

**Comparative performance of prediction model, non-expert and tele-diagnosis of common external and middle ear disease using a patient cohort from Cambodia that included one hundred and thirty-eight ears.**

### **Models for the diagnosis of otological disease**

James Schuster-Bruce<sup>1</sup>

Prajwal Shetty<sup>2</sup>

James O'Donovan<sup>3</sup>

Rishi Mandavia<sup>4</sup>

Touch Sokdavy<sup>5</sup>

Mahmood F Bhutta<sup>6</sup>

1. Department of Ear, Nose and Throat surgery, St George's Hospital, London, United Kingdom
2. Prajwal Shetty, Department of Ear, Nose and Throat Surgery, Worthing Hospital, Worthing, United Kingdom
3. Department of Education, University of Oxford, Oxford, United Kingdom
4. National Institute for Health Research, University College London Hospitals Biomedical Research Centre, London, United Kingdom
5. Department of Ear, Nose and Throat surgery, The Children's Surgical Centre, Phnom Penh, Cambodia.
6. Department of Ear, Nose and Throat surgery, Royal Sussex County Hospital, Brighton, United Kingdom

Words: 1499

### **Corresponding Author:**

Mr James Schuster-Bruce

Department of Ear, Nose and Throat surgery, St George's Hospital, London, United Kingdom

James.schuster.bruce@gmail.com

This article has been accepted for publication and undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the [Version of Record](#). Please cite this article as [doi: 10.1111/COA.13695](#)

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### **Acknowledgements and Funding**

We thank Sitha Heng for translation of the questionnaire from English to Khmer and the Association of Surgeons in Training (ASiT) for the travel bursary awarded to JSB.

### **Conflict of interest statement**

JSB received funding from Cupris to cover part of his travel costs in conducting this study. MB has previously received funding from Cupris for participation in market research. None of the authors are in receipt of ongoing employment, funding or other personal financial interest in Cupris. Cupris played no part in the design, conduct, analysis or reporting of this study.

### **Availability of data and materials**

Data is available on request

### **Ethical Considerations**

Ethics was granted from the CSC ethics board

### **Author contribution statement**

MB, RM, JO'D and JSB designed the study. JSB, TS, PS and MB were involved in data collection and analysis. MB and JSB wrote the paper with all authors contributing edits.

### **Keywords**

Smartphone otoscope, digital otoscopy, remote diagnosis, symptom algorithm, prediction model, community health workers, ear disease.

DR JAMES SCHUSTER-BRUCE (Orcid ID : 0000-0001-8530-1643)

MR RISHI MANDAVIA (Orcid ID : 0000-0002-5839-2735)

Article type : Correspondence: Our Experience

**Key Points:**

1. Efforts to combat the large global burden of ear and hearing disorders are hampered by poor availability of expert diagnosis
2. We report the first study to directly compare prediction model, non-expert and tele-diagnosis of middle and external ear disorders.
3. A prediction model based upon a novel automated otological symptom questionnaire performed poorly, but absence of otorrhoea was found to reliably exclude a diagnosis of chronic suppurative otitis media.
4. Both on-site non-expert and expert tele-diagnosis had high diagnostic specificity, but low sensitivity.
5. Future work could explore how the validity of these diagnostic methods may be improved.

## Introduction

Globally 6% of the population, or around 466 million people, suffer from disabling hearing loss ([www.who.int/pbd/deafness/estimates/en/](http://www.who.int/pbd/deafness/estimates/en/)), and the majority reside in low- and middle-income countries. One barrier to tackling this large burden is the lack of specialist health workers to recognise and manage ear and hearing disorders.<sup>1</sup>

Fuelled by technological advances, there is growing interest in remote or non-expert diagnosis in ear and hearing care. However, studies to date have been limited, mostly focused on screening for hearing loss, without exploring additional symptoms or signs which may signify middle or external ear disorders. This is important: of those suffering disabling hearing loss, around half (200 million) are thought to be due to chronic suppurative otitis media (CSOM),<sup>2</sup> where surgical intervention may be indicated.

Diagnosis of external and middle ear disorders is based on clinical history, including symptoms of otalgia, otorrhoea, and hearing loss, supplemented by otoscopy to visualise the ear canal and tympanic membrane. Diagnostic accuracy relates to expertise and experience of the assessor. Specialist assessment with appropriate tools (including a good quality otoscope) is the gold standard, but due to resource gaps in equipment and specialist health personnel, is not universally available.

We recognise three alternative strategies (Figure 1):

- Telediagnosis,<sup>3</sup> where relevant diagnostic information is captured (usually by a non-specialist) and sent to a specialist
- Non-expert diagnosis, where a non-specialist (defined as someone with less training or experience than a specialist) makes a diagnosis.
- Prediction model diagnosis,<sup>4</sup> where data from a diagnostic tool is fed into an algorithm, with no or minimal human interpretation

*Figure 1: Diagram showing gold standard assessment and three further strategies for diagnosis: tele-diagnosis, non-specialist diagnosis and prediction model diagnosis.*

To date few studies have explored the validity or feasibility of such approaches for diagnosis of external and middle ear disease.



Tele-diagnosis using otoscopic images captured with an endoscope or smartphone has been shown to be successful if users are trained in image capture,<sup>5</sup> but appears unsuccessful if users are given only instructions on use.<sup>6,7</sup> The requirement for training may therefore limit the scalability of this approach.

Studies of non-expert diagnosis of outer and middle ear disorders are sparse, but results to date are not encouraging. In high income settings, medical students, audiology students and general practitioners report low self-confidence in diagnostic otoscopy.<sup>8</sup> A recent study found poor reliability of nurse otoscopic diagnosis in Malawi.<sup>9</sup> Validity of non-expert diagnosis therefore remains uncertain.

Prediction model diagnosis is largely unexplored in this field. For example (and to our knowledge) no study has explored if an algorithm using patient history is valid to diagnose or differentiate external or middle ear disorders.

Here we devised a novel study to directly compare the utility of tele-diagnosis, non-expert diagnosis, and prediction model diagnosis as a screening tool for common external and middle ear disorders. Our test bed was a cohort of patients presenting to an Ear, Nose and Throat (ENT) clinic in the Children's Surgical Centre hospital in Cambodia ([www.csc.org](http://www.csc.org)), a setting with high prevalence of otological disease.

## Methods

### Ethics

The ethics board of the Children's Surgical Centre granted approval.

### Participants

We invited participation from consenting consecutive adult and paediatric patients presenting with self-declared ear or hearing symptoms to ENT outpatients at the Children's Surgical Centre hospital between 21st – 30th May 2018.

### Data capture

Each participant underwent sequential assessment using platforms to capture data on symptoms and otoscopic appearance (figure 2).

*Figure 2: Diagrammatic representation of study design.*

First, symptom history was captured using an automated questionnaire (devised by author MB in English, piloted in ENT clinics in Tanzania and England, and then translated into Khmer). An initial

question asked participants to report ear discharge (otorrhoea), ear pain (otalgia) and/or hearing loss (we also captured symptoms of dizziness and tinnitus, but did not use these in subsequent analysis). Forced responses and skip logic (figure 3) were used to further define each symptom, including laterality, chronology, and (for otorrhoea) consistency. The algorithm also collected data on demographical information, past medical and surgical history and drug and allergy history. For this study the questionnaire was hosted on the Cupris platform (Cupris Ltd, London), and data were captured by one of three ENT specialist nurses who were native Khmer language speakers and had worked at CSC hospital for 1-6 years.

*Figure 3: Diagrams displaying symptom-specific algorithms for (left to right) ear pain, runny ear and hearing loss. Demographic data, as well as past medical history, drug history and allergies were also collected by the questionnaire.*

Next, the same nurse administering the questionnaire captured brief otoscopic videos of each ear using the Cupris TYM device (a platform we have previously shown to be valid for expert tele-diagnosis<sup>5</sup>) attached to an Apple iPhone 5S mobile phone (Apple Inc., Cupertino). Videos were encrypted and stored. Because nurses were naïve to the device, they were given a short tutorial (author JSB) before the study commenced. The training included advice on what constitutes an adequate view of the tympanic membrane, and feedback on three live cases each.

Finally, each participant underwent assessment by one of two resident ENT specialists. They took a targeted clinical history (with open responses) in the Khmer language, performed oto-endoscopy, and recorded their diagnosis. In cases where wax precluded a full oto-endoscopic view, data were removed from final analyses. Specialist assessment was blind to previous assessment, and conducted in a separate area.

#### Outputs

Captured data were used to ascertain three outputs for non-expert diagnosis (figure 2).

The first output was data captured by the questionnaire, which was used for prediction model diagnosis. We also recorded time taken to administer the questionnaire.

The second output was diagnosis reached by the non-expert (nurse), using data they captured with the questionnaire and video otoscopy on the Cupris device. They were asked to select a diagnosis from a

list: normal / acute otitis media (AOM) / CSOM / inactive tympanic membrane perforation / otitis externa / otitis media with effusion / cholesteatoma / other.

The third output was tele-diagnosis by an expert, using data captured by the non-expert (nurse). Questionnaire results were translated back into English, and these and the otoscopic videos were anonymised and sent to specialist otologists in the UK (authors MB and PS). Owing to logistical constraints, data were reviewed 9-11 months after capture. Experts were asked to select a diagnosis from the same list as the nurse.

All assessments were blind to diagnosis by other methods.

Comparison to gold standard

Performance of each diagnostic method was compared to the gold standard, which here was diagnosis by the resident ENT specialist. For each method we calculated sensitivity, specificity, positive and negative predictive value, and likelihood ratio. For the questionnaire we evaluated performance of: each major symptom in isolation; sub-characteristics of each symptom; and combinations of major symptoms. For analysis we classified each ear as independent.

## Results

We recruited 85 participants (15 children and 70 adults), giving a total of 170 ears. One case was lost-to-follow up.

Completion of the questionnaire took an average of 6 minutes (range 1-14 minutes). In 23 ears wax precluded a diagnosis, and in a further 9, technical faults interrupted data capture. Therefore, we analysed questionnaire data for 138 ears.

For otoscopy, we obtained video data for 138 ears. However, experts considered three videos inadequate for tele-diagnosis (poor view of ear canal and/or tympanic membrane), leaving 135 cases for this method.

Gold standard diagnosis was achieved in all remaining cases, with 56 classified as normal external and middle ear, 22 CSOM, 21 otitis externa, 9 inactive tympanic membrane perforation, and 4 AOM. Performance of each diagnostic method compared to the gold standard is summarised in Table 1.

For prediction model diagnosis using the questionnaire, no particular symptom or combination of symptoms reliably differentiated diagnostic categories. For a symptom in isolation, sensitivity for a named diagnosis varied from 33-76%, except for presence of otorrhoea, which had 95% sensitivity for CSOM. Specificity varied between 67-80%.

Non-expert (nurse) diagnosis performed well for specificity (90-99% for all diagnoses) but poorly for sensitivity (<43% for all diagnoses).

Remote expert diagnosis also performed well for specificity (86-99%), but again less well for sensitivity (32-100%).

## **Discussion**

This is the first study to directly compare performance of different methods for diagnosis of external and middle ear disorders. We report variable results. Prediction model diagnosis using automated history performed poorly, but an absence of otorrhoea (perhaps unsurprisingly) was found to reliably exclude CSOM. This implies that individuals who deny a history of ear discharge could confidently be excluded in programs screening for CSOM.

Whereas both nurse-led and remote expert diagnosis showed high specificity, they had low sensitivity. The reasons for poor sensitivity can only be surmised, but could include poor performance of the questionnaire, poor quality video capture on otoscopy, insufficient video resolution, low experience or confidence of the assessor, or diagnostic uncertainty or disagreement. Poor image capture and diagnostic incongruity has been reported in other studies.<sup>7</sup>

We recognise our study as a preliminary exploration of methods for screening or diagnosis of middle or external ear disease (where on-site specialist assessment is unavailable). Future work could look to evaluate if and how validity of these approaches may be improved, including through improving the quality or platform for image capture to assist remote diagnosis; targeted training of non-experts to develop diagnostic skills; or providing additional or enhanced information to facilitate prediction model diagnosis (in particular the addition of automated interpretation of otoscopic images).<sup>10</sup>

## **Conclusions**

We report the first study to directly compare three approaches for non-specialist diagnosis of disorders of the middle or external ear. Our results show suboptimal but comparable performance

using an automated questionnaire, on site non-expert diagnosis, or remote expert diagnosis. Future work should look to enhance the performance of these approaches, including through training of non-experts, or through enhanced algorithms for diagnosis including incorporation of automated otoscopy.

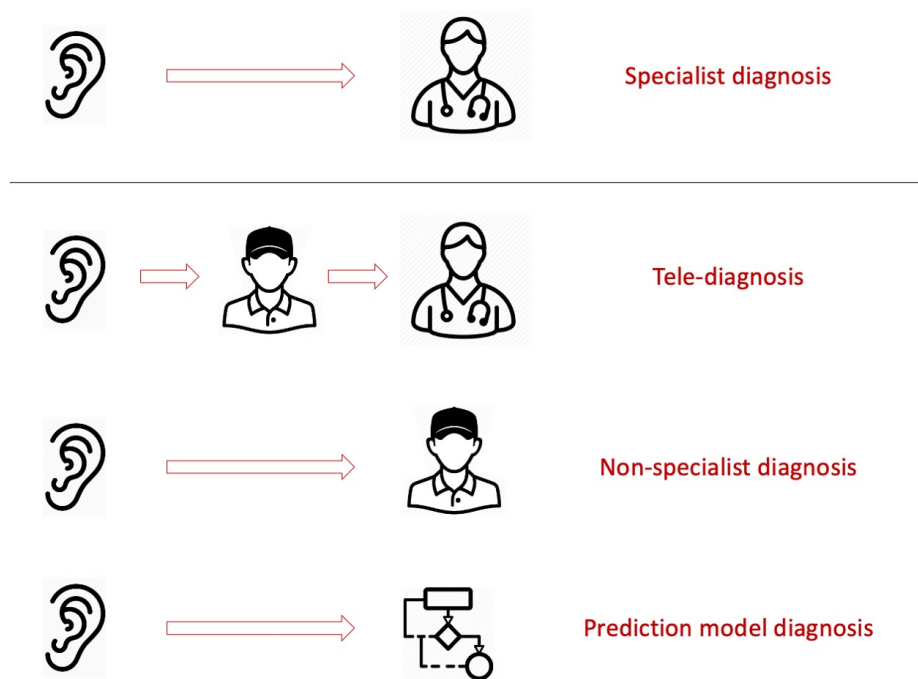
## References:

1. Bhutta MF, Bu X, de Muñoz PC, Garg S, Kong K. Training for hearing care providers. *Bull World Health Organ*. 2019 Oct 1;97(10):691–8.
2. World Health Organization. Chronic suppurative otitis media : burden of illness and management options [Internet]. 2004 [cited 2020 May 31]. Available from: [https://www.who.int/pbd/publications/Chronicsuppurativeotitis\\_media.pdf?ua=1](https://www.who.int/pbd/publications/Chronicsuppurativeotitis_media.pdf?ua=1)
3. Hersh WR, Hickam DH, Severance SM, Dana TL, Krages KP, Helfand M. Diagnosis, access and outcomes: Update of a systematic review of telemedicine services. *J Telemed Telecare*. 2006 Sep;12(2\_suppl):3–31.
4. Collins GS, Reitsma JB, Altman DG, Moons KGM. Transparent reporting of a multivariable prediction model for individual prognosis or diagnosis (TRIPOD): the TRIPOD statement. *BMJ*. 2015 Jan 7;350(jan07 4):g7594–g7594.
5. Mandavia R, Lapa T, Smith M, Bhutta MF. A cross-sectional evaluation of the validity of a smartphone otoscopy device in screening for ear disease in Nepal. *Clin Otolaryngol*. 2016;43(1):31–8.
6. Erkkola-Anttinen N, Irjala H, Laine MK, Tähtinen PA, Löyttyniemi E, Ruohola A. Smartphone Otoscopy Performed by Parents. *Telemedicine and e-Health*. 2019 Jun;25(6):477–84.
7. Demant MN, Jensen RG, Bhutta MF, Laier GH, Lous J, Homøe P. Smartphone otoscopy by non-specialist health workers in rural Greenland: A cross-sectional study. *International Journal of Pediatric Otorhinolaryngology*. 2019 Nov;126:109628.
8. Niermeyer WL, Philips RHW, Essig GF, Moberly AC. Diagnostic accuracy and confidence for otoscopy: Are medical students receiving sufficient training?: Otoscopic Accuracy and Confidence. *The Laryngoscope*. 2019 Aug;129(8):1891–7.
9. Bright T, Mulwafu W, Phiri M, Ensink RJH, Smith A, Yip J, et al. Diagnostic accuracy of non-specialist *versus* specialist health workers in diagnosing hearing loss and ear disease in Malawi. *Trop Med Int Health*. 2019 May 9;tmi.13238.
10. Livingstone D, Talai AS, Chau J, Forkert ND. Building an Otoscopic screening prototype tool using deep learning. *J of Otolaryngol - Head & Neck Surg*. 2019 Dec;48(1):66.

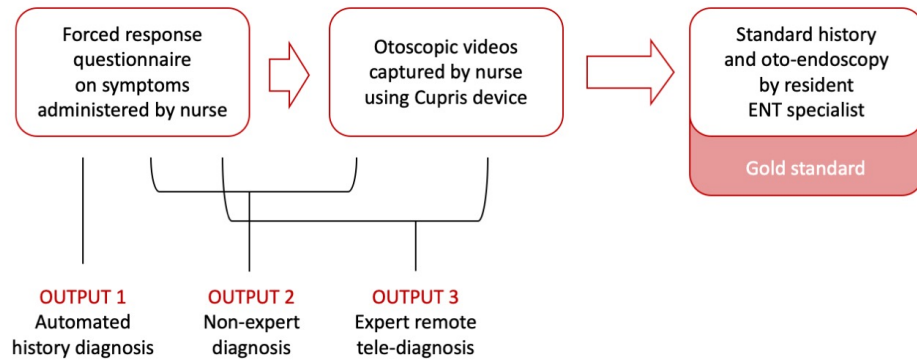
All	CSOM (n=22)						TM perf (n=9)						OE (n=21)						AOM (n=4)						Normal (n=56)						
present	present	Sens	Spec	PPV	NPV	LR	present	Sens	Spec	PPV	NPV	LR	present	Sens	Spec	PPV	NPV	LR	present	Sens	Spec	PPV	NPV	LR	present	Sens	Spec	PPV	NPV	LR	
Prediction model diagnosis, based upon symptom history algorithm (n=138)																															
44	21	0.95	0.80	0.48	0.99	1.19	5	0.56	0.70	0.11	0.96	0.80	11	0.52	0.72	0.25	0.89	0.73	3	0.75	0.69	0.07	0.99	1.08	1	0.02	0.48	0.02	0.41	0.04	
29	15	0.68	0.88	0.52	0.94	0.78	5	0.33	0.80	0.1	0.94	0.42	5	0.24	0.79	0.17	0.85	0.30	3	0.75	0.81	0.1	0.99	0.93	0	0	0.64	0	0.49	0	
21	14	0.64	0.94	0.67	0.33	0.68	0	0	0.84	0	0.92	0	5	0.24	0.86	0.24	0.86	0.28	1	0.25	0.85	0.05	0.97	0.29	0	0	0.74	0	0.52	0	
7	0	0	0.94	0	0.83	0	0	0	0.95	0	0.93	0	5	0.24	0.98	0.71	0.88	0.24	2	0.5	0.96	0.29	0.98	0.52	0	0	0.91	0	0.99	0	
35	21	0.95	0.88	0.6	0.99	1.08	5	0.56	0.77	0.14	0.96	0.72	5	0.24	0.74	0.14	0.84	0.32	1	0.25	0.75	0.03	0.97	0.34	1	0.02	0.59	0.03	0.99	0.03	
46	9	0.41	0.69	0.2	0.86	0.59	3	0.33	0.67	0.07	0.94	0.49	16	0.76	0.75	0.36	0.95	1.01	2	0.5	0.68	0.04	0.98	0.74	4	0.07	0.5	0.09	0.99	0.14	
29	3	0.14	0.78	0.2	0.83	0.18	3	0.33	0.80	0.1	0.94	0.42	11	0.53	0.85	0.38	0.91	0.62	1	0.25	0.79	0.03	0.97	0.32	2	0.04	0.67	0.07	0.99	0.05	
16	6	0.27	0.91	0.39	0.87	0.30	0	0	0.87	0	0.93	0	5	0.24	0.91	0.31	0.87	0.26	1	0.25	0.88	0.06	0.98	0.28	2	0.04	0.55	0.13	0.99	0.04	
39	6	0.68	0.79	0.38	0.87	0.86	4	0.44	0.73	0.1	0.95	0.61	8	0.38	0.74	0.21	0.87	0.52	3	0.75	0.73	0.08	0.99	1.03	2	0.03	0.55	0.05	0.99	0.07	
9	0	0	0.92	0	0.83	0	1	0.11	0.94	0.11	0.94	0.12	4	0.19	0.96	0.44	0.87	0.20	1	0.25	0.94	0.11	0.98	0.27	0	0	0.89	0	0.99	0	
30	15	0.68	0.87	0.5	0.83	0.78	3	0.33	0.79	0.1	0.94	0.42	4	0.19	0.78	0.13	0.84	0.24	2	0.5	0.79	0.07	0.98	0.63	2	0.04	0.66	0.07	0.99	0.05	
22	1	0.83	0.5	1	1.25	8	0.89	0.72	0.18	0.99	1.23	18	0.85	0.78	0.41	0.97	1.10	3	0.75	0.69	0.07	0.99	1.08	5	0.09	0.52	0.11	0.46	0.17		
21	0.95	0.80	0.48	0.99	1.19	7	0.78	0.71	0.16	0.98	1.09	15	0.71	0.75	0.34	0.94	0.95	4	1	0.70	0.09	1	1.43	3	0.05	0.5	0.07	0.44	0.11		

Table 1: Table displaying sensitivity (sens), specificity (spec), positive predictive values (PPV), negative predictive values (NPV) and likelihood ratios (LR) across diagnoses for three different diagnostic models. Diagnoses include Chronic Suppurative Otitis Media (CSOM), Inactive TM perforation (TM perf), Otitis Externa (OE) and Acute Otitis Media (AOM).

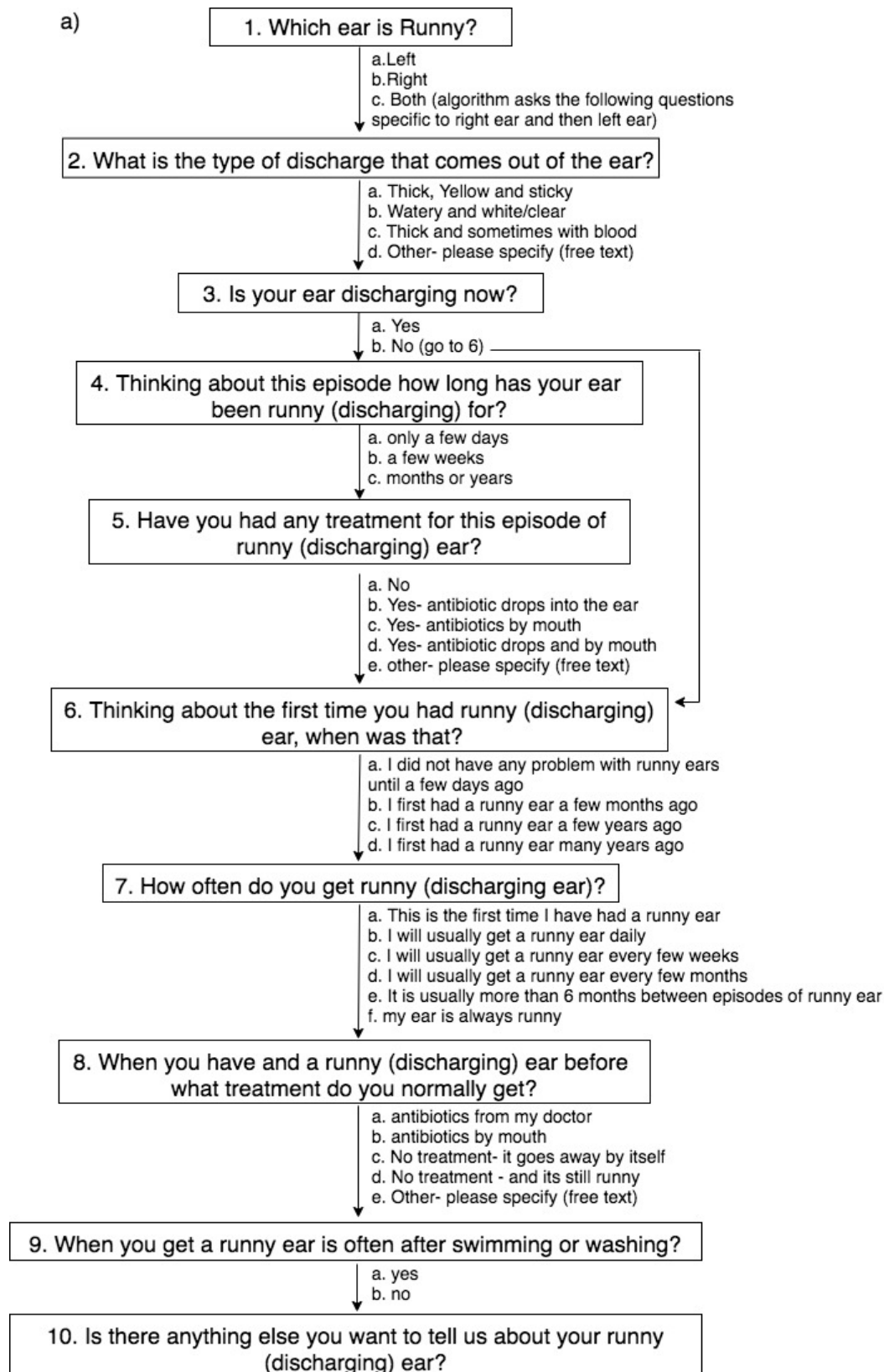


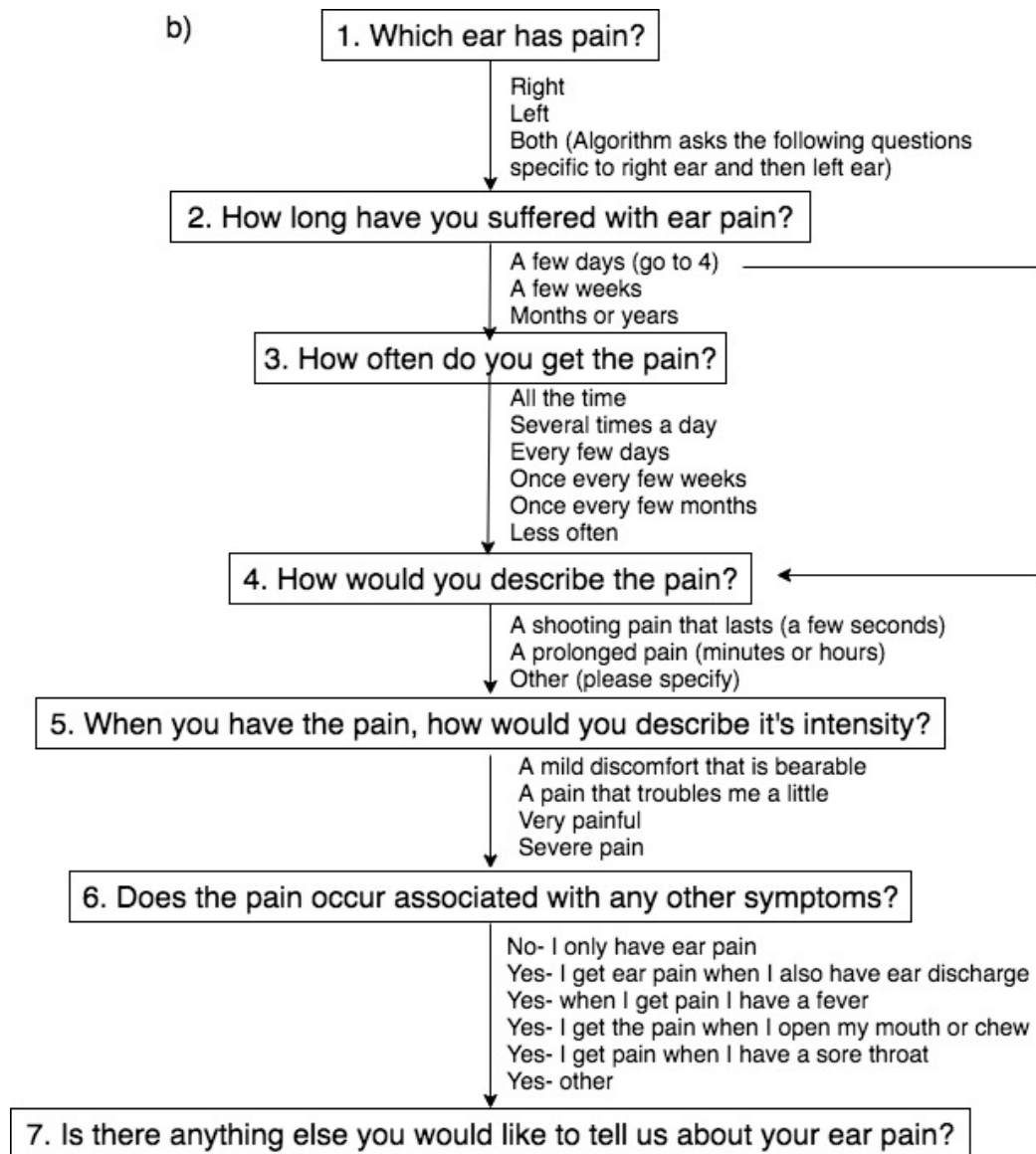


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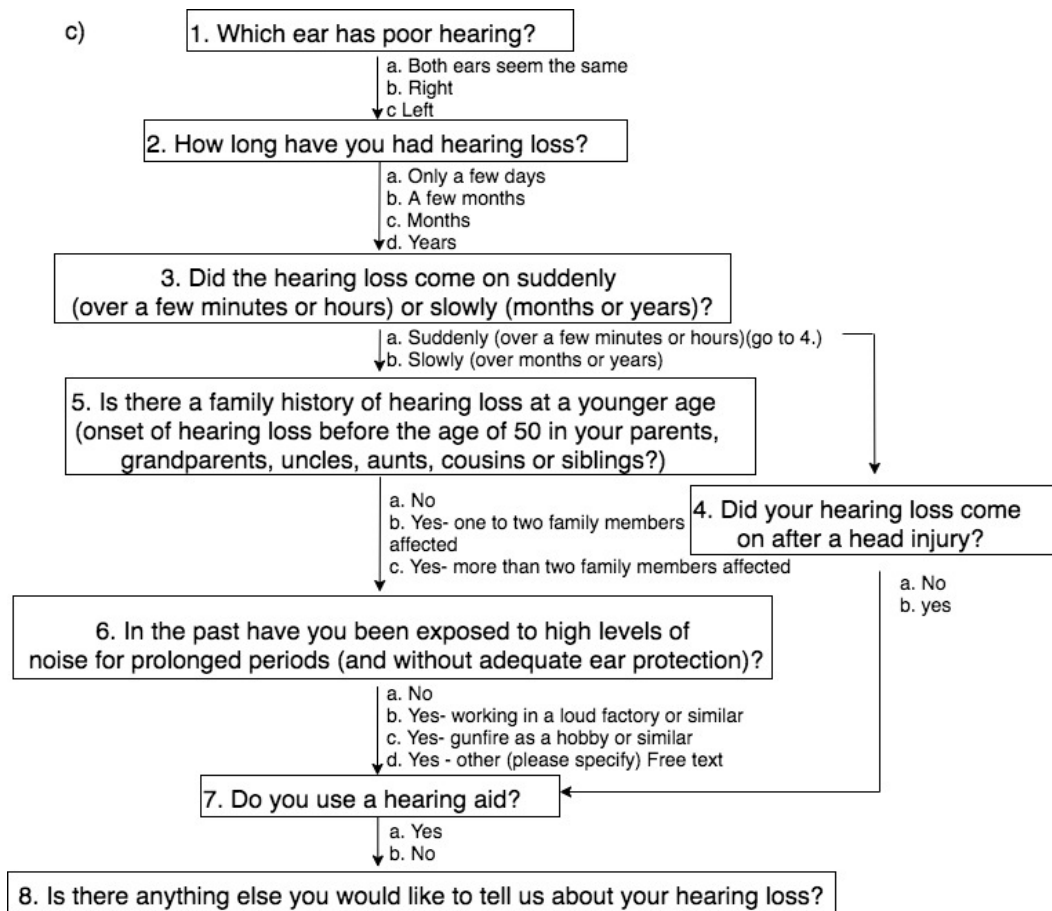


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