



Research Paper

BPPV Viewer: A downloadable 3D BPPV model for study of otolith disease

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Abstract *Objective:* To develop a downloadable three-dimensional (3D) study tool of the membranous labyrinth in order to facilitate the study of benign paroxysmal positional vertigo (BPPV).

Background: The diagnosis and treatment of BPPV depend on an understanding of the anatomy of the vestibular labyrinth and its position relative to the head. To date, many illustrations have been made to explain principles of diagnosis and treatment of BPPV, but few have been based on anatomical studies of the membranous labyrinth.

Methods: A previously reported 3D model of a human labyrinth was transposed to a 3D development software to allow the creation of markers along the semicircular ducts and utricle. These markers represent otoliths at different positions during movement of the model within the 3D environment. User-friendly tools were created to navigate the model, to allow clear documentation and communication of studied problems, and to study the model across relevant planes. The final model can be downloaded and is available for general use at <https://bppvviewer.com/download/>.

Results: The model allows visualization of true membranous labyrinth anatomy in both ears simultaneously. The dependent portion of each semicircular duct, the planes of the cristae, and the position of the utricle can easily be visualized in any head position. Moveable markers can mark the expected progress of otolith debris with changes in head position and images can be captured to document simulations in various draw styles.

Conclusion: This simple model could offer insights that lead to more accurate diagnosis and treatment of BPPV. It may also be useful as a tool to teach BPPV.

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Introduction

Benign paroxysmal positional vertigo (BPPV) is the most common cause of vestibular symptoms in humans. Initially thought to be caused by vertebro-basilar insufficiency, Dix and Hallpike proved that symptoms could be produced without head on body rotation.¹ Attention was then diverted to histologic evidence of cupulolithiasis and finally,² after 20 years, canalolithiasis was established as the main cause of BPPV.³ This prolonged insight timeline gives testimony to the fact that clinical BPPV can be confusing. Any canal can be involved with either cupulo- or canalolithiasis. In canalolithiasis otolith debris is freely moveable within the semicircular ducts, whereas in cupulolithiasis the debris is adherent to the gelatinous cupola. Multiple canals may be involved in the same patient and cause eye movements with different intensities, directions and time courses to be superimposed on one another. Even after diagnosis, treatment maneuvers for repositioning of misplaced otoliths have proliferated and have generated considerable confusion among practitioners. Illustrations offered by the authors of new maneuvers often bear no relation to true anatomy of the membranous labyrinth. Also, illustrations often use the bony labyrinth instead of the semicircular duct to illustrate problems in otolith disease, despite the distinct morphological differences in their anatomy. This may have clinical implications for the diseases being described.⁴ Clarity in scientific communication around BPPV has been compromised because of the absence of a reference model for labyrinthine anatomy.

While the anatomy of the bony labyrinth has been studied for more than a century using a variety of techniques,^{5,6} it is the morphology of the membranous labyrinth that is most important to the study of BPPV. Studies of the morphology of the human membranous labyrinth have been more recent and limited. As the membranous semicircular ducts cannot be practically delineated with current MRI or clinical CT, the most detailed of these studies have employed histologic techniques. These recent investigations have focused on canal plane analysis, estimates of endolymph volume, measures of canal cross-sections and sensory epithelia, biomechanical modeling of semicircular canal function and an anatomical model of the temporal bone for teaching.^{7–11}

An anatomic model of BPPV based on human membranous labyrinthine anatomy correctly associated with the human head can clarify communications related to BPPV and result in better teaching, innovation and refinements of treatment. We have created and previously reported on a 3D model of a human vestibular labyrinth for the study of BPPV.¹² The method of its creation from histologic sections has been outlined. Continued development and deployment

of a downloadable version available to any interested clinician is reported herein.

Methods

Histologic sections to digital network

As previously reported,¹² this model was created from histologic sections from a one-day old female, born at fortyweeks of gestation in the collection of The Temporal Bone Foundation (Boston, MA). The temporal bone was harvested 2 h postmortem. Following fixation in formalin, the specimen was decalcified using 5% trichloroacetic acid, embedded in celloidin, and serially sectioned in the axial plane at a thickness of 20 μm . Each of 780 sections was stained with hematoxylin and eosin, mounted on glass slides and digitized at 5590 x 3473 pixel resolution. The image stack was transferred into our workstation memory and into Amira 5.2.2 (Thermo Fisher Scientific Inc., OR, USA). The images in the stack were aligned. The utricle, saccule, semicircular ducts, the endolymphatic sac and duct, the maculae and cristae were segmented. A manual technique of segmentation was required for the cristae and otolith organs in which the indistinct segmentation boundary on the digital image was guided by direct observation of the corresponding histologic section on a microscope mounted next to the workstation. Because the cupulae of the cristae become dehydrated in histologic preparation their position is distorted so were not segmented. After smoothing the resulting surface maps, the endolymphatic sac, duct, the saccule and saccular macula were removed. The labyrinth was cloned to create its opposite, and fused with a skull and soft tissue surface taken from the MRI of a male cadaveric specimen. In the Amira 3D environment, the membranous labyrinths were aligned with the MRI images of the skull using anatomic norms averaged from 21 subjects.¹³

Transposition of the model to Unity 3D

The Amira model network was transposed as .obj files to Unity3D (2017.1.1p2) game development software (Unity Technologies, CA, USA) because of its versatility and multiple platform compatibility. This required reassembly of the model including re-alignment. In this environment, the labyrinths were colored to enhance visualization of the canals, and the labyrinth surface maps were separated into individual components that can be turned off in case they obscure details under observation. These sub-segments include: right and left anterior, lateral and posterior ducts, common crus, utricle, vestibular organs, and the

skull and skin surfaces. All subparts of the model are listed in a taxonomy that is easily accessed or hidden with a click.

Creation of canalith markers

Spherical otolith markers were then created in fixed positions around the circumference of the semicircular ducts and in the utricle. Fourteen otolith positions were created for each of the semicircular ducts, four in the common crus, and six in the utricle. These markers were programmed to remain invisible until the cursor rolls over their domain. An otolith marker is present in the center position of each crista and the utricular macula; these are referred to in the taxonomy as the 0-position markers. If clicked, the otolith position markers will remain visible until clicked again. In this way otoliths can be turned on and off without referring to the taxonomy of the labyrinthine subparts. A large or small otolith marker size can be specified.

Creation of a navigation tool

While the model can be manipulated freehand, a navigation tool was developed within Unity 3D that allows rotation of the head freely or at specific angular increments around the x, y or z axes, and around the RALP and LARP planes. The tool can be operated with a mouse or on the trackpad. The mouse scroll wheel offers extra functionality and fine control. This tool facilitates the finding of head positions commonly used in diagnosis and treatment of BPPV. The x, y and z axis deviations are listed at the bottom of the screen.

Marking crista planes

Crista planes were approximated by first setting a line along the curved crest of each crista. Because the crista is curved from end to end, any three points along the crest line of the saddle will describe a plane which can be carried to the opposite ampulla roof. This is a subjective but valuable process, as there is little information on the planes of the cristae.

Appearance

The background color of the model can be changed. The model view can be changed from orthographic to perspective as needed for clarity. The model can be viewed in color, in black and white, or in line drawing mode. The latter modes are present to facilitate the creation of illustrations for publication or other discussion. The navigation tool can be hidden to remove unnecessary information from screenshots.

Reducing model size for download

Although the model was created with high definition data sets, reduction of surface map detail of the skull was necessary to reduce the model to a workable size for download (48 MB for PC, 95 MB for Mac). After

development, the BPPV Viewer network was exported from Unity 3D as a freestanding network for use in Mac and PC environments.

Results

The resulting model functions within the Unity-based BPPV Viewer environment on a PC or Mac computer. Using a mouse or a trackpad, the head and its membranous labyrinths can be rotated in any plane as a unit. The canal markers can be indicated in any desired starting or ending position within the environment to mark and illustrate free otolith debris that could cause symptoms. Sequential screenshots allow a record of problem analysis (Figs. 1 and 2).

Discussion

The evolution of clinicians' understanding of BPPV over the last 50 years has been hampered by the lack of an accurate visual reference that all can refer to for conceptualization and communication. The BPPV Viewer model is both accurate and detailed. Based on human membranous labyrinthine anatomy obtained from histologic sections, the model can be used for analysis in 2D mode. The creation of the BPPV model in a 3D environment ensures the configuration accuracy of the model. This model will allow a clear analysis of many different problems encountered by clinicians interested in BPPV. It can be used for consideration of simultaneous phenomena in multiple canals, on one or both sides. With the use of this model, we have reported on optimization of the Dix-Hallpike maneuver to prevent simultaneous canal stimulations in multi-canal BPPV.¹⁴ Moreover, newer more efficient repositioning strategies may be devised, with the potential to improve treatment of patients.

A clear model allows finer distinctions to be made between predicted phenomena and observed clinical phenomena. There is no better example than the history of the understanding of BPPV itself to illustrate this point. An initial conceptual model of BPPV as cupulolithiasis existed for 20 years despite dramatic discrepancies between results predicted by the cupulolithiasis theory and observed clinical behavior. During this period, confusing and contradictory clinical observations that were essential to progress were discarded, and delayed the conceptualization of canalolithiasis as the predominant form of otolith disease.

The BPPV Viewer may facilitate the creation of an accurate repository of techniques of BPPV diagnosis and treatment. Common assumptions, such as the assumption that all otoliths assume the most dependent position in any head position or in response to movement, may be wrong. Mathematical modeling of BPPV, or clinical experience may contradict model predictions. In these cases, there will be an opportunity for greater insight.

The aVOR app developed in Sydney was made as a teaching, training and testing tool for the vestibulo-ocular reflex (VOR), the vestibular system and its disorders,

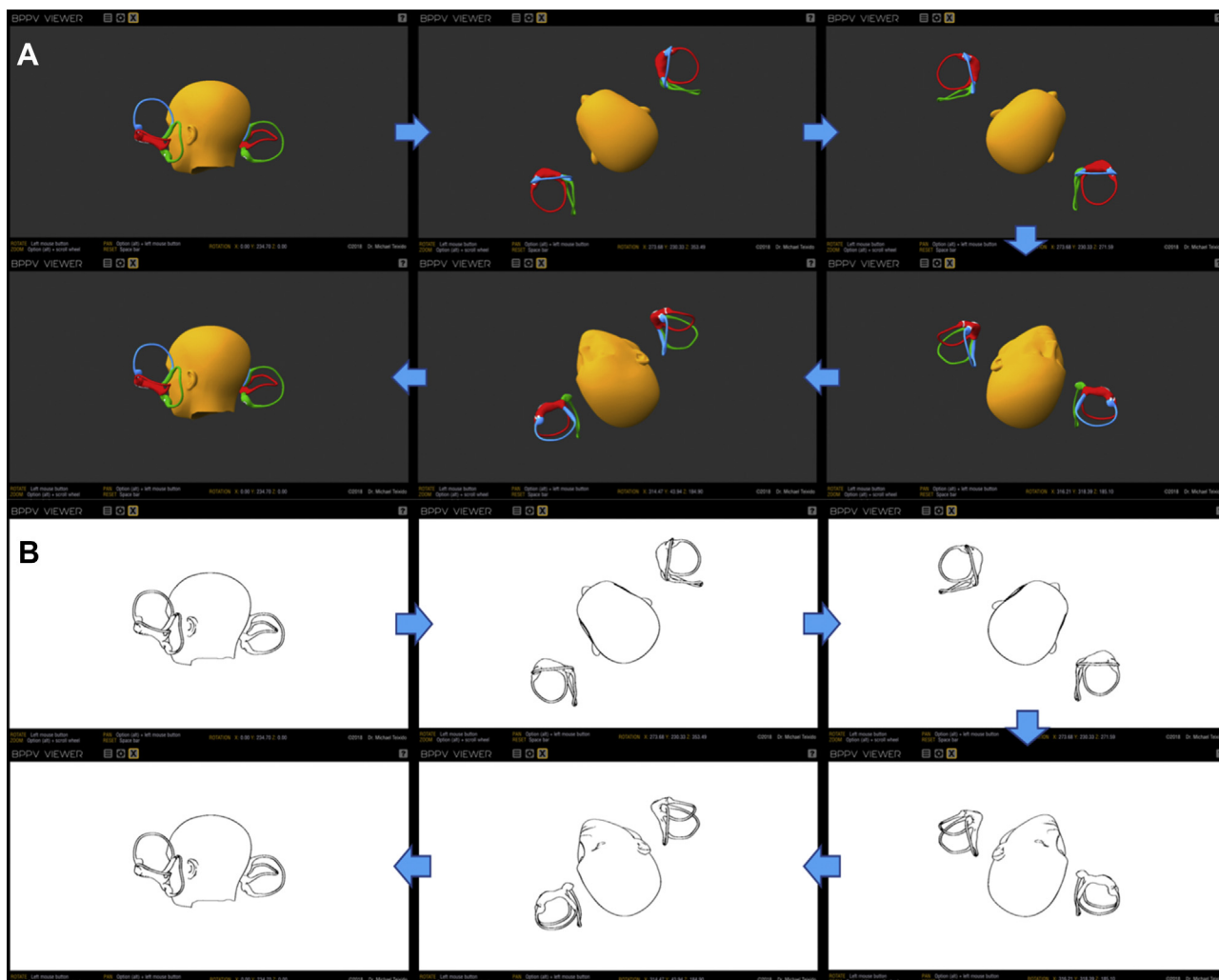


Fig. 1 A: a series of screenshots from the BPPV Viewer demonstrating an expanded Dix-Hallpike maneuver.¹⁴ This sequence will clearly separate posterior canal responses from one another, and anterior canal responses from posterior canal responses. B: the same sequence in draw mode is suitable for making images for publication.

including BPPV. It demonstrates eye movements, including those caused by canalithiasis.¹⁵ The app has been downloaded more than 50,000 times and its usefulness in teaching BPPV to medical students has been demonstrated.¹⁶ BPPV Viewer is a simpler model but offers a clearer visualization. Because it is based on a membranous labyrinth, it allows distinction of problems in BPPV which are not addressed by the app. For example, the BPPV Viewer allows the user to understand the difference between geotropic and ageotropic lateral canal BPPV based on the position of canaliths, whether they are anterior or posterior to the anterior inflection point of the lateral canal. BPPV Viewer also allows representation and therefore consideration of otoliths in multiple canals. General representation of the cristae planes allows consideration of cupulolithiasis in the BPPV Viewer. The viewer is useful as a communication tool and is anticipated to enhance discussion of BPPV in the way that images from the 3D Model of the Temporal Bone have enhanced the discussion of temporal bone anatomy.¹⁷ The BPPV Viewer is a 3D BPPV Model that is now available to any interested clinician at <https://bppvviewer.com/download/>.

Limitations

The authors acknowledge that although the model is based on a human membranous labyrinth the model is based on only a single labyrinth. It resides within the bony labyrinth which itself has small but significant variations of position within the human skull. As such, the model may not be said to be a final predictor of all possible otolith movement phenomena related to BPPV. There are limitations in the accuracy of the crista planes represented in the model. An attempt was made to derive the crista planes by selecting points along the saddle crest of each crista and fitting a plane to the marked points, but the variation between trials was too great to be relied on. The represented crista planes are instead defined by the null positions observed in clinical experience for the lateral and posterior canals. They are rather arbitrary for the anterior canals, since their involvement with cupulolithiasis has not been documented with clarity. The inclusion of crista planes in the model is important, however, for clinicians to incorporate cupulolithiasis and light cupula of all cristae in their clinical thinking.

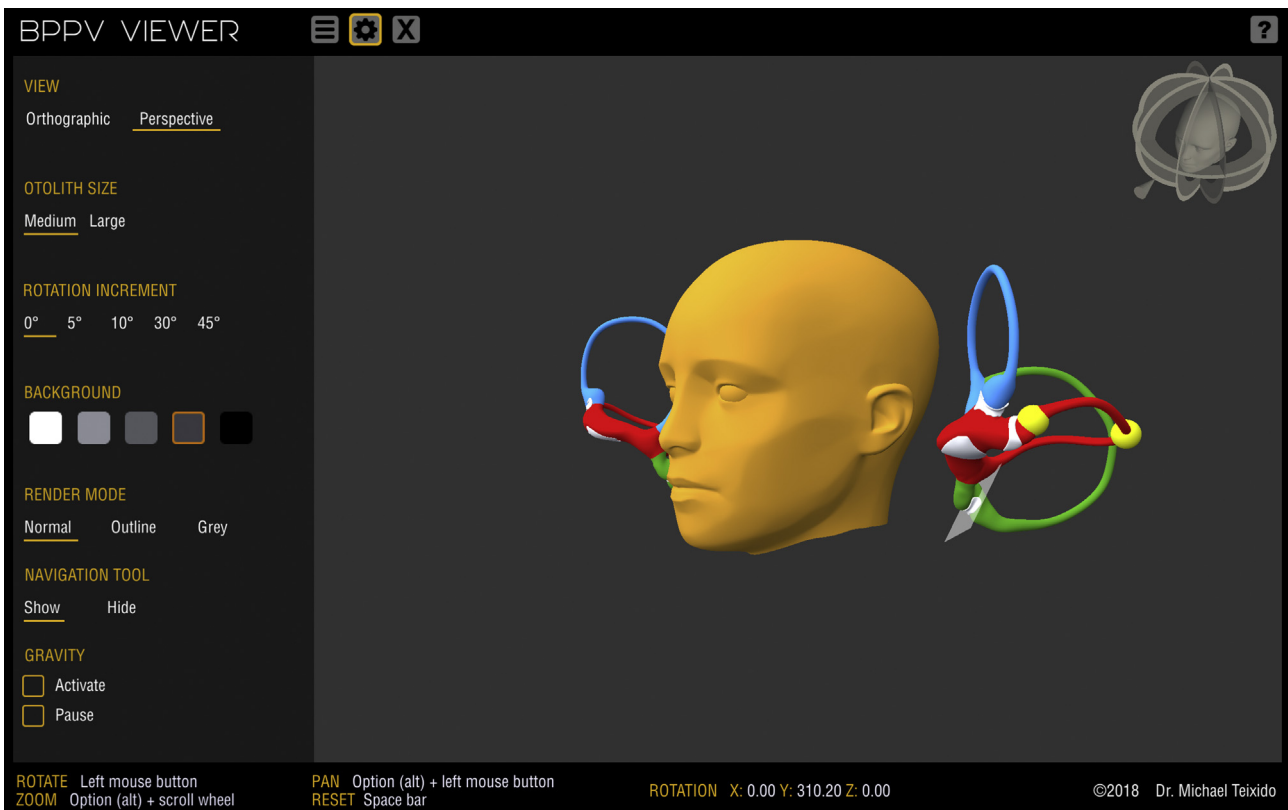


Fig. 2 An illustration of otolith positions anterior and posterior to the anterior inflection point of the lateral semicircular duct. These starting positions correspond to ageotropic and geotropic lateral canalithiasis. The plane of the crista of the left posterior canal is shown.

While useful, there are many possible improvements that can be explored; some of these focus on teaching, training and study, and others on treatment. The current model does not attempt to simulate eye movements. The model could facilitate the creation of simple animations from stored sequences of head positions and otolith movements. The model may also be improved to allow better visualization of simulated disease by creating a gravity environment with varying latency and duration of response. These improvements are relatively simple and require programming efforts and expertise. A more complicated version of the model could include biomechanical modeling of otolith and cupula movement based on known physical parameters such as otolith mass, duct diameter and endolymph viscosity.

In its current form, BPPV Viewer is a study tool that is also useful for teaching. Future planned development as a training tool will include stereo visualization in a virtual reality environment, with eye movements and with a dummy head, or haptic gloves that allow simulated manipulation of the subject head, as in the clinic environment.

Conclusion

The BPPV Viewer is a simple model based on human anatomy and is freely available. It has been created to allow careful study of BPPV pathophysiology and treatment.

Going forward, this tool will enhance communication, support innovation and teaching in BPPV, and could offer insights that may lead to more accurate diagnosis and treatment.

Declaration of Competing Interest

None.

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