The Evidence-based Management of Bilateral Ethmoid Osteomas: Diagnosis, Endoscopic Resection and Review of the Literature

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ABSTRACT

A 56-year old woman presented with headaches and nasal congestion secondary to bilateral ethmoid osteomas and she made a full recovery following endoscopic resection. Paranasal osteomas are common, benign, encapsulated tumours that are often asymptomatic, but may present similarly to rhinosinusitis and occasionally cause complications depending on their anatomical location. Bilateral ethmoidal osteomas, however, are a rare occurrence and have not been previously reported in the English literature as far as the authors are aware. Ethmoidal and fronto-ethmoidal osteomas, like other paranasal osteomas, are easily diagnosed using computed tomography and are increasingly amenable to endoscopic resection. An evidence-based management plan is presented.

Keywords: Ethmoid, osteoma, paranasal

Tratamiento Basado en la Evidencia para el Osteoma Etmoidal Bilateral: Diagnóstico, Resección Endoscópica y Revisión de la Literatura

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RESUMEN

Una mujer de 56 años se presentó con los dolores de cabeza y congestión nasal secundaria a un osteoma etmoidal bilateral, tras su recuperación total a partir de una resección endoscópica. Los osteomas paranasales son tumores comunes, benignos, y encapsulados, a menudo asintomáticos, pero que pueden presentarse de modo similar a la rinosinusitis, y ocasionalmente causar complicaciones, en dependencia de su localización anatómica. Los osteomas etmoidales bilaterales, sin embargo, son una ocurrencia rara y hasta donde sabemos, no ha sido reportado con anterioridad en la literatura en inglés. Los osteomas etmoidales y fronto-etmoidales – al igual que otros osteomas paranasales – se diagnostican fácilmente usando CT, y son cada vez más tratables mediante resección endoscópica. Se presenta un plan de tratamiento basado en la evidencia.

Palabras claves: Osteoma etmoidal, paranasales

West Indian Med J 2010; 59 (2): 188

INTRODUCTION

Osteomas are benign and are the most common neoplasms of the nose and paranasal sinuses (1). They resemble normal bone histologically, contain varying amounts of fibrous tissue and occur as two variants: round, lobulated, ivory-like cortical osteomas, and less dense, cancellous osteomas which contain cancellous bone and fibro-fatty marrow in abundance (2, 3). They are thought to arise during embryologic development or secondarily to trauma or infection. A combination of the latter two is thought to be most likely (4, 5).

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CASE REPORT

A 56-year old Caucasian woman was referred by her general practitioner to our clinic with a four-month history of facial pain centred on the nasal bridge associated with increasing nasal stuffiness and rhinorrhoea. She denied anosmia, epistaxis and ocular symptoms. There was no clinical evidence of proptosis or facial swellings. Endonasal examination revealed a mildly rhinitic mucosa but was otherwise unremarkable. There was no other relevant medical history.

Computed tomography (CT) of the paranasal sinuses was reported as showing mild mucoperiosteal thickening of

the right sphenoid sinus and moderate disease within the ethmoid air cells, the frontal sinus and the right maxillary antrum, with involvement of both middle turbinates and osteomeatal complexes. Two well-defined, rounded, calcified lesions were seen lying within the anterior ethmoid sinuses bilaterally, the left measuring 0.7 cm and the right 0.8 cm. They were identified as ethmoid osteomas (Figs. 1, 2)

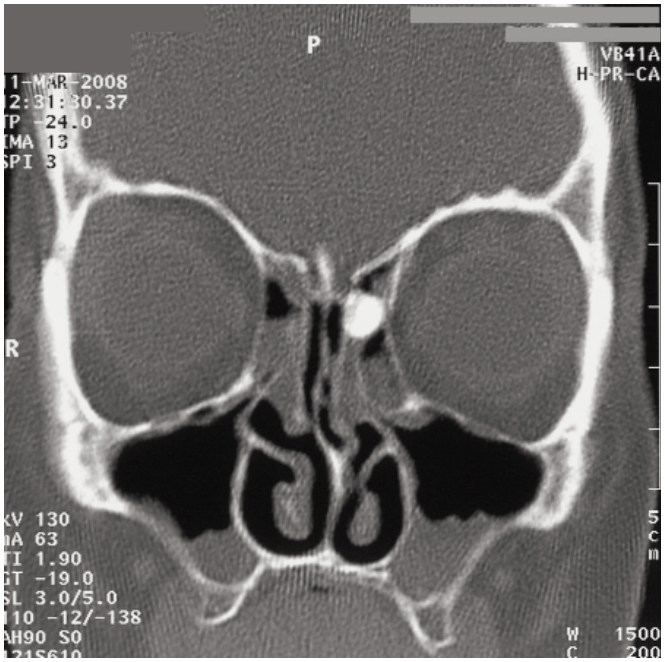

Fig. 1: Coronal CT scan of paranasal sinuses showing osteomas of left ethmoid sinuses.

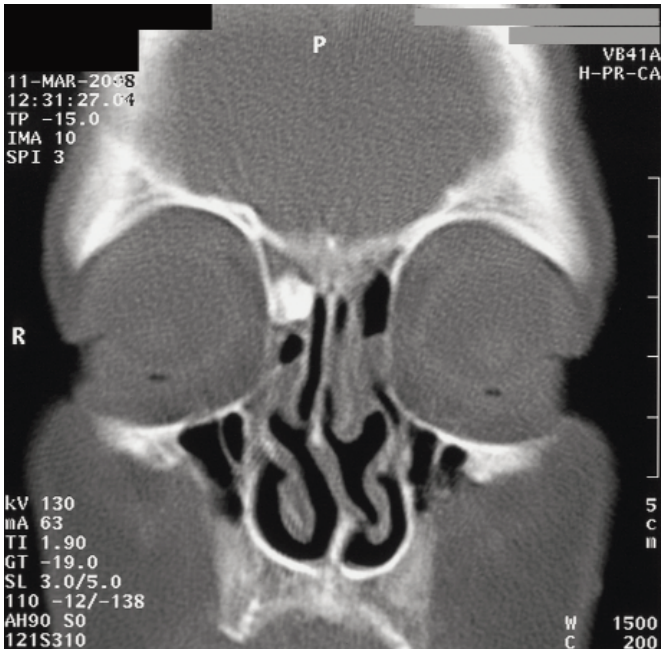

Fig. 2: Coronal CT scan of paranasal sinuses showing osteomas of right ethmoid sinuses.

Under general anaesthesia, endoscopic sinus surgery was performed. The left osteoma was found to be situated in the anterior ethmoid air cell adjacent to the lateral process of the crista galli, and the right one within the middle ethmoid air cell. Neither was attached to the lamina papyrecea. Polyps were found within the ethmoid air cells bilaterally. A middle meatal antrostomy and spheno-ethmoidectomy were performed bilaterally. The osteomas were removed in an antero-posterior fashion by using a curette. Biopsies of the osteomas and polyps were sent for pathology. The osteomas revealed a well-circumscribed nodule of mature bony trabaculae with evidence of fatty marrow spaces in keeping with an osteoma (Fig. 3). The polyp was shown to be inflam-

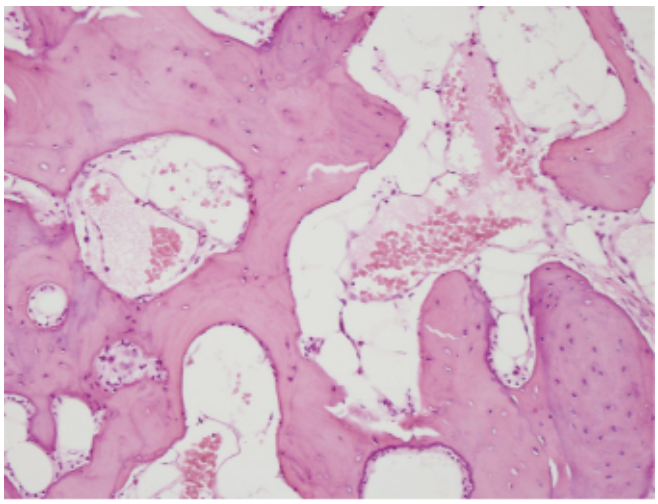

Fig. 3: High power histopathological slide of osteoma (H&E x 100).

matory in nature by demonstration of inflamed, oedematous and ulcerated respiratory mucosa.

Postoperatively, the patient was advised on nasal irrigation with saline and the use of topical nasal mometasone furoate monohydrate (Nasonex®, Schering Corporation, USA) long term. She made a good recovery with complete resolution of her symptomatology.

DISCUSSION

Osteomas tend to occur slightly more frequently in males, with a ratio of 1.3 to 1, and usually within the fifth to sixth decades (6). They are most commonly found in the frontal sinus, followed secondly by the ethmoid sinus, but are a relatively rare occurrence in the maxillary and sphenoid sinuses (7, 8). The occurrence of multiple osteomas in the facial skeleton may occur in Gardener's syndrome together with other stigmata, namely intestinal polyps, cutaneous fibromas, epidermal cysts and impacted permanent or supernumerary teeth (9). The presence of these should prompt further investigation.

Most osteomas are clinically asymptomatic and present as an incidental finding on a plain sinus radiograph (1%) or CT [3%] (1). When symptoms do arise, the most common

complaint is headache or facial pain, usually localized to the area of occurrence. There is some doubt, however, about whether paranasal osteomas can cause these symptoms in the absence of concomitant sinus disease, since most osteomas present incidentally (6). Other symptoms include facial deformity, symptoms of rhinosinusitis and occasionally ocular symptoms such as epiphora and orbital cellulitis (1, 10). Osteomas occurring in the ethmoid sinus tend to present earlier than others, possibly due to the constricted anatomy of the ethmoid air cells (11). The occurrence of bilateral ethmoid osteomas is rare.

Because osteomas are continually enlarging tumours, they may eventually cause complications due to involvement of surrounding anatomical structures. Fronto-ethmoidal osteomas are most commonly involved with such complications. Orbital involvement is possible with complications such as proptosis, periosteal orbital abscess and dacrocystitis having all been described (12–15). Fronto-ethmoidal osteomas have also been associated with spontaneous cerebrospinal fluid leaks and pneumocephalus due to erosion of the skull base and breach of the dura mater (16, 17). Nelson *et al* advise caution during lumbar puncture if pneumoencephaly is found radiologically (18). Other intracranial complications include meningitis, mucocoeles and abscesses (19–21). A preoperative CT scan, including coronal and axial images, is very helpful in determining the sites of the osteoma attachment and any co-existing complications, and aids in planning the operation (22). Increasingly, surgical intervention is being performed endoscopically, though limitations exist. Recurrence, though rare, has been reported (2).

The management of pasanasal osteomas, especially those of the fronto-ethmoidal region, remains controversial. There is conflicting opinion regarding not only when they should be operated on but what surgical procedure is ideal.

Traditionally, it was thought that frontal osteomas should be removed before they became symptomatic or caused complications (23) but the current trend is to remove them only when symptomatic, if a complication occurs, or if they fit certain radiological criteria. With respect to fronto-ethmoidal osteomas, Savic and Djeric suggest removal for osteomas extending beyond the boundaries of the frontal sinus, for continually enlarging osteomas, for those localized in the region of the frontonasal recess and those associated with chronic sinusitis (24). They also state that all ethmoid osteomas should be removed regardless of size. Hehar *et al* modify this list by suggesting that the following indications should justify fronto-ethmoidal osteoma removal: cosmetic deformity, frontonasal recess obstruction with evidence of sinus disease either by history related to upper respiratory tract infection or by mucosal disease apparent on CT, displacement of the orbital contents and mucocoele formation (6). The index patient fit these criteria for removal on three accounts: the osteomas were ethmoid in origin and there was the concurrent presence of mucosal disease apparent both on CT scan and endoscopically.

In deciding the surgical approach to an osteoma, its size and location are important, as is the surgeon's experience (24). Giant osteomas are considered those exceeding a size of 3 cm in diameter (25). Historically, an external approach *via* the use of an osteoplastic flap has been the method of choice for excision of fronto-ethmoidal osteomas (26). With the advent of endoscopic sinus surgery, a minimally invasive approach has gained popularity mainly because of its decreased morbidity and superior cosmetic advantage. In 2001, Schick *et al* suggested that an endonasal approach should be used for fronto-ethmoidal osteomas if sufficient frontal sinus access can be achieved endonasally; the osteoma is placed medial to a virtual sagittal plane through the lamina papyracea; the tumour base is at the inferior part of the posterior frontal sinus wall (27). These parameters have been shown to result in successful outcomes in two recent studies (28, 29). With regards to frontal osteomas, while some still favour an osteoplastic frontal sinus approach *via* a coronal incision if an external approach becomes necessary, as in the case of large osteomas. Dubin and Kuhn have further shown that endoscopic resection of frontal osteomas can be achieved with preservation of the natural sinus outflow instead of obliteration of the sinus, even with large osteomas (30).

Once an endoscopic approach has been embarked upon, resection of the osteoma may be carried out with the use of intranasal drill or *via* curettage (1). The use of a sonopet omni-ultrasonic bone curette has also been recently described (31). The left and right osteomas in the case in this report were 0.7 cm and 0.8 cm respectively and therefore considered small in size. We thus considered them amenable to endoscopic removal. Their ethmoid location allowed resection *via* a trans-ethmoidal approach lateral to the middle turbinate. The use of a curette proved a safe and successful method of removal in this case, more so because of non-involvement of the lamina papyracea.

CONCLUSION

Overall, paranasal osteomas are relatively uncommon but may have important consequences. With endoscopic surgery becoming increasingly popular in rhinology, most osteomas will be removed *via* this approach, though there will be those that are ideally suited to a more traditional external approach. The evidence presented was mainly based on large case series and therefore only represented Level 4 evidence (Oxford Centre for Evidence-Based Medicine Levels of Evidence, May 2001). This, however, is due in part to the lack of large numbers of these types of cases, and to the improbability of comparing the surgical treatment of these cases to a medical intervention or placebo. While each individual case should be treated on its own merit, it is still useful to have clear evidence-based guidelines to aid in the

decision-making process. This helps to ensure optimal patient care based on published, peer-reviewed prior knowledge.

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