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Transcanal sound recordings as a screening tool in the clinical management of patients with pulsatile tinnitus: A pilot study of twenty patients with pulsatile tinnitus eligible for digital subtraction angiography

1 | INTRODUCTION

Tinnitus is a common problem, with a prevalence of about 8% in the general population. A pulsatile tinnitus occurs in less than 10% of tinnitus patients. It has an extensive differential diagnosis. In general, the underlying pathology for a pulsatile tinnitus is found in up to 70% of the cases.¹ Most frequently, the diagnosis can be made based on non-invasive imaging techniques (CT or CT-angiography (CTA); MRI or MR-angiography (MRA)). However, evaluation of flow dynamics is limited on MRI/MRA and CTA and particularly dural arteriovenous fistulas (dAVFs) are difficult to recognise.

With an estimated incidence of 0.16 per 100 000 adults per year dAVFs are a rare intracranial vascular malformation.² In more than 10% of the cases of patients with a dAVF, a pulsatile tinnitus is the only initial symptom.³ A dAVF with cortical venous involvement (Borden 2 or 3) can, if left untreated, have an aggressive natural disease course with neurological sequelae and even death.⁴ Therefore, patients with pulsatile tinnitus lacking a clear diagnose after non-invasive imaging should be referred for digital subtraction angiography (DSA), which is considered as the gold standard for detection of dAVFs (Figure 1). On the other hand, DSA is an invasive imaging modality, with a procedural risk of 1%-2% of neurological complications, of which 0.5% are permanent.⁵ In a tertiary care setting, a dAVF was found in 24% of patient with pulsatile tinnitus that underwent a DSA.⁶ As such, the majority of patients with pulsatile tinnitus indicated for a DSA are unnecessarily exposed to these risks. It would be valuable to have a screening tool to narrow indication for a DSA in patients with pulsatile tinnitus.

In some patients, the pulsatile sound is externally detected by a clinician, for example, by auscultation with a stethoscope and sometimes even with bare-ear listening. Tinnitus is then referred to as objective. The incidence of objective tinnitus in patients with pulsatile tinnitus varies from 6% to 42%.⁷ The incidence of an objective tinnitus in patients diagnosed with dAVF is higher.⁸ Suggesting that an objectified pulsatile tinnitus may be a unique characteristic for patients with dAVF, recently a new method of objectifying pulsatile

tinnitus was introduced by measuring sound with a sensitive microphone in the external auditory canal; transcanal sound recordings.⁹ We investigated whether with transcanal sound recordings the indication for DSA in patients with pulsatile tinnitus could be narrowed.

2 | MATERIALS AND METHODS

2.1 | Ethical considerations

The Medical Ethics Committee of the UMCG evaluated the research proposal and concluded that the study was not subjected to the Dutch Law on Medical Research with Humans. The board waived the need for patient consent. The study was conducted in accordance with the Declaration of Helsinki and applicable Dutch laws.

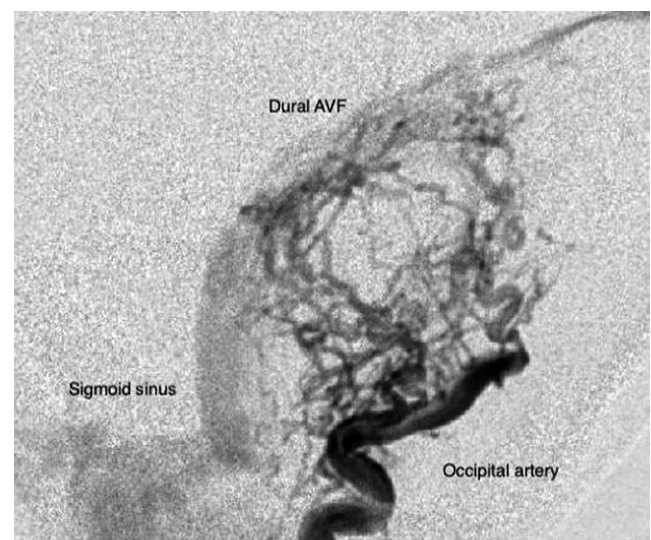


FIGURE 1 Lateral projection of a digital subtraction angiography examination demonstrating a dural arteriovenous fistula (dAVF)

2.2 | Patient selection

The patient cohort for this study consisted of 20 consecutive patients with pulsatile tinnitus that were referred for a DSA to rule out neurovascular pathology. Patients who declined a DSA were excluded from this study. All patients reported a pulse-synchronous sound that matched their heartbeat. They were all assessed with a physical examination at the ENT department (including otoscope inspection, hearing evaluation, peri-auricular and neck auscultation with stethoscope), followed by non-invasive imaging (Table 1). In all patients, the aforementioned examinations did not lead to a definitive diagnosis for the pulsatile tinnitus. Eventually, all patients in this selected cohort underwent a six-vessel catheter angiography and a transcanal sound recording.

2.3 | Transcanal sound recordings

The transcanal sound recordings were obtained preceding the DSA. The neurosurgeon who proposed the DSA to these patients was blinded for the result of the recordings. All sound measurements were performed in a sound-isolated chamber. A microphone (Model ER10B, Etymotic Research Inc, Elk Grove Village, IL, USA) was placed in the external ear canal of the affected ear and a sound registration was made. The signal was amplified and filtered with a bandpass filter (30 Hz-15 kHz) and sampled at 44.1 kHz. A pulsatile tinnitus was regarded as objective (positive sound measurement) if a pulse-synchronous sound was present in the recorded signal. If no pulse-synchronous sound could be detected in the registration, it was regarded as subjective tinnitus.

2.4 | DSA examination

For the six-vessel wall catheter DSA, patients were admitted in day-care. During the procedure the cranial arteries were visualised by using a standard biplane fluoroscopy (ARCADIS Orbic, Siemens, Germany). All DSA examinations were evaluated by a neuroradiologist. If a dAVF was found, it was classified according to the Borden classification.

2.5 | Statistical analysis

Sensitivity and specificity values were calculated (with 95% confidence intervals; Clopper-Pearson interval) to analyse the relation of the transcanal sound recording with the outcome of the DSA.

3 | RESULTS

3.1 | General characteristics

Between December 2015 and November 2018, 20 patients underwent a DSA to rule out a dAVF and participated in our study. The study included 7 males and 13 females with a mean age of 54 years (SD \pm 15). The pulsatile tinnitus was unilateral in 16 out of 20 patients. A CT-scan was made in 10 of 20 patients. All patients had an MRI/MRA examination. In seven cases MRI/MRA raised a suspicion

Keypoints

- In pulsatile tinnitus, the differential diagnosis includes neurovascular pathology, which can be occult on non-invasive imaging techniques. Therefore, if a clear diagnosis is lacking, digital subtraction angiography (DSA) is indicated to rule out a potentially hazardous vascular lesion, particularly a dural arteriovenous fistula (dAVF). However, a DSA carries a procedural risk of 1%-2%.
- In a tertiary care setting, the incidence of a dAVF in pulsatile tinnitus patients lacking a diagnosis after non-invasive imaging is about 25%-35%. Therefore, the majority of this group of patients is unnecessarily exposed to the risks of DSA.
- We report on 20 consecutive patients in a tertiary care setting with pulsatile tinnitus who were referred for DSA to rule out neurovascular pathology. We found that the absence of a pulsatile sound detected by transcanal sound recordings, excludes a dAVF (100% sensitivity).
- Consequently, the use of transcanal sound recordings as a screening tool may prevent patients for the unnecessary risks of DSA in the diagnostic work-up of pulsatile tinnitus.
- Conventional peri-auricular and neck auscultation with stethoscope is not always sufficient to objectify the presence of a pulsatile tinnitus.

of a dAVF. None of the patients had non-invasive time-resolved imaging (trMRA of 4D-CTA).

3.2 | Transcanal sound recordings

A pulsatile sound was detected in the external ear canal of ten patients. In seven patients, DSA revealed an arteriovenous shunt (70%). The angiographic diagnosis was a dAVF in six patients, and an extracranial (subcutaneous) AVF in one patient. In three patients, no abnormalities were found on DSA (false positives). In the remaining ten patients, the sensitive microphone did not detect a pulsatile sound in the ear canal. In none of them, abnormalities were found on DSA.

The sensitivity of sound measurement on the outcome of finding an AVF on DSA was 100% (CI 59%-100%). Its specificity was 77% (CI 46%-95%), indicating that in this cohort an objective pulsatile sound yielded no false negatives in detecting an AVF-shunt. In the same cohort, the sensitivity of detecting an AVF for MRI/MRA and auscultation by stethoscope was calculated 86% and 80%, respectively (Table 2).

4 | DISCUSSION

4.1 | Synopsis of key findings

Transcanal sound recordings can accurately detect pulsatile sounds in patients complaining of pulsatile tinnitus. In this study, DSA showed

TABLE 1 Clinical image and sound characteristics of the patient cohort

| # | Age | Hearing evaluation PTA (dB) | Tinnitus side | Auscultation | CT/CTA | MRI/MRA | DSA (Grade, lateralisation) | Sound measurement |
|----|-----|--------------------------------|---------------|-----------------------|------------------------------|-------------------|-------------------------------|-------------------|
| 1 | 29 | R:17.5, L:10.0 | R | No bruit | Normal | Normal | Normal | Positive |
| 2 | 66 | R:18.8, L:21.3 | R | Bruit retro-auricular | n.a. | Suspicion of dAVF | dAVF (Borden II, R) | Positive |
| 3 | 66 | R:20.0, L:18.8 | L | Bruit carotid | n.a. | Suspicion of dAVF | dAVF (Borden I and III, L) | Positive |
| 4 | 61 | R:30.0, L:37.5 | R > L | No bruit | Fibrous dysplasia | Suspicion of dAVF | Normal | Negative |
| 5 | 62 | R:26.3, L:21.3 | R | No bruit | n.a. | Normal | Normal | Negative |
| 6 | 71 | R:25.0, L:23.8 | L | No bruit | normal | Suspicion of dAVF | dAVF (Borden I, R) | Positive |
| 7 | 69 | R:13.8, L:11.3 | L | No bruit | Normal | Normal | Normal | Negative |
| 8 | 54 | R:18.8, L:17.5 | R | n.a. | n.a. | Normal | Normal | Negative |
| 9 | 30 | R:11.3, L:16.3 | L | n.a. | n.a. | Normal | Normal | Negative |
| 10 | 42 | R:10.0, L:10.0 | L | Bruit retro-auricular | n.a. | Suspicion of dAVF | dAVF (Borden II, L) | Positive |
| 11 | 53 | R:16.3, L:17.5 | L > R | No bruit | Normal | Normal | Normal | Negative |
| 12 | 53 | R:11.3, L:25 | L | n.a. | Dehiscence ssc | Normal | Cutaneous AVF (-, L) | Positive |
| 13 | 29 | R:10.0, L:10.0 | R | No bruit | Thin tegmen tympani | Normal | Normal | Positive |
| 14 | 62 | R:56.3, L:30.0 | L | Bruit retro-auricular | n.a. | Suspicion of dAVF | dAVF's (L) (Borden I and III) | Positive |
| 15 | 53 | R:28.8, L:20.0 | R | No bruit | Normal | Normal | Normal | Negative |
| 16 | 61 | R:27.5, L:27.5 | L&R | n.a. | Normal | Normal | Normal | Positive (L&R) |
| 17 | 37 | R:40.3, L:43.8 | L | No bruit | Fenestral otosclerosis (L&R) | Normal | Normal | Negative |
| 18 | 36 | R:10.0, L:10.0 | R | No bruit | n.a. | Normal | Normal | Negative |
| 19 | 67 | R:18.8, L:18.8 | L&R | No bruit | n.a. | Normal | Normal | Negative |
| 20 | 73 | R:32.5, L:33.8 | R | Bruit (L&R) | n.a. | Suspicion of dAVF | dAVF (R) (Borden I) | Positive (L&R) |

DSA, digital subtraction angiography; dAVF, dural arteriovenous fistula; n.a., not available; PTA, pure tone average 0.5, 1, 2, 4 kHz; ssc, superior semi-circular canal.

TABLE 2 Diagnostic value of examinations on an arteriovenous fistula in PT patients

| Examination | % Sensitivity (95% CI) | % Specificity (95% CI) |
|----------------------------|------------------------|------------------------|
| Transcanal sound recording | 100 (59-100) | 77 (46-95) |
| MRI/MRA | 86 (42-100) | 93 (66-100) |
| Auscultation ^a | 80 (28-99) | 100 (69-100) |

^aIn five patients, the outcome for auscultation could not be retrieved.

no abnormalities if no sound was recorded, while in seven of ten patients with a detectable sound, DSA revealed an AVF. Three patients with an objective pulsatile tinnitus remained without a diagnosis despite non-invasive imaging and DSA. Our encountered prevalence of dAVF was 35%, comparable with the findings of 't Veld et al.⁶

We argue that DSA should be considered if a cause for pulsatile tinnitus is lacking following a conventional diagnostic work-up, including non-invasive imaging (CT/CTA and/or MRI/MRA). Particularly in case of a pulse-synchronous tinnitus, the clinician should rule out a dAVF, a neurovascular entity that may be occult on cross-sectional imaging. A dAVF regularly presents with a pulsatile tinnitus. If left untreated, a dAVF can have an aggressive natural course with neurological sequelae and even death. On the other hand, DSA is also notorious for its risk on neurological complications. Therefore, it is valuable to have a diagnostic tool to narrow the amount of PT patients eligible for DSA.

4.2 | Comparisons with other studies

Dural arteriovenous fistulas can be occult on MR imaging. In a tertiary care setting, 't Veld et al⁶ found a sensitivity of 75% of MRI/MRA in detecting dAVFs in patients with pulsatile tinnitus. In this study, we found a sensitivity of 86%. This supports that lack of support of an AV-shunt on MRI/MRA does not preclude the need for a DSA in these patients.

In case a bruit is heard in patients with pulsatile tinnitus, the incidence of vascular pathology is high. It has already been demonstrated that using a conventional stethoscope for auscultation is not sufficient in the detection of objective tinnitus.¹⁰ In our population of 20 patients eligible for DSA, in 10 patients a bruit could be detected by using transcanal sound recordings. In only four patients, a bruit was objectified by conventional auscultation. These findings demonstrate that simple auscultation is not always sufficient for objectifying the presence of a pulsatile tinnitus.

4.3 | Strength of the study and limitations

To our knowledge, this is the first study that provides data of a perfect association (100% sensitivity) between objective pulsatile tinnitus, as established by transcanal sound recordings, and a positive DSA. However, the patient numbers are small and further research with a larger patient cohort is necessary to determine the diagnostic accuracy of sound measurements as a diagnostic test.

4.4 | Clinical applicability of the study

Our study strongly suggests that the absence of a pulsatile sound detected by transcanal sound recordings, excludes the presence of an AVF and therefore the need for a DSA. In contrast, the presence of a pulsatile sound provides a strong motivation to perform a DSA. As such, transcanal sound recordings may prevent patients with pulsatile tinnitus the risks of an unnecessary DSA.

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CONFLICT OF INTEREST

The authors declare no conflict of interest in connection with this article.

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Juvenile nasopharyngeal angiofibroma: A retrospective study of 27 cases in the ENT department of IPO-PORTO

1 | INTRODUCTION

Juvenile nasopharyngeal angiofibroma (JNA) is a highly vascular and histologically benign tumour and the most common benign tumour of the nasopharynx.¹ However, it is a rare tumour accounting for 0.05% to 0.5% of all head and neck tumours.² They appear almost exclusively in males between the ages of 10 and 20.³ It is thought that JNA originates from the posterosuperior margin of the sphenopalatine foramen.¹⁻⁶ Due to their anatomical location near the skull base and their tendency towards local invasion, these tumours tend to originate extensive tissue destruction, bone remodelling⁶ and frequent recurrence. This growth pattern and spreading to local structures is unpredictable and at the time of diagnosis, most tumours are locally advanced.¹ The most common clinical presentation is a painless nasal obstruction initially unilateral, profuse and recurrent epistaxis and rhinorrhoea.^{1,4} Diagnosis is based on a careful history and nasal examination, supplemented by imaging studies. Biopsies may lead to massive bleeding and are not recommended.⁴

There are several staging classifications for JNA,¹ the most popular are Andrews-Fisch and Radkowski classifications.⁷

Surgical resection is considered the primary treatment option for JNA, and the objective is to achieve tumour exposure and complete resection with the least possible morbidity.^{1,4}

Recurrence rates reported in the literature are around 20% (range 5%-50%).⁴ The main reason for tumour recurrence is strongly associated with incomplete resection and extension of tumours to critical areas such as the infratemporal fossa, sphenoid, foramen lacerum, medial cavernous sinus and anterior cranial fossa.⁸

Radiotherapy might be used as an alternative treatment in recurrences, locally advanced tumours with high risk of significant surgical morbidity, residual/recurrent disease or tumour in neuro-critical areas.⁸

The aim of this paper is to demonstrate our experience in the management of JNA.

2 | MATERIAL AND METHODS

In this retrospective study, we reviewed medical records of all patients diagnosed with JNA and treated in the Department of Otolaryngology—Head and Neck Surgery of the *Instituto Português de Oncologia* in Porto between 1989 and 2017.

Data collection was based on the review of clinical records, noting data on age, gender, previous surgery, clinical presentation, tumour size and staging according to the Radkowski classification system, mean time from the start of symptoms until surgery, surgical approach, complications, recurrence and persistence rate and length of follow-up. Blood loss during surgery and transfusion requirements was also quantified.

We included in our study all patients with complete clinical records, preoperative clinical examination and CT or MRI, dates of surgical approach, histological records and adequate follow-up.

In this study, we considered persistence of tumour when complete tumour excision was not achieved during surgery or in cases that tumours were present in postoperative image (CT or MRI) in immediate follow-up. Tumour recurrence was considered if after complete excision and after 12 months of follow-up without clinical evidence of tumour or without clinical symptoms, we observe recurrence of disease by clinical observation or image.

About the monitoring strategy, along the first year, patients were observed every three months, with physical complete examination including endoscopy. After one year, and during two years, we observed patients every 6 months, with the same methodology. After