which was managed endoscopically.

Conclusions

Paranasal sinus osteoma is the most common tumor of the paranasal sinuses and might produce significant symptoms and complications, including chronic rhinosinusitis and symptoms related to orbital or intracranial extension. Whenever applicable, surgical management of paranasal sinus osteomas might require meticulous preoperative planning, which in certain cases might result in external approach being safer for the patient than endoscopic approach. In this case, successful excision of ethmoid and frontal osteomas was performed. The latter was managed with osteoplastic flap technique, preceded by thorough preoperative planning with the application of 3D CT reconstruction for determining the location and measurements of the bone flap. It resulted in successful excision with no intraoperative adverse events and postoperative complications limited to frontal sinus outflow stenosis.

Author contributions

Conceptualization: M.R., M.P., P.H., M.K. and J.T.; methodology: M.R., M.P. and J.T.; validation M.R., M.K. and J.T.; resources: J.H., M.M. and M.B.; writing—original draft preparation: M.R., J.H., M.M., J.P., K.M. and M.B.; writing—review and editing: M.R., M.K., P.H. and J.T.; visualization: J.H., M.M., K.M. and M.B.; supervision: M.R., M.P., P.H., M.K. and J.T.; project administration: J.H., M.M., M.B., J.P. and K.M.; funding acquisition: J.P., K.M. and J.T. All authors have read and agreed to the published version of the manuscript.

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Conflict of interest

None declared.

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